The Perception of Leprosy

Studies on assessment, impact and interventions at individual, family and community level

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De Perceptie van Lepra

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Leprosy

Leprosy, or Hansen's disease, is an infectious disease caused by *Mycobacterium leprae* or *Mycobacterium lepromatosis* [1,2]. Although the transmission of *M. leprae* is not fully understood, it is believed to be facilitated by close and long-term exposure to (infected) airborne droplets [1,3]. The first manifestation of leprosy is often the occurrence of hypopigmented, anaesthetic skin patches. Without timely diagnosis and treatment, the disease can be progressive and lead to permanent damage of the peripheral nervous system, the soft tissue of the nose and throat, and vision. Due to a loss of sensation in the extremities, people easily injure themselves. These, often painless, injuries may become infected, which may eventually result in tissue loss in affected areas such as in the fingers and toes [1].

Leprosy mainly affects people in resource-poor countries and is closely linked to health inequalities and poverty [4]. In 2019, the number of new leprosy patients reported globally was 202,185 [5]. The geographical distribution of leprosy is uneven, 80% of the world's leprosy patients live in three countries: India (57%), Brazil (14%) and Indonesia (9%) [5].

Treatment for leprosy (called 'multidrug therapy' or 'MDT') was introduced in 1982. Since 1995, Novartis provides MDT to leprosy patients free of costs, the distribution is managed by the World Health Organization (WHO) [1]. Leprosy patients are no longer infectious within days after taking their first dose of MDT [6]. Up until today, rifampicin, the most important component of MDT, is known as the most effective bactericidal agent against *M. leprae*, also for prevention [6]. In several studies, a single dose of rifampicin (SDR-PEP) has been used as chemoprophylaxis in the prevention of leprosy among close contacts of leprosy patients [7–9]. SDR-PEP only has a protective effect of 57% among contacts, in the first two years after administration [7].

In 2017, the PEP++ project was launched, to find a more effective regimen for the prevention of leprosy. The PEP++ project aims to test the efficacy of an enhanced chemoprophylaxis regimen as compared to SDR-PEP, and to stop the transmission of leprosy in endemic areas in India, Indonesia, and Brazil by using a package of innovative tools and best practices. This package of innovative tools consists of:

- Identifying high-endemic clusters of leprosy through Geographic Information System (GIS) based mapping
- Context-specific community education materials to increase the community acceptance of the study and adherence to treatment, and to change the perception and knowledge of leprosy (this thesis)
- An enhanced preventive medication regimen, consisting of three doses of rifampicin and clarithromycin, given to close contacts of leprosy patients
- Blanket treatment of non-close contacts of patients in the high endemic clusters with SDR-PEP

The PEP++ project will run until 2024.

CHAPTER 1

Introduction

> *"...I was anxious. I was afraid I wouldn't be able to recover like other conditions such as diabetes, high blood pressure and cholesterol ... I was afraid. First, I was afraid I wouldn't get better. Second, I was afraid my family would shun me. Third, I heard that leprosy can cause death..."*
>
> *– Person affected by leprosy, Indonesia*

Perception

'Perception' refers to how individuals or groups "see" an object, person, event or institution [10–12]. Perception is a broad concept; social perception, for example, is how an individual or group "sees" others [10,11]. Perception can also refer to illness; a person's interpretation and understanding of a disease and its potential consequences [12]. Perception comprises knowledge, beliefs, attitudes and emotions which are in turn influenced by personal factors (e.g. personality, experience) and environmental factors (e.g. culture, religion) [10,11,13,14]. These concepts are interrelated and intertwined (Figure 1).

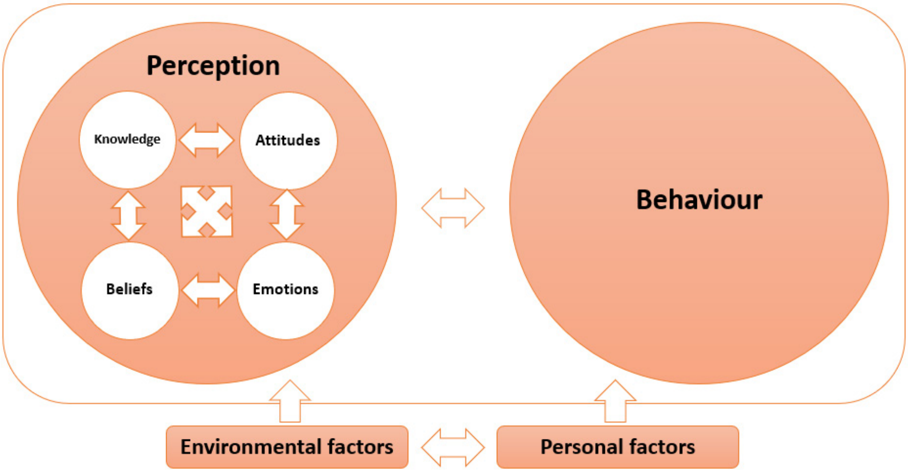

Figure 1. Components of perception and the relation between perception and behaviour.

Knowledge and beliefs

Knowledge and beliefs are mental representations that arise from experiences (everyday encounters or formally constructed through learning) [15,16]. Knowledge and beliefs give meaning to uninterpreted experiences, it is information that is stored, or represented in a person's memory, that can be and is used during cognitive processes [11,16,17]. This information is often unconsciously activated and shapes people’s impressions, judgments and feelings [17]. Beliefs link an object (such as a person, group of people, disease, institution, or behaviour) to an attribute. For example the belief “leprosy is dangerous”, links “leprosy” (object) to “dangerous” (attribute) [18]. Knowledge refers to theoretical or practical understanding of a subject (facts, skills, or objects). Truth and belief are a prerequisite for possessing knowledge: one has a belief in something, and that belief must be true (based on observable and measurable evidence). For example, if you know that leprosy is an infectious disease, then you must believe this, and your belief must be true [19].

Attitudes

An attitude refers to a person's feelings toward and evaluation of an aspect of the person's world, for example an object, person, event, or towards performing specific behaviours [13,18]. It refers to “a person’s location on a dimension of affect or evaluation” and falls on a continuum from very favourable to very unfavourable [18]. Attitudes are based on beliefs about an attitude object, that in turn stem from learning and experiences [10,18]. A person’s attitudes about leprosy, for example, is a function of his beliefs that leprosy has certain attributes and his evaluation of those attributes. If leprosy is associated with primarily favourable attributes, the person’s attitude will tend to be positive [18]. Attitudes help us define how we see and behave towards a situation, and can influence what we remember [10]. Attitudes can include affective (feelings, evaluations), cognitive (knowledge, beliefs), and conative (intentions) components [13,14]. An attitude is both a personal disposition and a societal product (for example, myths, legends, scriptures and folklores all shape attitudes and beliefs) [20].

Emotions

Emotions are inner states such as anger, joy, fear, or love. Several emotions can be experienced at the same time. Emotions can be consciously experienced, but can also be repressed, inhibited or unconscious [21]. Emotions are often intentional, they are directed at an event, object or person [22]. Recreating experiences that evoked strong emotional feelings is important for defining our self-concept and for behaviour intention [23]. Emotions motivate us to act in a certain way, for example, fear motivates avoiding the danger [22]. With over 90 definitions of the term “emotions”, there is little consensus about how to define it [21]. In this thesis, we use the following definition: emotion is an episode of interrelated changes in the states of all or most of the following components of emotion: (1) appraisal of an event, object or person, (2) bodily sensations, (3) motor (facial and vocal) expressions, (4) motivation to act, and (5) subjective (feelings) and emotional experience [24].

Environmental and personal factors

Environmental factors refer to factors in the socio-cultural context –the physical, social, and attitudinal environment- in which individuals live [25]. Examples of environmental factors are attitudes of others, legislation, religion, and culture. Personal factors is a very broad concept and refer to the background of an individual's life and living that are not part of a health condition [25,26], such as gender, age, education, religiosity, experience and personality. Some factors are both personal and environmental, such as worldview. In addition, environmental and personal factors are related and influence each other. For example, personality (the set of psychological traits and mechanisms within individuals), a personal factor, influences how people interact with, and adapt to, the environment [27]. Another example is personal experience; personal experiences are influenced by environmental factors, such as support and services [25].

An example of a concept that influences all aspects of both perception and behaviour is worldview. Worldviews are the basic, largely implicit, assumptions people make to explain reality. Worldviews emerge from our interaction with the world and shape how we see the world [11]. Worldviews are maps (or mental blueprints) people have of reality that they use for living. These maps structure perceptions of reality and guide behaviour [11]. Worldviews in turn are influenced by culture and religion, including beliefs, values, moral codes, symbols and traditions [28]. Both worldviews and culture include socially constructed realities and assumptions individuals use to make sense of the world (however, unlike worldviews, culture also refers to artifacts, technologies and institutions) [28].

Perception is closely related to the concepts in Figure 1, which are in turn related to each other. Perception is an unconscious process. When a person perceives others, information about the other and his or her social group is instantly activated – including attitudes, beliefs, emotions (e.g. fears) and experiences related to this group [17]. An individual’s perception can be substantially different from reality [10]. Social perception influences and is influenced by (but does not include) behaviour, including interactions between individuals [11,29].

Perception is expressed through the following components: knowledge, beliefs, attitudes, emotions. Even though we consider knowledge a component of perception, in this thesis we have sometimes mentioned knowledge separately (“perception and knowledge”). We did this because knowledge can be measured more accurately (knowledge about the disease leprosy refers to facts about the disease) and because other studies found that it is easier to improve knowledge than to change attitudes and behaviour [30,31].

The importance of studying perception

Perceptions of 'others' towards persons affected by stigmatised health conditions, such as leprosy, include local misconceptions, cultural and religious beliefs, and fears that may be linked to these beliefs [32–34]. These perceptions influence behaviour towards persons affected [11,29]. Perceptions and behaviour have a major impact on persons affected, their families and the effectiveness of public health efforts to combat the disease [35]. For many people with a (stigmatised) health condition, the psychosocial consequences of their condition are harder or just as hard to bear as the physical consequences [35,36]. Interpersonal relationships (such as friendships and family relations, marriage), social participation (including education, employment and participation in social events), mobility and leisure activities may be affected [35]. In addition, stigma is associated with poor psychosocial health outcomes such as low mental wellbeing, reduced quality of life and low self-esteem [36,37]. The origin of stigma lies in public perceptions about people who are stigmatised. Given the profound impact of perceptions on stigma and on people's lives, perception (and ways to change perception) is an important subject to study.

Stigma

The process of stigma

Perception is an important driver of stigma [38]. In this thesis, we consider perception a driver and component of stigma. Social stigma contributes to a hidden burden of many health conditions. Many health conditions are associated with social stigma, including cancer, epilepsy, mental health conditions, disability and infectious diseases such as HIV/AIDS, tuberculosis and leprosy [39]. Stigma is a social process with multiple dimensions [38,40,41] and a global phenomenon [35,42,43]. There are different definitions of stigma. Two often used definitions include:

- "a social process, experienced or anticipated, characterized by exclusion, rejection, blame or devaluation that results from experience, perception or reasonable anticipation of an adverse social judgment about a person or group" [38] and
- "when elements of labeling, stereotyping, separation, status loss, and discrimination co-occur in a power situation that allows the components of stigma to unfold" [41].

Stigma includes both cognitive (e.g. knowledge, attitudes, labelling) and behavioural (e.g. rejection, secrecy, withdrawal) elements [44,45]. Stigma consist of five components: labelling, stereotyping, separation, status loss and discrimination, and power [41]. The first of the five components of stigma, labelling, occurs when people identify and label differences between (groups of) people. Examples of differences that are labelled include race, skin colour and sexual preference. Stereotyping occurs after people have been labelled: the labelled person or group is linked to undesirable characteristics – and these characteristics are deemed the same for all members of the group (for example “all persons with leprosy are dangerous”) [41]. Stereotypes –beliefs and expectancies about a group– are a type of interpersonal perception [17,29]. The labels and stereotypes modify the actions of others towards those who have been labelled. Separation occurs when a distinction is made between those who stigmatise (“us”) and the stigmatised group (“them”). People are stigmatised when the labelling, stereotyping and separation leads them to experience status loss and discrimination. The final component of stigma, power, is essential for stigmatisation to occur: power to allow the separation, rejection and discrimination to occur [41]. The people who stigmatise must have the power to make a distinction between "us" from "them" and to have the designation stick [41].

Even though status loss and discrimination are one of the five components of stigma according to Link and Phelan [41], we believe that there can also be stigma without discrimination. This is the case when stigma is for example anticipated or internalised, described in more detail below, in which people do not actually experience unfair treatment or social exclusion. The knowledge that stigma is present in the community, can impact individuals even if they have not directly experienced stigma or discrimination (internalised stigma). Stigma, however, always has a behavioural element – this is often experienced or anticipated discrimination, but can also be secrecy, withdrawal or treatment avoidance [44,45].

Different types of stigma

Weiss [46], extending Scambler’s Hidden Distress Model of Stigma [47], developed a model in which he identified six types of stigma. In this model a distinction is made between those who stigmatise and those who are stigmatised [46]. It should be noted that it is sometimes difficult to make a distinction between people who stigmatise and those who are stigmatised [48]. For example, family members and health workers can be both a source and a target of stigma.

For those who stigmatise, Weiss’ model distinguishes between enacted, endorsed, and accepted stigma. Enacted stigma refers to directly, or actively engaging in stigmatising others (acting on attitudes). Those who do not directly stigmatise others may do this indirectly; they may accept or endorse it. Accepted stigma refers to disagreeing with stigmatisation of others, but not speaking out against it; thus, accepting it without endorsement (Weiss, 2008). Endorsed stigma refers to justifying and supporting the exclusion of others, but not actively engaging in the process [46]. Discrimination is enacted stigma, it is acting on negative attitudes, and refers to unfair treatment or negative behaviour [41,49]. Stigma does not always have to result in actual discrimination [41,49].

For those who are stigmatised, the model distinguishes between enacted, internalised, and anticipated stigma. Anticipated stigma refers to the feeling that discrimination and exclusion will happen and the anticipation that other people will have a negative perception of you, it is the anticipation of being stigmatised by others. Internalised and enacted stigma are actually experiencing stigma. Internalised stigma refers to self-stigma, the stigmatised person "accepts perceived exclusionary views of society and self-stigmatises himself or herself" [46]. Enacted stigma on the other hand, is the extent to which the person who is stigmatised actually experiences discrimination and exclusion [46].

Stigma can occur at different levels. These can broadly be categorised in intrapersonal (affected person), interpersonal (the affected person's environment), organisational/institutional, community and governmental/structural level [42].

Perception and stigma

Perception and stigma are related, perception is an important driver of stigma [38]. Perceptions (knowledge, attitudes, beliefs, emotions) play an important role in stigma [32,33,50,51]. In leprosy for example, people's attitudes towards (persons affected by) leprosy are strongly influenced by cultural and religious beliefs and fears that may be linked to these beliefs. In some cultures leprosy is attributed to supernatural causes, karma, witchcraft, sins or immoral behaviour, or is believed to be hereditary [32–34,51,52]. These beliefs influence how people behave towards persons affected by leprosy.

An important difference between perception and stigma, is that perception is a neutral term, it can be positive or negative, while stigma is characterised by exclusion and rejection – and therefore always negative. Some researchers have argued that the concept of 'stigma' itself is stigmatising – since it implies that something is wrong with the person who is stigmatised [53]. In addition, stigma includes both cognitive (e.g. knowledge, attitudes) and behavioural (e.g. rejection, withdrawal) elements [44,45], while perception is a solely cognitive concept (Figure 1) [18–20]. Perception influences and is influenced by but does not include behaviour. Another limitation of the term 'stigma' is the multiple meanings that are attached to it [53]. We consider stigma a combination of negative perceptions and negative behaviour (Figure 1). In order to assess perception, one should assess knowledge, attitudes, emotions and beliefs. In contrast, assessing community stigma usually only entails assessing behavioural intention and/or attitudes towards persons affected and their health condition, because it is difficult to measure actual behaviour [35]. Understanding local perceptions is crucial to understand explanatory models, attitudes and behaviour [34,54].

The health and social impact of perception and stigma of leprosy

Drivers of perceptions and stigma

While the unfair treatment and social exclusion is very similar for all health-related stigma, the origin of stigma varies between conditions [55]. Stigma against leprosy is strong and universal, and dates to centuries ago. Researchers have identified several drivers (factors that contribute to) leprosy-related stigma. Drivers of stigma are always negative [56]. According to Rao [55], there are references in the Bible that leprosy was caused by a divine intervention, or a curse. A common perception of leprosy is that it (always) causes disabilities and disfigurements [55,57,58]. Leprosy is also thought to be incurable, hereditary or highly contagious [57,58]. The perception of leprosy, and leprosy-related stigma are deeply rooted in social, religious, cultural and economic domains [55].

Drivers of stigma include external manifestations of leprosy, fear, poor knowledge about leprosy, and religious and cultural beliefs [57,58]. These aspects (knowledge, beliefs, fears, perception of external manifestations) all reflect perceptions of leprosy. Persons affected are not perceived as ‘different from the norm’, until their acts or attributes are perceived as different [59]. Table 1 provides an overview.

Table 1. Drivers of perceptions and stigma of (persons affected by) leprosy, based on [55,57,58,60].

Pathways through which stigma can develop	Meaning	Examples of drivers in leprosy
Concealability	Visibility of / ability to hide condition	Concealment is only possible when there are no visible impairments
Course	Persistence over time	Perception that leprosy is incurable, people still experience discrimination even when they have completed treatment
Disruptiveness	Interference with social interactions	Social participation restrictions, often related to fear (of transmission or external manifestations) and cultural and religious beliefs
Peril	Perceived dangerousness of condition, fear	Perception that leprosy is highly contagious, fear of transmission
Origin	Cause / origin of condition	Perception that leprosy is caused by a curse or personal wrongdoing, or is hereditary
Aesthetics	Displeasing nature or appearance	Perception of leprosy as progressively disabling, unattractive

External manifestations of leprosy, the visible impairments associated with the disease, are among the main factors reported to contribute to stigma [61,62]. The external manifestations of leprosy identify persons affected as different from others in the community, persons with physical impairments can for example be perceived as dangerous or low in competence, and make people want to keep their distance. The unpleasant smell caused by infected ulcers and/or the flies attracted by these ulcers may also prevent people from getting close to persons affected [57,58]. As a result, persons affected often attempt to hide their condition [57,58]. For persons with visible impairments, for whom it is difficult to hide their condition, this may mean that they prefer wearing closed shoes instead of sandals.

Another important contributor to stigma is fear of getting infected with leprosy, this often leads to prejudice [57,63]. Fear of getting infected is closely linked to visible impairments that can exaggerate fear of danger or infection, and can exaggerate misconceptions [46,57,58]. For example, the misconceptions that leprosy is incurable, hereditary or highly contagious contribute to fear and stigma [57,58]. Indeed, low knowledge about leprosy and cultural beliefs (or misconceptions) have been found to be associated with negative attitudes and stigma [33,64,65]. Cultural and religious beliefs, such as the belief that leprosy occurs because of wrongdoing, sins or witchcraft, that leprosy is a curse from God increase stigma [57,64]. Finally, inequalities between people in class, age, gender and sexuality can also contribute to and increase stigma [66].

Facilitators of perceptions and stigma

Facilitators refer to positive or negative influences on perception and stigma [56]. Examples include the presence or absence of negative attitudes of health workers, positive or negative beliefs (e.g. perceived controllability of leprosy) and discriminatory or ‘positive’ legislation [56]. Facilitators are similar to drivers, but where drivers are always negative, facilitators can be both negative and positive influences (e.g. the presence or absence of certain beliefs or situations) [56].

Manifestations (experiences and practices)

The manifestations of stigma mirror the types of stigma, stigma can be enacted, internalised, perceived and anticipated [46]. Manifestations influence a number of outcomes for persons affected [56].

Outcomes

Impact on individuals and families

Many persons affected by leprosy experience negative consequences of their condition [58,66]. This is caused by the very widespread negative perceptions and practices towards persons affected and their disease [35]. Stigma can affect and disrupt many areas of the affected person's life and may impact the lives of their family members. In addition, perception of leprosy influences disease management [35,58,66,67].

As a result of physical impairments, pain, negative (self-)perceptions and stigma, persons affected may experience mental distress, suffer from depression or anxiety, and have a low self-esteem and poor quality of life [68]. Fear of infecting others can prevent emotional closeness to family members [58]. Stigma can exacerbate or undermine the availability of resources, social relationships and psychological resources – that can lead to adverse health outcomes, such as stress or depression [69]. Furthermore, restrictions in social participation are common such as restrictions in education, employment and participation in social events [35]. Researchers have found that leprosy and its stigma can affect mental wellbeing (e.g. cause anxiety or depression), quality of life, marriage (prospects and ongoing marriages), problems in finding or keeping a job, social relationships, leisure activities and participation in community life (e.g. attendance at social and religious functions) [58,70].

These factors are interrelated. For example, when someone loses their employment because of physical or social consequences of leprosy, they also lose their income and means of supporting their family, which may cause a loss of respect in their communities and negatively impact their self-esteem [71]. In addition, it imposes a social and economic burden on marginalised families [72,73]. Family members and friends may experience public disapproval as a consequence of being associated with their affected family member or friend (often referred to as courtesy stigma) [68,74] or may lose (family) income [72,73]. The quality of life of the whole family may be affected [75]. In addition, marriage prospects of family members may be reduced [76].

Impact on disease management

There is evidence that perceptions and (fear of) stigma cause delays in seeking treatment. In a study in Brazil, the authors found that persons affected by leprosy who feared community isolation were ten times as likely to wait longer before going to a health centre to get their symptoms checked [67]. A study in Nepal found that stigma indirectly influences perception and disease management [77].

The anticipation of stigma and fears of leprosy can cause persons affected to hide their condition, which only people with less visible signs are able to do successfully. As a result, the image of leprosy as a disease with deformities is not challenged, since persons with visible signs are often not able to hide their condition and new patients do not expect that they have leprosy, because they are not aware of the symptoms [77]. People's knowledge influences their awareness that signs and symptoms are due to leprosy, which, if awareness is lacking, hampers early reporting of leprosy [34,78,79].

Stigma and perceptions can also impede adherence to treatment [80]. People's perception and interpretation of their disease and its treatment, impacts the way they deal with them (for example help seeking and adherence) [81]. For example, people's understanding of leprosy treatment, their belief in the efficacy of treatment or perceived risks or side effects of treatment, can influence their adherence to treatment. Especially side effects of medication, such as darkening of the skin or feelings of weakness, can cause people to discontinue treatment [81]. Other factors that can contribute to non-adherence to treatment include "inaccessibility of treatment centres, unfriendly staff, lack of patient education skills, shortages and irregularities in drug supplies, and logistics other than patient awareness about the disease" [54]. Misdiagnosis (inadequate knowledge) and stigma among health workers also contribute to late diagnosis [78]. It is important to address and avoid delays in seeking treatment, as these may result in permanent, visible disabilities [1]. This in turn can influence the perception of leprosy [61,62].

Factors that can reduce the effects of perceptions and stigma on people's lives

There are several factors that can have a mitigating effect on the impact of stigma on persons affected. Important factors include but are not limited to social support, psychological support and counselling, socio-economic rehabilitation, resilience, and personal empowerment. These factors are related and can strengthen each other.

Social support enables people to cope with stress [82]. Quality relationships with others, such as family, friends and peers, can help people regulate their emotions, and can provide practical support and help solve problems [83,84]. Peer support in particular can help mitigate the impact of stigma, by creating a sense of autonomy and community [85], and by helping stigmatised individuals to develop a positive self-image [85]. Peer support enhances people's sense of empowerment. Social support is positively associated with psychosocial wellbeing [86], quality of life [87,88] and resilience [89].

Psychological support and counselling are important for the successful treatment of persons diagnosed with leprosy [90]. Counselling can help individuals cope with hardship and challenges [91]. (Lay and peer) counselling has also been found to reduce internalised stigma, create hope, improve self-image, improve social participation and improve quality of life [92].

Socio-economic rehabilitation can diminish the (social) consequences of leprosy – it has been found to increase financial means, dignity, independence and social participation [93]. Persons affected by leprosy often hold a marginalised position in their communities, poverty and a lack of resources for income generation are challenges [94,95]. Costs for treatment and reduced ability to work may cause a financial burden on the entire household [95]. Micro-credit loans and vocational training, for example, can improve the financial situation of persons affected, but can also reduce community stigma by protecting people against loss of social value and by facilitating their engagement in daily (social) life [96,97].

Resilience is the process of successfully adapting to or managing challenging life experiences and adversity, especially through mental, emotional and behavioural flexibility [98,99]. Resilience is not a personality trait, but a dynamic process that changes across the lifespan [98]. Resilience-related skills and thinking can help people cope with and overcome adversity, such as experiences of stigma [100].
Social support networks are important for fostering resilience also [83]. In addition, counselling may also help people to become more resilient [91].

Empowerment is the opposite of internalised stigma. It involves “power, control, activism, righteous indignation, and optimism” [101]. Disclosure about one’s condition can reduce worry and concern over secrecy, and can promote a sense of power and control [101]. Resilience and empowerment are related. However, where empowerment is enacted socially – aimed at external change to relationships and power dynamics, resilience consists of internal goals aimed at intrapersonal actions and outcomes [102]. The factors mentioned above, e.g. counselling, building resilience and peer support are all mechanisms of empowerment also [66].

There are also more static factors that mitigate the effect of stigma and perceptions. The impact of perceptions and stigma largely depend on a person’s position in the family and community. Stigma is not a constant, it takes different forms in different stages of a person’s life, and in different networks of relationships [48]. Stigmatisation is based on interactions between people, and hierarchy can in part explain the difference in impact of stigma between people. Stigma feeds on and strengthens already existing inequalities between people in class, age, gender and sexuality [66]. Factors like gender, wealth, higher social status can potentially mitigate the impact of perceptions also.

Changing perception

There are several strategies and interventions to change the perception of leprosy. Many of these are similar to interventions for stigma reduction. Interventions that aim to reduce stigma often address the causes of stigma, such as beliefs and attitudes that lead to labelling, stereotyping and discrimination [41]. Strategies are interrelated. Interventions in one area often have effect in other areas also. For example, improving knowledge and changing beliefs, can in turn change attitudes. In addition, interventions often have benefits beyond their original aim. For example, socio-economic empowerment has been found to not only improve the economic situation of persons affected, but to also improve mental wellbeing and reduce community stigma [96,97]. Socio-economic rehabilitation can increase people's dignity and social position in society [93]. The strategies discussed below are interrelated, just like the components of perception (knowledge, attitudes, emotions and beliefs).

Crucial to changing perceptions are understanding the local context, and understanding and addressing the (social, political and economic) causes of these perceptions [41,55,103]. Interventions should fit the audience [104]. Contextualised interventions are more effective than generic interventions [54]. In addition, effective interventions affect multiple levels of an ecological system in multiple ways [105].

Changing disease perception

The image of leprosy, the way leprosy is seen (i.e. disease or illness perception), is often one of impairments and disabilities [77]. Because only people with less visible signs of leprosy are able to hide their condition, the image of leprosy as a disease with deformities is not challenged [77]. Leprosy is also sometimes perceived as an incurable or highly contagious disease [57,58]. These perceptions may elicit emotional responses such as fear. It is likely that the distribution of post-exposure prophylaxis can challenge some of these beliefs and fears. Physical and socio-economic rehabilitation can also positively influence the community perception of leprosy, and self-perception and self-esteem of persons affected [58]. The mitigating factors mentioned previously (social support, counselling, socio-economic rehabilitation, resilience and personal empowerment) can help persons affected to regain their social identity and can positively influence the perception of (persons affected by) leprosy.

The way persons affected are portrayed in the media reflects, defines, and perpetuates public perceptions of those who are portrayed [59]. This applies to language, terminology and images [106]. The internet is filled with negative images of leprosy. These images, often of persons affected by leprosy with disabilities, are used to raise awareness and donations. These pictures, but also discriminatory language, reinforce the negative perception of leprosy and are not representative of the disease [107,108]. Positive images of persons affected can challenge and change the perceptions of negative differences between social groups [59]. An example of a project that promotes a positive image of leprosy, is the recently launched "New Face of Leprosy Project", in which persons affected are portrayed enjoying life and functioning in society [107,108]. The media can be a source of stigma through the negative portrayal of persons affected, and can play a role in reducing stigma by raising awareness [109–111].

Changing the self-perception of persons affected

Interventions that aim to reduce internalised negative stereotypes aim to change individual characteristics such as knowledge, behaviour and coping skills [42]. Interventions and strategies such as counselling can help persons affected to cope with their disease and can reduce internalised stigma. Strategies to change self-perception include changing illness perception, improving self-esteem, empowerment, support groups, and help-seeking behaviour and altering negative beliefs and attitudes of the person affected [42,112]. During cognitive behavioural therapy for example, individuals are trained to identify and challenge negative beliefs and interpretations [42]. Resilience and resilience-related protective factors such as social resources, emotional regulation and problem solving abilities can also help people cope with and overcome adversity, such as experiences of stigma [100]. In addition, peer support can create a sense of autonomy and community [85], and can help individuals to develop a positive self-image [85].

Improving knowledge and changing beliefs

Knowledge and beliefs are formed by direct observation and experience, and by information provided by outside sources such as newspapers, books, radio and interaction with other people [18]. Improving knowledge and changing beliefs is often done through health education. The purpose of health education is usually twofold: increasing awareness of leprosy and changing negative (community) attitudes towards persons affected by leprosy. It is important that educational needs and socio-cultural beliefs of the target group are understood when materials are developed. Health education can be incorporated into posters and flyers, but also into songs, movies, games and puppet shows [42,54]. It is important that education about leprosy is directed at both those with and without leprosy. Teaching persons affected about their condition empowers them and provides them with the knowledge and confidence to counteract discriminatory behaviour and misconceptions they may face from others. This can also decrease internalised stigma [92]. Community members of persons affected need to be educated as well, ideally by a local person. It can help to target specific groups, such as village leaders (the information will ‘filter down’). Knowledge about for example treatment and cause of leprosy can change people’s perspective on leprosy [58]. Because beliefs form the basis for the formation of attitudes, changing beliefs can result in changing attitudes also [18].

Changing attitudes

Since attitudes refer to a person's feelings toward and evaluation of an aspect of the person's world, and are based on beliefs about an attitude object, that in turn stem from learning and experiences [10,18], they can also be changed. Attitudes can change when we are exposed to new experiences and information [20]. They are however, complex and difficult to change, especially when rooted in in religious belief systems and when functional, for instance when they help us fit in with a social group [20]. Views and attitudes of key influential people or opinion leaders, such as health workers or religious leaders, can influence how people perceive leprosy [58]. Negative attitudes among health workers can in part be addressed by leprosy awareness training and by improving knowledge about leprosy [58]. In addition, compassion training could potentially reduce health worker stigma. Randomised controlled trials have demonstrated significant effects of compassion-based training in improving compassionate responses to suffering, both in clinical and non-clinical settings [113–115].

Attitudes are a societal product and are influenced by social communication and learning [20]. “Contact interventions”, interactions between persons affected and the public, are therefore an effective strategy to reduce negative attitudes – because it changes people’s experience with (and beliefs about) persons affected. Contact with persons affected makes people realise their attitudes are no longer functional and thus opens the way to change [20]. Contact demystifies incorrect information, and challenges stereotypes [42]. Interventions that stimulate interaction between people with and without leprosy, ‘contact interventions’, have been found to be effective in increasing knowledge and changing public attitudes regarding leprosy also [116].

Aim and research questions

The overall aim of this thesis is to contribute to a better understanding of the perception of leprosy and to explore interventions to change the perception of leprosy at individual, family, and community level. This thesis includes studies on the assessment of the perception of leprosy, the impact of leprosy on marital and family life, and interventions to change this perception and reduce this impact.

This thesis consists of three parts. Part 1 of this thesis addresses the perception of leprosy. Part 2 of addresses the impact of leprosy on marital and family life. In part 3, interventions to change perception at community level and interventions to reduce the impact of leprosy at individual and family level are explored.

This thesis will address the following research questions:

1. What are determinants of perception and knowledge of leprosy in endemic communities?
2. How does leprosy impact marital and family life?
3. How effective are posters and community meetings in changing perception and knowledge of leprosy at community level?
4. What interventions have the potential to reduce the impact of leprosy at individual and family level?

Outline of this thesis

This thesis consists of 12 chapters. **Chapters 2 and 3** describe the perceptions and knowledge of leprosy in endemic districts in India and Indonesia. **Chapter 4** covers the methodology used to assess perception of leprosy. **Chapters 5 and 6** covers the experiences of persons affected and their family members regarding the impact of leprosy on their lives, to explore the impact of leprosy on marital and family life. **Chapter 7** describes the development and evaluation of two interventions, posters, and community meetings, to change perception and knowledge of leprosy at community level in endemic districts in India. **Chapter 8** describes the development and evaluation of a family-based intervention for prevention and self-management of disabilities due to three diseases, including leprosy, in Ethiopia. **Chapter 9** explores sources of strength and resilience for persons affected by leprosy in Brazil. **Chapter 10** describes the development and evaluation of an intervention to strengthen individual and family resilience against leprosy-related discrimination. The main findings of chapter 2 to 10 are discussed and reflected on in **chapter 11**.

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References

1. Suzuki K, Akama T, Kawashima A, Yoshihara A, Yotsu RR, Ishii N. Current status of leprosy: epidemiology, basic science and clinical perspectives. J Dermatol 2012;39:121–9.
2. Han XY, Seo Y-H, Sizer KC, Schoberle T, May GS, Spencer JS, et al. A new Mycobacterium species causing diffuse lepromatous leprosy. Am J Clin Pathol 2008;130:856–64.
3. Lastória JC, Abreu MAMM de. Leprosy: review of the epidemiological, clinical, and etiopathogenic aspects-part 1. An Bras Dermatol 2014;89:205–18.
4. Bhutta ZA, Sommerfeld J, Lassi ZS, Salam RA, Das JK. Global burden, distribution, and interventions for infectious diseases of poverty 2014.
5. World Health Organization. Weekly Epidemiological Record, 4 September 2020. Global leprosy (Hansen disease) update, 2019: time to step-up prevention initiatives. 2020;95:417–40.
6. Bakker MI, Hatta M, Kwenang A, Van Benthem BHB, Van Beers SM, Klatser PR, et al. Prevention of leprosy using rifampicin as chemoprophylaxis. Am J Trop Med Hyg 2005;72:443–8.
7. Moet FJ, Pahan D, Oskam L, Richardus JH. Effectiveness of single dose rifampicin in preventing leprosy in close contacts of patients with newly diagnosed leprosy: cluster randomised controlled trial. Bmj 2008;336:761–4.
8. Richardus R, Alam K, Kundu K, Roy JC, Zafar T, Chowdhury AS, et al. Effectiveness of single-dose rifampicin after BCG vaccination to prevent leprosy in close contacts of patients with newly diagnosed leprosy: A cluster randomized controlled trial. Int J Infect Dis 2019;88:65–72.
9. Mieras L, Singh MK, Manglani PR, Arif MM, Banstola NL, Pandey B, et al. A single-dose of rifampicin to prevent leprosy; quantitative analysis of impact on perception, attitudes and behaviour of persons affected, contacts and community members towards leprosy in India, Nepal and Indonesia. Lepr Rev 2020;91:314–27.
10. Pickens J. Attitudes and perceptions. Organ Behav Heal Care 2005;4.
11. Hiebert PG. Transforming worldviews: An anthropological understanding of how people change. Baker Academic; 2008.
12. Broadbent E, Wilkes C, Koschwanez H, Weinman J, Norton S, Petrie KJ. A systematic review and meta-analysis of the Brief Illness Perception Questionnaire. Psychol Health 2015;30:1361–85.
13. Ajzen I, Fishbein M, Lohmann S, Albarracín D. The influence of attitudes on behavior. Handb. attitudes Vol. 1 Basic Princ., Routledge/Taylor & Francis Group; 2019, p. 197–255.
14. Eagly AH, Chaiken S. The psychology of attitudes. Harcourt brace Jovanovich college publishers; 1993.
15. Alexander PA, Dochy FJRC. Conceptions of knowledge and beliefs: A comparison across varying cultural and educational communities. Am Educ Res J 1995;32:413–42.
16. Hutner TL, Markman AB. Proposing an operational definition of science teacher beliefs. J Sci Teacher Educ 2016;27:675–91.
17. Ferguson MJ, Bargh JA. How social perception can automatically influence behavior. TRENDS Cogn Sci 2004;8:33.
18. Fishbein M, Ajzen I. Belief, attitude, intention, and behavior: An introduction to theory and research. Addison-Wesley: Reading, MA, USA; 1975.
19. Pritchard D. What is this thing called knowledge? Routledge; 2018.

20. Dalal AK. Social interventions to moderate discriminatory attitudes: the case of the physically challenged in India. Psychol Health Med 2006;11:374–82.
21. Plutchik R. The nature of emotions: Human emotions have deep evolutionary roots, a fact that may explain their complexity and provide tools for clinical practice. Am Sci 2001;89:344–50.
22. Helm BW. Emotions as evaluative feelings. Emot Rev 2009;1:248–55.
23. Ochsner KN, Schacter DL. A social cognitive neuroscience approach to emotion and memory. Neuropsychol Emot 2000:163–93.
24. Scherer KR. What are emotions? And how can they be measured? Soc Sci Inf 2005;44:695–729.
25. Organization WH. A practical manual for using the International Classification of Functioning, Disability and Health (ICF). Geneva WHO 2013.
26. Grotkamp SL, Cibis WM, Nüchtern EAM, von Mittelstaedt G, Seger WKF. Personal factors in the international classification of functioning, disability and health: prospective evidence. Aust J Rehabil Couns 2012;18:1–24.
27. Larsen RJ, Buss DM, Wismeijer A, Song J. Personality psychology: Domains of knowledge about human nature 2005.
28. Johnson KA, Hill ED, Cohen AB. Integrating the study of culture and religion: Toward a psychology of worldview. Soc Personal Psychol Compass 2011;5:137–52.
29. Snyder M, Tanke ED, Berscheid E. Social perception and interpersonal behavior: On the self-fulfilling nature of social stereotypes. J Pers Soc Psychol 1977;35:656.
30. Bay JL, Mora HA, Sloboda DM, Morton SM, Vickers MH, Gluckman PD. Adolescent understanding of DOHaD concepts: a school-based intervention to support knowledge translation and behaviour change. J Dev Orig Health Dis 2012;3:469–82.
31. Barr RG, Barr M, Fujiwara T, Conway J, Catherine N, Brant R. Do educational materials change knowledge and behaviour about crying and shaken baby syndrome? A randomized controlled trial. Cmaj 2009;180:727–33.
32. Ebenso B, Newell J, Emmel N, Adeyemi G, Ola B. Changing stigmatisation of leprosy: an exploratory, qualitative life course study in Western Nigeria. BMJ Glob Heal 2019;4:e001250.
33. Singh R, Singh B, Mahato S. Community knowledge, attitude, and perceived stigma of leprosy amongst community members living in Dhanusha and Parsa districts of Southern Central Nepal. PLoS Negl Trop Dis 2019;13:e0007075.
34. Opala J, Boillot F. Leprosy among the Limba: illness and healing in the context of world view. Soc Sci Med 1996;42:3–19.
35. Van Brakel WH. Measuring health-related stigma—a literature review. Psychol Health Med 2006;11:307–34.
36. Litt E, Baker MC, Molyneux D. Neglected tropical diseases and mental health: a perspective on comorbidity. Trends Parasitol 2012;28:195–201.
37. Staring ABP, Van der Gaag M, Van den Berge M, Duivenvoorden HJ, Mulder CL. Stigma moderates the associations of insight with depressed mood, low self-esteem, and low quality of life in patients with schizophrenia spectrum disorders. Schizophr Res 2009;115:363–9.
38. Weiss MG, Ramakrishna J, Somma D. Health-related stigma: rethinking concepts and interventions. Psychol Health Med 2006;11:277–87.

39. Van Brakel WH, Cataldo J, Grover S, Kohrt BA, Nyblade L, Stockton M, et al. Out of the silos: identifying cross-cutting features of health-related stigma to advance measurement and intervention. BMC Med 2019;17:13.
40. Goffman E. Stigma: Notes on the management of spoiled identity. Simon and Schuster; 2009.
41. Link BG, Phelan JC. Conceptualizing stigma. Annu Rev Sociol 2001;27:363–85.
42. Heijnders M, Van Der Meij S. The fight against stigma: an overview of stigma-reduction strategies and interventions. Psychol Health Med 2006;11:353–63.
43. Rao D, Elshafei A, Nguyen M, Hatzenbuehler ML, Frey S, Go VF. A systematic review of multi-level stigma interventions: state of the science and future directions. BMC Med 2019;17:1–11.
44. Link BG, Struening EL, Rahav M, Phelan JC, Nuttbrock L. On stigma and its consequences: evidence from a longitudinal study of men with dual diagnoses of mental illness and substance abuse. J Health Soc Behav 1997:177–90.
45. Thornicroft G, Rose D, Kassam A, Sartorius N. Stigma: ignorance, prejudice or discrimination? Br J Psychiatry 2007;190:192–3.
46. Weiss MG. Stigma and the social burden of neglected tropical diseases. PLoS Negl Trop Dis 2008;2:e237.
47. Scambler G. Stigma and disease: changing paradigms. Lancet 1998;352:1054–5.
48. Staples J. Interrogating leprosy'stigma': why qualitative insights are vital. Lepr Rev 2011;82:91.
49. Deacon H. Understanding HIV/AIDS stigma: A theoretical and methodological analysis. HSRC press; 2005.
50. Sermrittirong S, van Brakel WH, Bunders-Aelen JFG. How to reduce stigma in leprosy–a systematic literature review. Lepr Rev 2014;85:149–57.
51. Tabah EN, Nsagha DS, Bissek A-CZ-K, Njamnshi TN, Njih IN-N, Pluschke G, et al. Community knowledge, perceptions and attitudes regarding leprosy in rural Cameroon: The case of Ekondotiti and Mbonge health districts in the South-west Region. PLoS Negl Trop Dis 2018;12:e0006233.
52. van't Noordende AT, Korfage I, Lisam S, Arif MA, Kumar A, van Brakel WH. The role of perceptions and knowledge of leprosy in the elimination of leprosy: A baseline study in Fatehpur district, northern India. PLoS Negl Trop Dis 2019;13:e0007302.
53. Sayce L. Stigma, discrimination and social exclusion: What's in a word? J Ment Heal 1998;7:331–43.
54. Brown W. Can social marketing approaches change community attitudes towards leprosy? Lepr Rev 2006;77:89–98.
55. Rao PS. Study on differences and similarities in the concept and origin of leprosy stigma in relation to other health-related stigma. Indian J Lepr 2010;82:117–21.
56. Stangl AL, Earnshaw VA, Logie CH, van Brakel W, Simbayi LC, Barré I, et al. The health stigma and discrimination framework: a global, crosscutting framework to inform research, intervention development, and policy on health-related stigmas. BMC Med 2019;17:1–13.
57. Sermrittirong S, Van Brakel W. Stigma in leprosy: concepts, causes and determinants. Lepr Rev 2014;85:36–47.
58. Rafferty J. Curing the stigma of leprosy. Lepr Rev 2005;76:119–26.
59. Susman J. Disability, stigma and deviance. Soc Sci Med 1994;38:15–22.
60. Jones EE. Social stigma: The psychology of marked relationships. WH Freeman; 1984.

61. Navon L. Beggars, metaphors, and stigma: a missing link in the social history of leprosy. Soc Hist Med 1998;11:89–105.
62. Adhikari B, Kaehler N, Raut S, Marahatta SB, Ggyanwali K. Risk factors of stigma related to leprosy-A systematic review. J Manmohan Meml Inst Heal Sci 2013;1:3–11.
63. Hofstraat K, van Brakel WH. Social stigma towards neglected tropical diseases: a systematic review. Int Health 2016;8:i53–70.
64. Adhikari B, Shrestha K, Kaehler N, Raut S, Chapman SR. Community attitudes towards leprosy affected persons in Pokhara municipality of western Nepal. J Nepal Health Res Counc 2014.
65. Seshadri D, Khaitan BK, Khanna N, Sagar R. The tangled web: a study of knowledge and attitude towards leprosy from a tertiary care hospital in India. Indian J Lepr 2014:27–41.
66. Heijnders ML. The dynamics of stigma in leprosy. Int J Lepr 2004;72:437–47.
67. Henry M, GalAn N, Teasdale K, Prado R, Amar H, Rays MS, et al. Factors contributing to the delay in diagnosis and continued transmission of leprosy in Brazil–an explorative, quantitative, questionnaire based study. PLoS Negl Trop Dis 2016;10:e0004542.
68. Somar PMW, Waltz MM, van Brakel WH. The impact of leprosy on the mental wellbeing of leprosy-affected persons and their family members–a systematic review. Glob Ment Heal 2020;7.
69. Hatzenbuehler ML, Phelan JC, Link BG. Stigma as a fundamental cause of population health inequalities. Am J Public Health 2013;103:813–21.
70. van Brakel WH. Measuring leprosy stigma-a preliminary review of the leprosy literature1. Int J Lepr Other Mycobact Dis 2003;71:190.
71. Rao VP, Rao IR, Palande DD. Socio-economic rehabilitation programmes of LEPRA India-methodology, results and application of needs based socio-economic evaluation. Lepr Rev 2000;71:466–71.
72. Awofeso N. Stigma and socio-economic reintegration of leprosy sufferers in Nigeria. Acta Leprol 1996;10:89–91.
73. van't Noordende AT, Aycheh MW, Schippers A. The impact of leprosy, podoconiosis and lymphatic filariasis on family quality of life: A qualitative study in Northwest Ethiopia. PLoS Negl Trop Dis 2020;14:e0008173.
74. Goffman E. Stigma and social identity 1963.
75. Van't Noordende AT, Aycheh MW, Schippers AP. An exploration of family quality of life in persons with leprosy-, lymphatic filariasis–and podoconiosis-related disabilities and their family members in Ethiopia. Trans R Soc Trop Med Hyg 2020;114:1003–12.
76. Try L. Gendered experiences: marriage and the stigma of leprosy. Asia Pacific Disabil Rehabil J 2006;17:45–6.
77. Engelbrektsson U-B, Subedi M. Stigma and leprosy presentations, Nepal. Lepr Rev 2018;89:1–8.
78. Da Silva Souza C, Bacha JT. Delayed diagnosis of leprosy and the potential role of educational activities in Brazil. Lepr Rev 2003;74:249–58.
79. Robertson LM, Nicholls PG, Butlin R. Delay in presentation and start of treatment in leprosy: experience in an out-patient clinic in Nepal. Lepr Rev 2000;71:511–6.
80. Barrett R. Self-mortification and the stigma of leprosy in northern India. Med Anthropol Q 2005;19:216–30.
81. Heijnders ML. Experiencing leprosy: perceiving and coping with leprosy and its treatment. A qualitative study conducted in Nepal. Lepr Rev 2004;75:327–37.

82. Gurung R, Sarason B, Sarason I. Close personal relationships and health outcomes: A key to the role of social support. Handb Pers Relationships Theory, Res Interv (2nd Ed) Chichester, UK Wiley 1997:547–73.
83. Earvolino-Ramirez M. Resilience: A Concept Analysis. Nurs. Forum, vol. 42, 2007, p. 73.
84. Cohen S. Psychosocial models of the role of social support in the etiology of physical disease. Heal Psychol 1988;7:269.
85. Mead S, Hilton D, Curtis L. Peer support: A theoretical perspective. Psychiatr Rehabil J 2001;25:134.
86. Thoits PA. Mechanisms linking social ties and support to physical and mental health. J Health Soc Behav 2011;52:145–61.
87. Baumeister RF, Leary MR. The need to belong: desire for interpersonal attachments as a fundamental human motivation. Psychol Bull 1995;117:497.
88. Connell J, Brazier J, O'Cathain A, Lloyd-Jones M, Paisley S. Quality of life of people with mental health problems: a synthesis of qualitative research. Health Qual Life Outcomes 2012;10:138.
89. Southwick SM, Sippel L, Krystal J, Charney D, Mayes L, Pietrzak R. Why are some individuals more resilient than others: the role of social support. World Psychiatry 2016;15:77.
90. Organization WH. Guidelines for strengthening participation of persons affected by leprosy in leprosy services. WHO Regional Office for South-East Asia; 2011.
91. Enache R. Psychological counseling and developing resilience for young people with psychological trauma in childhood or adolescence. J Exp Psychother 2016;19:73.
92. Lusli M, Peters R, van Brakel W, Zweekhorst M, Iancu S, Bunders J, et al. The impact of a rights-based counselling intervention to reduce stigma in people affected by leprosy in Indonesia. PLoS Negl Trop Dis 2016;10:e0005088.
93. Ebenso B, Fashona A, Ayuba M, Idah M, Adeyemi G, S-Fada S. Impact of socio-economic rehabilitation on leprosy stigma in Northern Nigeria: findings of a retrospective study. Asia Pacific Disabil Rehabil J 2007;18:98–119.
94. Deepak S, Hansine PE, Braccini C. Self-care groups of leprosy-affected people in Mozambique. Lepr Rev 2013;84:283.
95. Tembei AM, Kengne-Ouaffo JA, Ngoh EA, John B, Nji TM, Deribe K, et al. A comparative analysis of economic cost of podoconiosis and leprosy on affected households in the northwest region of Cameroon. Am J Trop Med Hyg 2018;98:1075–81.
96. Ebenso B, Ayuba M. "Money is the vehicle of interaction": insight into social integration of people affected by leprosy in northern Nigeria. Lepr Rev 2010;81:99.
97. Dadun D, Van Brakel WH, Peters RMH, Lusli M, Zweekhorst M, Bunders JGF, et al. Impact of socio-economic development, contact and peer counselling on stigma against persons affected by leprosy in Cirebon, Indonesia–a randomised controlled trial. Lepr Rev 2017;88:2–22.
98. Windle G. What is resilience? A review and concept analysis. Rev Clin Gerontol 2011;21:152–69.
99. Sivilli TI, Pace TWW. The Human Dimensions of Resilience: A Theory of Contemplative Practices. Garrison Inst 2014.
100. Shih M. Positive stigma: Examining resilience and empowerment in overcoming stigma. Ann Am Acad Pol Soc Sci 2004;591:175–85.
101. Corrigan PW, Rao D. On the self-stigma of mental illness: Stages, disclosure, and strategies for change. Can J Psychiatry 2012;57:464–9.

102. Brodsky AE, Cattaneo LB. A transconceptual model of empowerment and resilience: Divergence, convergence and interactions in kindred community concepts. Am J Community Psychol 2013;52:333–46.
103. Parker R, Aggleton P. HIV and AIDS-related stigma and discrimination: a conceptual framework and implications for action. Soc Sci Med 2003;57:13–24.
104. Birbeck G. Interventions to reduce epilepsy-associated stigma. Psychol Health Med 2006;11:364–6.
105. Cook JE, Purdie-Vaughns V, Meyer IH, Busch JTA. Intervening within and across levels: A multilevel approach to stigma and public health. Soc Sci Med 2014;103:101–9.
106. van Brakel WH, Gopal PK. Portraying a positive image of persons (previously) affected by leprosy. Int J Lepr Other Mycobact Dis 1999;67:477.
107. Kumar A, Lambert S, Lockwood DNJ. Picturing health: a new face for leprosy. Lancet 2019;393:629–38.
108. Lockwood DNJ, Bradley T. New face for leprosy “A positive image for leprosy.” Lepr Rev 2020;91:2–13.
109. Chapple A, Ziebland S, McPherson A. Stigma, shame, and blame experienced by patients with lung cancer: qualitative study. Bmj 2004;328:1470.
110. Bekalu MA, Eggermont S, Ramanadhan S, Viswanath K. Effect of media use on HIV-related stigma in sub-Saharan Africa: A Cross-Sectional Study. PLoS One 2014;9:e100467.
111. Niederkrotenthaler T, Reidenberg DJ, Till B, Gould MS. Increasing help-seeking and referrals for individuals at risk for suicide by decreasing stigma: the role of mass media. Am J Prev Med 2014;47:S235–43.
112. Mittal D, Sullivan G, Chekuri L, Allee E, Corrigan PW. Empirical studies of self-stigma reduction strategies: a critical review of the literature. Psychiatr Serv 2012;63:974–81.
113. Kirby JN, Tellegen CL, Steindl SR. A meta-analysis of compassion-based interventions: Current state of knowledge and future directions. Behav Ther 2017;48:778–92.
114. Kirby JN. Compassion interventions: The programmes, the evidence, and implications for research and practice. Psychol Psychother Theory, Res Pract 2017;90:432–55.
115. Ferrari M, Hunt C, Harrysunker A, Abbott MJ, Beath AP, Einstein DA. Self-compassion interventions and psychosocial outcomes: A meta-analysis of RCTs. Mindfulness (N Y) 2019;10:1455–73.
116. Peters RMH, Zweekhorst MBM, Bunders JFG, van Brakel WH. A cluster-randomized controlled intervention study to assess the effect of a contact intervention in reducing leprosy-related stigma in Indonesia. PLoS Negl Trop Dis 2015;9:e0004003.

PART 1

The perception of leprosy

CHAPTER 2

The role of perceptions and knowledge of leprosy in the elimination of leprosy: a baseline study in Fatehpur district, northern India

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Abstract

Background

With the introduction of new interventions to prevent leprosy, such as post-exposure prophylaxis (PEP) given to contacts of leprosy patients, it is necessary to update our understanding of knowledge and perception of leprosy among the populations where these interventions will be introduced, in order to tailor communication optimally to the current situation. This study is a baseline study of the PEP++ project and aimed to assess the knowledge, attitudes and practices regarding leprosy in Fatehpur, India.

Methodology

The study used a community-based cross-sectional design with a mixed-methods approach. We assessed knowledge, attitudes, and practices with the KAP measure, and stigma with the Explanatory Model Interview Catalogue community stigma scale (EMIC-CSS) and the Social Distance Scale (SDS). In addition, semi-structured interviews and focus group discussions were conducted with all participant groups. The quantitative data were analysed using stepwise multivariate regression. The qualitative data were analysed using open, inductive coding and content analysis.

Findings

A total of 446 participants were included in the study: 100 persons affected by leprosy, 111 close contacts, 185 community members and 50 health care workers. In addition, 24 in-depth interviews were conducted and 35 people were included in focus group discussions. 12.5% of the participants had adequate knowledge of leprosy, while 22% had poor knowledge. Knowledge on cause (answered correctly by 10% of the participants), mode of transmission (5%) and symptoms of leprosy (16%) was especially poor. The mean EMIC-CSS score was 15.3 (95%CI 14.6-16.0) and mean SDS score 7.2 (95%CI 6.6-7.8). Better knowledge of leprosy was associated with lower levels of social distance towards persons affected by leprosy.

Conclusion

This study revealed poor knowledge regarding leprosy and high levels of stigma and fear and desire to keep social distance towards persons affected by leprosy. Community education that takes cultural beliefs, knowledge gaps and fears into consideration could improve knowledge, reduce misconceptions and positively influence the perception of leprosy.

Introduction

Leprosy is an infectious disease caused by *Mycobacterium leprae*. Leprosy primarily affects the peripheral nerves and skin. The damage of the nerves may affect the sensory, motor and autonomic functions of the nerves, resulting ultimately in disability [1,2]. In addition to the physical consequences of leprosy, social stigmatization is a challenge for many persons affected by leprosy, especially since this often remains once the medical treatment is finished [3–5].Transmission of the bacteria is believed to occur through long-term exposure of the respiratory system to airborne nasal droplets [1,2,6].

With over 210,000 new patients diagnosed in the world each year, leprosy is still a public health problem in many low and middle income countries [2,7]. To interrupt the transmission of *M. leprae* and to reduce the number of new leprosy patients globally early detection and prompt treatment with multi-drug therapy (MDT) are essential [2,8,9]. Early detection is also necessary to reduce the physical and social consequences of the disease as the complications of leprosy depend on how timely in the disease process leprosy is diagnosed and treated [8,10]. Prevention of disability thus begins with early detection of leprosy [8].

Late detection of leprosy is associated with misdiagnosis, inadequate or incorrect knowledge about the disease as well as negative beliefs about leprosy among persons affected and health care workers [10]. People's perception of leprosy influences their awareness that certain signs and symptoms may be due to leprosy [10,11]. Indeed, voluntary and early reporting of leprosy requires awareness of leprosy and its treatment facilities [12]. Several studies attributed delayed diagnosis of leprosy to the use of traditional medicine and/or low awareness of modern treatment, ignorance of leprosy, a belief in self-cure, unavailability of services or skilled health care workers, stigma and the influence of traditional or community leaders [10,13–17]. In addition, because of the stigma associated with leprosy, persons affected by leprosy often delay seeking treatment until they develop permanent, visible disabilities [2]. This set of interrelated factors suggests that sufficient knowledge of leprosy presentation, clinical features and services and stigma reduction are essential for early detection of leprosy.

Improving the knowledge and perception of leprosy and reducing stigma seem essential to improve strategies for early case detection in leprosy. There have been several studies already that looked into the knowledge, attitudes and practices regarding leprosy of persons affected, the general community, students as well as health care workers. In India alone, over 14 studies that looked into this have been conducted after the year 2000 [12,18,27–30,19–26]. Most of these studies found low levels of knowledge about leprosy and negative attitudes towards persons affected by leprosy. However, most studies administered questionnaires only: only two of these studies conducted in-depth interviews [21,24] and one study used a mixed-methods approach [30]. With the introduction of new interventions to prevent leprosy, such as post-exposure prophylaxis (PEP) given to contacts of leprosy patients, it is necessary to update our understanding of knowledge and perception of leprosy among the populations where these interventions will be introduced, in order to tailor communication optimally to the current situation.

The current study aimed to assess the knowledge, attitudes and practices regarding leprosy in a leprosy endemic district (Fatehpur district in Uttar Pradesh, India) using a mixed-methods approach. A mixed-methods approach will allow us to quantify the knowledge levels as well as gain more insight into the rationale behind people's perceptions, beliefs and attitudes. We expect that the findings of this study will give insight in the existing knowledge of leprosy, myths or misconceptions, as well as the prevailing attitudes, beliefs and specific fears and concerns people may have about leprosy.

Methods

Study design

The study used a community-based cross-sectional design. A mixed methods approach was used. Quantitative questionnaire interviews were used to assess the knowledge and attitudes of people towards leprosy and both semi-structured interviews and focus group discussions were used to ask in-depth questions.

Study site

The study was conducted in Fatehpur district, Uttar Pradesh, northern India between December 2017 and February 2018. Fatehpur is a district in Uttar Pradesh, a state in northern India, where the prevalence of leprosy is high compared to the national average. The prevalence of leprosy in India, that accounts for more than half (60%) of the global disease burden of leprosy, was 0.69 per 10,000 population in April 2015, the prevalence in Fatehpur was 0.77 per 10,000 population [31,32] indicating the need to put in extra efforts.

Study population and sample

Four groups of people were included in the study: (1) persons affected by leprosy or "index patients"; (2) close contacts of index patients; (3) community members; and (4) health care workers.

We collected quantitative and qualitative data. Epi Info StatCalc for cross-sectional studies was used to calculate the quantitative sample size. For the quantitative questionnaire interviews we aimed to include a random sample of at least 100 persons of each target group. This estimate is based on an assumed prevalence of 'negative attitudes' of 50% at baseline and wanting to be able to detect a reduction of 20% or more (i.e. prevalence of negative attitudes in the 2nd survey is 30% or less). Using these parameters, a significance level of 0.05 and a power of 80%, at least 186 subjects are needed in each group, 93 before and 93 after the community education intervention that will be implemented at a later stage as part of the larger research project (PEP++ project). In addition, we conducted interviews to gain more insight into the rationale behind people's perceptions, beliefs and attitudes. We aimed to have semi-structured interviews with six persons from each participant group. We also aimed to conduct one focus group discussion per participant group with seven to ten participants each. The participants in the qualitative sample will be a subset of those in the quantitative sample.

Eligibility criteria

Participants had to live in Fatehpur district. Index patients had to be diagnosed with leprosy within the last five years. Closest contacts included household contacts, family members, neighbours and/or social contacts who have intensive contact with the index patient (at least 20 hours per week for at least three months in the year before the index patient was diagnosed). Community members had to live in the same village or neighbourhood as the index patient. Health care workers had to work in the primary health care centre in the district.

Persons below the age of 16 and persons unwilling or unable to give informed consent were excluded. Close contacts, community members and health care workers were also excluded if they were or had ever been affected by leprosy. Participants who were listed as close contact of an index patient could not participate as community member also.

Sampling methods

The villages were selected by stratified systematic sampling with a random start from among the villages where one or more index patient lived. A list of 13 blocks and 242 villages in these blocks where new leprosy cases were reported in the year 2016 and 2017 was prepared. Every second village from the total number of villages in each block was selected. The first index patient and village that were visited were selected randomly from the list. A total of 114 villages, spread across the 13 blocks, connected to all the 17 primary health care centres in the district were included.

Participants for the quantitative questionnaire interviews were selected as follows:

1. The index patients included in the study were selected by stratified systematic sampling with a random start from a list of leprosy patients registered at the primary health care centre.
2. The close contacts of index patients, i.e. household members, neighbours or social contacts, were selected by the index patient because of their convenient accessibility and proximity. Close contacts were selected by convenience sampling. One contact per index patient was included in the quantitative questionnaire interviews.
3. The community participants were selected by convenience sampling from among those living in the same village or neighbourhood as an index patient. One or two community members per index patient were selected from within a radius of 500 meters of the house of the index patient in the villages where the index patients live. We aimed to select the community members as randomly as possible while trying to ensure an equal number of men and women.
4. Health care workers were selected based on convenience sampling. All primary health care centres in the district were visited, where health care workers were selected based on their availability. We included different types of health care workers: auxiliary nurse midwives, non-medical supervisors and assistants, physiotherapists, paramedical workers, medical workers and district leprosy consultants. Half of the health care workers (n= 25) included in this study had specific responsibilities for leprosy treatment services.

The participants for the qualitative interviews were selected using purposive sampling to ensure adequate representation of age, sex and villages. These participants were a subset of those in the quantitative sample. We used a sampling grid to ensure an equal number of men and women were included.

Data collection

Demographic information was obtained from each participant. In addition, three instruments were used in this study. A knowledge attitudes and practices (KAP) measure and two short stigma instruments: the Explanatory Model Interview Catalogue community stigma scale (EMIC-CSS) and the Social Distance Scale (SDS). All measures were interviewer-administered.

The KAP measure was developed to assess the knowledge, attitudes and perceived practices of index patients, contacts, community members and health care workers regarding leprosy. The KAP measure has been used in several leprosy studies between 2012 and 2017 but the results of these studies have not been published. The questionnaire has 17 items (and consists of single and multiple answer questions). Participants could give multiple answers to some of the KAP questions. Answer options were not suggested to respondents in advance. For the questions for which multiple answers could be given, we considered an answer correct only if the correct answer was given in the absence of incorrect answers. We defined adequate knowledge as 70% or more correct answers on the knowledge section of the KAP measure (≥5 out of 7 questions) Poor knowledge was defined as 30% or less correct answers (≤2 out of 7).

The 15-item EMIC-CSS was used to measure perceived attitudes and behaviour towards persons affected by leprosy. The EMIC-CSS has been validated among community members of persons affected by leprosy in India [33]. The 7-item SDS was used to assess the social distance the interviewee wants to keep towards persons affected by leprosy as a proxy for their attitudes. The SDS has not been formally validated for use with persons affected by leprosy in India, but has been validated among community members of persons affected by leprosy in Indonesia [34]. The SDS has been translated to Hindi, partially validated and used in a study in Uttar Pradesh, India (Ballering et al., in preparation). The EMIC-CSS and SDS were interviewer-administered to community members, close contacts and health care workers.

In addition, cross-sectional data on attitudes and perceptions of the participants towards leprosy were obtained using semi-structured in-depth interviews and focus group discussions.

All interviewers were trained in leprosy, in the instruments used and in the interviewing techniques prior to data collection. Pilot interviews were conducted prior to the final data collection and minor revisions to the interview guide were made. These participants were not included in the final sample and no changes were made to the questionnaires used. All participants were interviewed in their local language by a local interviewer in their home, or at a private space near their home. The in-depth interviews and focus group discussions were audio recorded. A district coordinator monitored the entire process.

Data analysis

Quantitative data were entered in a database created using Epi Info. Simple descriptive methods were used to generate a demographic profile of the study sample. In addition, mean total scores of the KAP, EMIC-CSS and SDS measures were calculated per participant group. Multivariate regression was done to examine which factors had an independent effect on the outcomes (knowledge, attitudes and perceived stigma). We used stepwise multivariate regression with backward elimination to see if there were associations between knowledge, stigma and social distance and the other variables in our dataset (e.g. gender, occupation, etc.). The multivariate analysis was carried out using a model with all variables potentially associated with the outcome with a *p*-value of <0.2 identified through univariate analysis. Variables with *p*-values of ≥0.05 were eliminated one-by-one until all variables that remained in the model were statistically significant (*p*<0.05). For dependent variables that were distributed non-normally we conducted bootstrapped stepwise multivariate regression with backward elimination, as bootstrapping corrects for non-normality by making no assumptions about the shape of the distribution. Data analysis was done in the software packages Epi Info version 7.2.2.2 and SPSS Statistics version 24.

The recordings of the in-depth interviews and focus group discussions were transcribed, translated to English and analysed using open, inductive coding and content analysis. Similar phrases with recurring themes were coded in the software programme Nvivo version 12 and clustered together in tables, to identify connections.

Ethical considerations

Ethical approval was obtained from the Institutional Ethics Committee. Ethical approval for this study was obtained as part of a larger research project: the Post-Exposure Prophylaxis project (PEP++ project). All participants were fully informed about the nature and objective of the study and of confidentiality of the data prior to data collection. Written consent for participation in the study was obtained from each participant in their local language. All persons approached agreed to participate in the study.

Results

Demographic information

A total of 446 participants, of which 285 men (64%) were included in the study. Four groups of people were included in the study: 100 persons affected by leprosy or "index patients" (22%), 111 close contacts of index patients (25%), 185 community members (41%) and 50 health care workers (11%). Most participants (n=395, 88%) lived in rural areas. The average age was 39.2 (range 16-90), men were on average older (41.2) than women (35.7). One fifth of the participants were illiterate (n=95, 21%) and almost one tenth could read and write, but did not have any formal education (n=37, 8%). Four hundred six participants were Hindu (91%). Over half (n=225, 65%) of the participants, excluding index patients, had a close relationship with someone with leprosy. Index patients were diagnosed on average 17.9 months ago (range 9-60 months). Half of the health care workers (n=25, 50%) who were included in this study had specific responsibilities for leprosy treatment services.

Twenty-four in-depth interviews were conducted to supplement the quantitative data. Six people from all four groups (index patients, close contacts, community members and health care workers) were interviewed. Half of them were men (n=12, 50%). The average age of the interviewees was 31 years for women (range 15-55) and 36 years for men (range 20-57). In addition, 35 people were included in focus group discussions (Table 1). Health care workers and community members were the main sources for participants to acquire leprosy relation information. All health care workers who were interviewed in-depth received training on leprosy.

Table 1. Number of participants included in the study, per participant group.

Participant type	Questionnaires*	In-depth interviews	Focus group discussion
Index patient	100	6	9
Close contact	111	6	10
Community member	185	6	7
Health care worker	50	6	9
Total	446	24[a]	35[b]

[a] Index patients were administered the KAP only, while the other participant groups received the KAP, SDS and EMIC-CSS.

[b] The qualitative respondents are a subset of those in the quantitative sample.

Knowledge of leprosy

Table 2 provides an overview of the responses given to the KAP measure.

Seventy participants (16%) correctly answered that both “loss of sensation” and “skin patches” are early symptoms of leprosy. One community member described the early symptoms of leprosy in one of the interviews as:

> *“…Hand or some body parts get numb, or they do not know if a needle is pricked and do not realize that a needle has been kept there. Malformed fingers, water discharge, melting of nails, the body becomes bowed…”*
>
> *(Community member, male, 42)*

When asked what participants thought was the cause of leprosy, two-thirds (67%, n=298) indicated they didn’t know. Few participants (n=43, 10%) only gave the correct answer, “germs or bacteria”, in the absence of wrong answers. A lack of hygiene or cleanliness and eating bad food were often mentioned as causes of leprosy during the in-depth interviews.

Participants were also asked how they thought leprosy is transmitted. There were 11 participants (2%) who only responded that leprosy is transmitted by air. Many of the people who were interviewed in-depth said that they or people from their communities thought that leprosy is transmitted by touch.

A community member described the transmission of leprosy as:

> *“…It can happen because of uncleanliness, by insects, it can spread through touch and by clothes also. (…) Leprosy is a disease which can spread even by touch and it casts effect on the people who live with a leprosy patient. We should stay away from leprosy patient otherwise it can happen to others also…”*
>
> *(Community member, male, 28)*

A health care worker explained:

> *“…Society says not to touch leprosy patients. Then we make them understand and show them by touching patients that it is not a disease that spreads by touch…”*
>
> *(Health care worker, female, 55)*

The majority of participants (n=416, 93%) were aware that leprosy can be treated. Almost all participants (n=412, 97%) who knew leprosy can be treated knew it can be treated by medication. Over half of the participants (n=208, 54%) were aware that leprosy is no longer contagious after a patient has started treatment. Two-thirds of the participants (n=291, 65%) said that the disabilities that some patients have can be prevented, which is correct. Furthermore, when asked if participants thought leprosy was more likely to be temporary or permanent, half (n=212, 48%) of the participants indicated that they thought leprosy was temporary, which is correct.

Table 2. An overview of the responses given per knowledge question. The responses in green are the correct answers.

Topic	**Responses given as percentage of participants who gave the answer as *n* (%).**					**Percentage of people who gave the correct answer only^a (n=446)**
		Persons affected (n=100)	**Contacts (n=111)**	**Community (n=185)**	**Health workers (n=50)**	
Early symptoms	Skin patches	62 (62)	49 (44)	83 (45)	34 (68)	16%
	Loss of sensation	54 (54)	20 (18)	25 (14)	35 (70)	
	Don't know	10 (10)	34 (31)	57 (31)	4 (8)	
	Itchiness	17 (17)	23 (21)	48 (26)	6 (12)	
	Other: tingling, coughing, bleeding, blisters, rashes	19 (19)	20 (18)	22 (12)	14 (28)	
Cause of leprosy	Don't know	82 (82)	83 (75)	119 (64)	14 (28)	
	Germs/bacteria	6 (6)	11 (10)	15 (8)	25 (50)	10%
	Unclean environment	5 (5)	9 (8)	21 (11)	5 (10)	
	Other: punishments for sins, karma, impure blood, hereditary	10 (10)	11 (10)	32 (17)	5 (10)	
Transmission of leprosy	Don't know	65 (65)	65 (59)	84 (45)	13 (26)	
	Skin contact	23 (23)	33 (30)	71 (38)	16 (32)	
	Eating together	11 (11)	13 (12)	17 (9)	4 (8)	
	Other: contaminated soil, insects, 'different'	19 (19)	12 (11)	6 (3)	5 (10)	
	By air	4 (4)	3 (3)	7 (4)	8 (16)	2%

Topic	Responses given as percentage of participants who gave the answer as *n* (%).					Percentage of people who gave the correct answer only[a] (n=446)
		Persons affected (n=100)	**Contacts (n=111)**	**Community (n=185)**	**Health workers (n=50)**	
Treatability of leprosy	Can be treated	97 (97)	102 (93)	168 (91)	49 (98)	93%
	Don't know	1 (1)	7 (6)	12 (6)	1 (2)	
	Can't be treated	2 (2)	1 (1)	5 (3)	0 (0)	
Treated how	By medication	96 (96)	105 (95)	162 (88)	49 (98)	97%
	Other	2 (2)	1 (1)	10 (5)	1 (1)	
Contagiousness	Not contagious when on treatment	45 (45)	56 (50)	81 (44)	26 (52)	54%
	Contagious when on treatment	35 (35)	33 (30)	56 (30)	20 (40)	
	Don't know	17 (17)	15 (14)	31 (17)	3 (6)	
Disabilities	Disabilities can be prevented	61 (61)	72 (65)	117 (63)	41 (82)	65%
	Don't know	22 (22)	28 (25)	43 (23)	2 (4)	
	Disabilities can't be prevented	17 (17)	11 (10)	25 (14)	7 (14)	
Duration of condition	Leprosy is temporary	50 (50)	44 (40)	92 (50)	26 (52)	48%
	Leprosy is permanent	28 (28)	37 (33)	35 (19)	17 (34)	
	Don't know	22 (22)	30 (27)	58 (31)	7 (14)	

[a] In the absence of incorrect answers. Participants could give multiple answers to some of the KAP questions. We choose to present the answers as percentage of participants who gave the answer, rather than as percentage of all the responses given to a particular question. Therefore, the percentages presented may exceed 100%.

Adequate knowledge of leprosy

Participants answered on average 3.2 out of the 7 KAP questions correctly (range 0-7). Two participants answered all seven questions correctly. With a mean of 4.2 correct answers, health care workers had significantly better knowledge scores (*p*=0.042) than the other participants (mean knowledge score 3.1). An overview of the number of correct answers per KAP question can be found in Figure A in S1 Figure.

One in eight participants (n=56, 12.5%) were considered to have adequate knowledge of leprosy, while almost one quarter of the participant (n=99, 22%) were considered to have poor knowledge of leprosy.

Multivariate analysis showed that participants who knew someone affected by leprosy, completed higher education and health care workers all had significantly higher mean levels of knowledge of leprosy (see Table 3).

Table 3. Correlations between level of knowledge about leprosy and the other variables in the dataset. This model explained 16% of the variability of knowledge of leprosy (R-squared= 0.15).

	Regression coefficient	Standard error	*p*-value	*N*
Constant	*2.678*	*.118*	*.000*	
Health care worker	.912	.206	.000	50
Completed higher education	.483	.148	.001	158
Knows someone affected by leprosy*	.345	.134	.011	225

*) The comparison category

Attitudes: questions for index patients only

The final five questions of the KAP measure, about attitudes people have towards persons affected by leprosy, were asked to index patients (*n*=100) only. Most index patients (87%) knew that leprosy can be treated in six to twelve months. Over half of the participants (56%) indicated they would prefer to keep people from knowing they have leprosy. Some participants (22%) indicated that they thought that neighbours, colleagues or others in their community have less respect for them because of their illness. A small proportion of participants (12%) said some people refuse to visit their home even after they have been treated. In addition, eight participants (8%) indicated they decided by themselves to stay away from work or a social group. It is worth noting that most participants replied that they were “not sure” about the answer to the question (ranging from 35% to 60% of the answers given).

Attitudes, stigma and social distance

The EMIC-CSS and SDS were used to assess attitudes and stigma in contacts, community members and health care workers regarding leprosy. An overview of the scores per participant group can be found in Table 4.

Table 4. Mean total scores per participants group. A high score on the KAP measure reflects higher knowledge, whereas high EMIC-CSS and SDS scores reflect higher levels of stigma and desired social distance respectively.

	KAP measure (up to 17 items), range 0-8		EMIC-CSS (17-items), range 0-30		SDS (7-items), range 0-21	
	Mean (95%CI)	Range	Mean (95%CI)	Range	Mean (95%CI)	Range
Index patient	3.3 (3.08-3.52)	0-6	-	-	-	-
Close contact	3.2 (3.00-3.41)	0-5	13.9 (12.7-15.1)	0-26	7.0 (5.99-8.01)	0-21
Community member	3.0 (2.83-3.17)	0-5	16.2 (15.2-17.2)	2-30	8.2 (7.36-9.04)	0-21
Health care worker	4.2 (3.80-4.60)	0-7	14.9 (13.4-16.4)	0-24	4.2 (3.22-5.18)	0-13
All groups	3.2 (3.13-3.35)	0-7	15.3 (14.6-16.0)	0-30	7.2 (6.61-7.79)	0-21

The mean EMIC-CSS score was 15.3 (95%CI 14.6-16.0). Answers to questions that related to marriage and avoidance of persons affected by leprosy most often indicated stigma (see Figure B in S2 Figure). We found that participants who knew a person affected by leprosy had higher mean EMIC-CSS scores and therefore higher levels of perceived stigma, compared to participants who did not know a person affected by leprosy (17.3 vs 14.2, $p<0.001$, independent samples t-test). In addition, being a close contact and doing paid work were associated with lower EMIC-CSS total scores and thus lower levels of stigma (see Table 5). We found that participants who thought that leprosy is caused by an unclean environment or a divine punishment for sins, and participants who thought leprosy transmits through skin contact or by air had significantly higher mean EMIC-CSS scores (see Table 5).

Table 5. Correlations between level of stigma and the other variables in the dataset. This model explained 15% of the variability of stigma towards persons affected by leprosy (R-squared=0.148).

	Regression coefficient	Standard error	*p*-value	*N*
Constant	*15.003*	*1.012*	*.000*	
Thinks leprosy transmits by air	4.461	1.531	.004	18
Thinks leprosy is a divine punishment for sins	3.974	1.667	.018	17
Thinks leprosy is caused by an unclean environment	2.873	1.253	.023	35
Knows someone affected by leprosy*	-2.393	.722	.001	224
Thinks leprosy transmits through skin contact	2.305	.731	.002	120
Indicate they don’t know what causes leprosy	2.208	.859	.011	216
Occupation is paid work	-1.710	.729	.020	115
Close contact	-1.576	.760	.039	110

*) The comparison category

The mean SDS score was 7.2 (95%CI 6.6-7.8). Questions that indicated the most negative attitudes related to marriage and having someone affected by leprosy as caretaker of your children (see Figure C in S3 Figure). We found that health care workers, participants who knew someone affected by leprosy, men, and people with a higher number of correct answers on the KAP measure had lower mean SDS total scores and thus a more positive attitude (see Table 6). Community members, women and illiterate participants had higher mean SDS total scores and thus on average more negative attitudes towards persons affected by leprosy (see Table 6). In addition, participants who said they didn't know the early symptoms of leprosy, participants who thought that leprosy is transmitted by air and participants who thought that leprosy is contagious after treatment also had more negative attitudes (see Table 6).

Table 6. Correlations between level of social distance and the other variables in the dataset. This model explained 19% of the variability of stigma towards persons affected by leprosy (R-squared=0.187).

	Regression coefficient	**Standard error**	***p*-value**	***N***
Constant	*2.356*	*.949*	*.018*	
Thinks leprosy transmits by air	3.915	1.695	.019	18
Illiterate	2.135	.791	.011	71
Doesn't know the early symptoms of leprosy	2.120	.713	.002	95
Health care worker	-2.035	.818	.013	50
Community member	1.931	.637	.003	185
Gender (women)*	1.722	.593	.006	122
Thinks leprosy is contagious	.642	.319	.050	109

*) The comparison category

From the in-depth interviews it became clear that none of the six index patients who were interviewed and only one of the nine index patients who participated in the focus group discussions knew anyone else who was affected by leprosy. In addition, participants often didn't want to disclose because of shame or to avoid negative reactions or social exclusion. One index patient explained:

> *"...No, I did not tell my friend. I kept it hidden (...) because people have a bad perception about leprosy in society. Later people start thinking bad about it [being affected by leprosy] for instance "don't keep him with us"..."*
>
> *(Index patient, male, 20)*

In addition, during the in-depth interviews and focus group discussions, many participants indicated that community members keep their distance from persons affected by leprosy or exclude them from social activities. Community members of participants don't want to talk to, eat with, sit with or touch persons affected by leprosy (six out of the seven community members in the focus group discussion). Persons affected are also often not invited to ceremonies or parties. Avoiding persons affected by leprosy was often linked to the idea of transmission of leprosy by touch (14 out of the 18 non-health workers in the in-depth interviews). Over half of the participants who were interviewed (14 out of the 24 participants in the in-depth interviews) indicated that community members would refrain from touching a person affected by leprosy. Many of them indicated they also thought leprosy transmits via touch (eight participants). "Untouchability" was mentioned often. There were also participants who were positive towards persons affected by leprosy, one community member explained:

> *"…Certainly it [being affected by leprosy] will not make any difference, everyone is given equal respect…"*
>
> *(Community member, male, 42)*

Discussion

Our study revealed poor knowledge regarding leprosy among index patients, close contacts, community members and health care workers in Fatehpur district, Uttar Pradesh, India. There were few participants with adequate knowledge of leprosy, defined as 70% or more correct answers on the knowledge section of the KAP measure (≥5 out of 7 questions). Knowledge on mode of transmission, cause and symptoms of leprosy was especially poor. In addition, we found high levels of perceived stigma and desired social distance towards persons affected by leprosy.

The present study revealed that only 12.5% of the participants had adequate knowledge of leprosy, while 22% had poor knowledge. Similar findings have been reported in other studies in India. Even though other studies did not report a quantified level of knowledge, several reported that knowledge levels among persons affected and their community members were low or inadequate [21,24,28,29]. Two studies found that persons affected by leprosy had higher levels of knowledge about leprosy than community members [22] and family members [19]. This difference was also found in the present study, but was not statistically significant. In the present study, health care workers were found to have better knowledge of leprosy than other participants. This finding is similar, even though knowledge levels appear to be lower, to findings from a study that looked at knowledge, attitudes, and reported practices of health care providers regarding leprosy in Assam, northeast India. In this study over 80% of the participants had attended training programmes on leprosy [30]. In the present study this was 50%. We expect that health care workers who receive training on leprosy will have higher levels of knowledge. This assumption is supported by a study by Rao and colleagues conducted in southeast India, who found that “Medical Officers who received training in leprosy and possessed reference material on leprosy have shown higher knowledge and practice” [23].

In the present study, levels of knowledge about cause (10%), mode of transmission (5%) and early symptoms of leprosy (16%) were poor. This is much lower than other studies, who report up to 28% of correct knowledge on mode of transmission [18,21,22], 26-44% correct knowledge on cause of leprosy [12,18,20–22,24] and 20-73% of correct knowledge on early symptoms [12,20,22,28,29]. Only one study, among community members of urban slums in southern India, reports similar low levels of knowledge on cause, mode of transmission and early symptoms [28]. These low levels of correct knowledge may in part reflect a lack of dissemination of relevant, correct information as participants in the present study reported that their main sources of leprosy-related information were health care workers and community members. Traditional beliefs are likely to be deeply rooted in the Indian culture and can vary from state to state. We believe that traditional beliefs and a lack of knowledge of leprosy play an important role in to the persistence of stigma.

The main misconception related to cause of leprosy was that people thought leprosy is caused by an unclean environment or by a lack of hygiene. An unclean environment, the belief that leprosy is hereditary and bad blood were causes often mentioned in other studies also [12,21,22,24]. In addition, in the present study many people believed that leprosy transmits by touch. Participants reported that community members would refrain from touching a person affected by leprosy and often linked exclusion to “untouchability”. A study among persons affected by leprosy, their family members and people with non-leprosy skin diseases in a tertiary care hospital in Delhi**,** found something similar, stating that “fear of the leprosy-affected and reluctance for physical contact (…) were prominent” [19]. We found only one study in in Madhya Pradesh, central India, from 1981, that explicitly stated that many persons affected by leprosy (62%) experienced stigma related to touch [35].

We found that 93% of the participants knew that leprosy can be treated. Of these people, 97% knew that leprosy can be treated by medication. This is higher than in other studies in India, which reported 29 to 90% of correct knowledge about curability and treatment of leprosy [12,18,20–22,24,28,29].

The levels of stigma and desired social distance towards persons affected by leprosy found in the present study were high. We found that more knowledge about leprosy was associated with lower levels of stigma, but that ‘knowledge’ explained only a small proportion of the variation in stigma levels. Other studies in India also report high levels of negative attitudes and stigma [12,19,22,24,29,36–38]. A study in a tertiary care hospital in Delhi also found that greater knowledge of leprosy is a positive predictor of attitude [19]. This suggests that improving knowledge about leprosy may also improve attitudes.

In our study participants who knew a person affected by leprosy perceived higher levels of community stigma while being a close contact to someone affected was associated with lower levels of stigma. This is a surprising finding, as one would expect that close contacts of someone affected by leprosy are people who know someone affected by leprosy. Next to higher perceived levels of community stigma, participants who knew a person affected also perceived lower levels of desired social distance compared to participants who did not know a person affected. We believe that perceived stigma in the community may increase when people know someone affected and see the difficulties they experience. At the same time knowing someone affected could potentially improve personal attitudes towards the person, thus reduce the desired social distance. Furthermore, considering the high levels of incorrect knowledge of our participants regarding leprosy, we believe that the higher levels of stigma among people who know someone affected by leprosy in our study could also be due to their misconceptions regarding leprosy.

Finally, the findings of this study have to be considered in the context of its limitations. A limitation of the study is that it was a cross-sectional study and could therefore not establish definite cause-and-effect relationships; we were only able to form hypotheses about cause and effect relationships. Furthermore, although interesting and potentially relevant, it was not possible to take into account factors such as disability status and leprosy classification as we needed to focus on background characteristics which are most relevant in the context of designing large group interventions, for instance gender and level of education. In addition, the SDS used in this study had not yet been formally validated in Hindi. However, the SDS has been translated, piloted, extensively used and had its psychometric properties assessed in a parallel baseline study (Ballering, in preparation). We therefore considered the SDS a valid measure of social distance. Finally, this study only assessed leprosy-related stigma in community members, contact and health care workers and did not assess self-stigma and enacted stigma. A strength of the present study is the mixed-method approach that allowed for triangulation of the data.

The present study has important implications for the development of leprosy control strategies. This study identified a lack of knowledge about leprosy and high levels of stigma towards persons affected by leprosy in Fatehpur district, Uttar Pradesh, India. The insights we obtained in this study in knowledge gaps, beliefs and misconceptions will be used to design effective community education methods to raise awareness, positively influence the perception of and improve knowledge regarding leprosy and reduce the stigma against leprosy. We recommend a combination of written health education materials in combination with behavioural change interventions, as written materials used in isolation are often not adequate to change behaviour [39,40] and because a high level knowledge of leprosy alone does not necessarily lead to more positive attitudes towards persons affected [41]. In addition, we identified a need for increased awareness of and information about leprosy among health care workers. Even though health care workers had higher levels of knowledge than the other participants in this study, they were still not adequate. We recommend standard training on leprosy for all health care workers and regular refresher courses in areas that are endemic for leprosy. We expect that these education methods will improve strategies for early case detection in leprosy thus improving the effectiveness of the National Leprosy Eradication Programme

Conclusion

This study revealed poor knowledge regarding leprosy among index patients, close contacts, community members and health care workers in Fatehpur district, northern India. Knowledge on mode of transmission, cause and symptoms of leprosy was especially poor. In addition, we found high levels of stigma towards persons affected by leprosy.

Several factors were associated with higher levels of negative attitudes towards persons affected by leprosy, including knowing a person affected by leprosy, being a community member, being a woman, being illiterate and specific conceptions regarding cause, transmission and contagiousness of the disease. We found that better knowledge of leprosy was associated with lower levels of social distance towards persons affected by leprosy.

In order to improve knowledge, reduce misconceptions and positively influence the perception of leprosy, community education is needed. Special emphasis needs to be placed on education regarding mode of transmission, cause and symptoms of leprosy. A multidisciplinary approach that takes cultural beliefs, knowledge gaps and fears into consideration is recommended.

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References

1. Lastória JC, Abreu MAMM de. Leprosy: review of the epidemiological, clinical, and etiopathogenic aspects-part 1. An Bras Dermatol 2014;89:205–18.
2. Suzuki K, Akama T, Kawashima A, Yoshihara A, Yotsu RR, Ishii N. Current status of leprosy: epidemiology, basic science and clinical perspectives. J Dermatol 2012;39:121–9.
3. Rafferty J. Curing the stigma of leprosy. Lepr Rev 2005;76:119–26.
4. Singh GP. Psychosocial aspects of Hansen's disease (leprosy). Indian Dermatol Online J 2012;3:166.
5. van Brakel WH. Measuring leprosy stigma-a preliminary review of the leprosy literature1. Int J Lepr Other Mycobact Dis 2003;71:190.
6. Bratschi MW, Steinmann P, Wickenden A, Gillis TP. Current knowledge on Mycobacterium leprae transmission: a systematic literature review. Lepr Rev 2015;86:142–55.
7. WHO. Global leprosy update, 2016: accelerating reduction of disease burden. vol. 92. 2017.
8. World Health Organization. WHO Expert Committee on leprosy: eighth report. Geneva: World Health Organization; 2012.
9. Duthie MS, Truman RW, Goto W, O'Donnell J, Hay MN, Spencer JS, et al. Insight toward early diagnosis of leprosy through analysis of the developing antibody responses of Mycobacterium leprae-infected armadillos. Clin Vaccine Immunol 2011;18:254–9.
10. Da Silva Souza C, Bacha JT. Delayed diagnosis of leprosy and the potential role of educational activities in Brazil. Lepr Rev 2003;74:249–58.
11. Opala J, Boillot F. Leprosy among the Limba: illness and healing in the context of world view. Soc Sci Med 1996;42:3–19.
12. John AS, Rao OS. Awareness and attitudes towards leprosy in urban slums of Kolkata, India. Indian J Lepr 2009;81:135–40.
13. Bekri W, Gebre S, Mengiste A, Saunderson PR, Zewge S. Delay in presentation and start of treatment in leprosy patients: a case-control study of disabled and non-disabled patients in three different settings in Ethiopia. Int J Lepr Other Mycobact Dis 1998;66:1.
14. Nicholls PG, Chhina N, Bro AK, Barkataki P, Kumar R, Withington SG, et al. Factors contributing to delay in diagnosis and start of treatment of leprosy: Analysis of help-seeking narratives in northern Bangladesh and in West Bengal, India. Lepr Rev 2005;76:35–47.
15. Reddy NB, Satpathy SK, Krishnan SA, Srinivasan T. Social aspects of leprosy: a case study in Zaria, northern Nigeria. Lepr Rev 1985;56:23.
16. Robertson LM, Nicholls PG, Butlin R. Delay in presentation and start of treatment in leprosy: experience in an out-patient clinic in Nepal. Lepr Rev 2000;71:511–6.
17. Van de Weg N, Post EB, Lucassen R, De Jong J, Van Den Broek J. Explanatory models and help-seeking behaviour of leprosy patients in Adamawa State, Nigeria. Lepr Rev 1998;69:382–9.
18. Saha G, Mandal NK, Dutta RN. Current Perceptions and Practices (KAP) about Leprosy among Leprosy Patients: A Comparative Study between High Prevalent & Low Prevalent Districts of West Bengal. Indian J Lepr 2015;87:1–16.
19. Seshadri D, Khaitan BK, Khanna N, Sagar R. The tangled web: a study of knowledge and attitude towards leprosy from a tertiary care hospital in India. Indian J Lepr 2014:27–41.

20. Stephen T, Selvaraj I, Parameswari PJ. Assessment of knowledge, attitude and practice about leprosy among patients and their families in a rural community in Tamil Nadu. Indian J Lepr 2014;86:7–14.
21. Grewal I, Negi Y, Kishore J, Adhish S V. Knowledge and attitude about Leprosy in Delhi in post elimination phase. Indian J Lepr 2013;85.
22. Mankar MJ, Joshi SM, Velankar DH, Mhatre RK, Nalgundwar AN. A comparative study of the quality of life, knowledge, attitude and belief about leprosy disease among leprosy patients and community members in Shantivan Leprosy Rehabilitation centre, Nere, Maharashtra, India. J Glob Infect Dis 2011;3:378.
23. Rao P V., Rao SL, Vijayakrishnan B, Chaudhary AB, Peril S, Reddy BP, et al. Knowledge, attitude and practices about leprosy among medical officers of Hyderabad urban district of Andhra Pradesh. Indian J Lepr 2007;79:27–43.
24. Kaur H, Gandhi A. People's perception of leprosy--a study in Delhi. Indian J Lepr 2003;75:37–46.
25. Jain M, Sharma A, Jain V, Virjee K, Singh S. Knowledge and Attitude about Leprosy among Indian Dental Students in Faridabad. J Clin Diagnostic Res JCDR 2016;10:ZC48.
26. Kanodia SK, Dixit AM, Shukla SR, Seth AK, Balothia V, Gupta R. A study on knowledge, beliefs and attitude towards leprosy in students of Jaipur, Rajasthan. Indian J Lepr 2012;84:277–85.
27. Giri PA, Phalke DB, Aarif SM. A study of knowledge, attitude and practices regarding leprosy among undergraduates and interns of a medical college and hospital from rural India. Indian J Lepr 2011;83:75–80.
28. Danturty I, Manchala S, Jairaj S. Community based study of knowledge, attitude and practices towards leprosy in an urban slum in Secunderabad, Telangana. JMSCR 2016;49:13527–31.
29. Barkataki P, Kumar S, Rao PS. Knowledge of and attitudes to leprosy among patients and community members: a comparative study in Uttar Pradesh, India. Lepr Rev 2006;77:62–8.
30. Kar S, Ahmad S, Pal R. Current knowledge attitudes, and practices of healthcare providers about leprosy in Assam, India. J Glob Infect Dis 2010;2:212.
31. National Leprosy Eradication Programme. NLEP Annual Report 2016-2017. 2017.
32. WHO. Weekly Epidemiological Record. vol. 93. 2018.
33. Rensen C, Bandyopadhyay S, Gopal PK, Van Brakel WH. Measuring leprosy-related stigma–a pilot study to validate a toolkit of instruments. Disabil Rehabil 2011;33:711–9.
34. Peters RMH, Van Brakel WH, Zweekhorst MBM, Damayanti R, Bunders JFG. The cultural validation of two scales to assess social stigma in leprosy. PLoS Negl Trop Dis 2014;8:e3274.
35. Kushwah SS, Govila AK, Upadhyay S, Kushwah J. A study of social stigma among leprosy patients attending leprosy clinic in Gwalior. Lepr India 1981;53:221–5.
36. Jacob JT, Franco-Paredes C. The stigmatization of leprosy in India and its impact on future approaches to elimination and control. PLoS Negl Trop Dis 2008;2:e113.
37. Barrett R. Self-mortification and the stigma of leprosy in northern India. Med Anthropol Q 2005;19:216–30.
38. Rao P V., Raju MS, Barkataki A, Nanda NK, Kumar S. Extent and Correlates of Leprosy Stigma in Rural India. Indian J Lepr 2008;80:167.
39. Seligman HK, Wallace AS, DeWalt DA, Schillinger D, Arnold CL, Shilliday BB, et al. Facilitating behavior change with low-literacy patient education materials. Am J Health Behav 2007;31:S69–78.

40. Noar SM, Benac CN, Harris MS. Does tailoring matter? Meta-analytic review of tailored print health behavior change interventions. Psychol Bull 2007;133:673.
41. Wong ML. Guest editorial: designing programmes to address stigma in leprosy: issues and challenges. Asia Pacific Disabil Rehabil J 2004;15:3–12.

CHAPTER 3

Leprosy perceptions and knowledge in endemic districts in India and Indonesia: differences and commonalities

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Abstract

Introduction

Understanding how knowledge, attitudes and practices regarding leprosy differ in endemic countries can help us develop targeted educational and behavioural change interventions. This study aimed to examine the differences and commonalities in and determinants of knowledge, attitudes, practices and fears regarding leprosy in endemic districts in India and Indonesia.

Methods

A cross-sectional mixed-methods design was used. Persons affected by leprosy, their close contacts, community members and health workers were included. Through interview-administered questionnaires we assessed knowledge, attitudes, practices and fears with the KAP measure, EMIC-CSS and SDS. In addition, semi-structured interviews and focus group discussions were conducted. The quantitative data were analysed using stepwise multivariate regression. Determinants of knowledge and stigma that were examined included age, gender, participant type, education, occupation, knowing someone affected by leprosy and district. The qualitative data were analysed using open, inductive coding and content analysis.

Results

We administered questionnaires to 2344 participants (46% from India, 54% from Indonesia) as an interview. In addition, 110 participants were interviewed in-depth and 60 participants were included in focus group discussions. Knowledge levels were low in both countries: 88% of the participants in India and 90% of the participants in Indonesia had inadequate knowledge of leprosy. In both countries, cause, mode of transmission, early symptoms and contagiousness of leprosy was least known, and treatment and treatability of leprosy was best known. In both countries, health workers had the highest leprosy knowledge levels and community members the highest stigma levels (a mean score of up to 17.4 on the EMIC-CSS and 9.1 on the SDS). Data from the interviews indicated that people were afraid of being infected by leprosy. Local beliefs and misconceptions differed, for instance that leprosy is in the family for seven generations (Indonesia) or that leprosy is a result of karma (India). The determinants of leprosy knowledge and stigma explained 10-29% of the variability in level of knowledge and 3-10% of the variability in level of stigma.

Conclusion

Our findings show the importance of investigating the perceptions regarding leprosy prior to educational interventions in communities: even though knowledge levels were similar, local beliefs and misconceptions differed per setting. The potential determinants we included in our study explained very little of the variability in level of knowledge and stigma and should be explored further. Detailed knowledge of local knowledge gaps, beliefs and fears can help tailor health education to local circumstances.

Introduction

Many health conditions are associated with social stigma, including epilepsy, mental illness, disability and infectious diseases such as HIV/AIDS, tuberculosis, leprosy, lymphatic filariasis and Buruli ulcer. Stigma occurs when "elements of labelling, stereotyping, separation, status loss, and discrimination occur together in a power situation that allows them" [1]. Stigma is characterized by social exclusion or separation, rejection, blame and loss of status of an individual or group [1,2]. For many stigmatized individuals, the psychosocial consequences of their health condition are harder to bear than the physical consequences [3,4]. Stigma is associated with poor psychosocial health outcomes such as reduced quality of life, low self-esteem, depression and social exclusion [3,5]. This is also the case for persons affected by leprosy who experience stigma [6].

Leprosy is an infectious disease that primarily affects and damages the peripheral nerves and skin, which can result in disabilities [7,8]. Leprosy has had a very negative image for hundreds of years and is known for being a very stigmatized condition [9]. Leprosy and its stigma may affect different areas of a person's life such as mobility, social relationships, marriage, employment and social participation [4]. Stigma and discrimination can lead to stress, anxiety, depression, suicide, isolation and problems in interpersonal relationships of persons affected [4]. Stigma in leprosy can also worsen already existing social inequalities due to age, gender and social class or status [10].

Stigma, as well as a lack of knowledge about leprosy, are obstacles to case finding and adherence to treatment [11,12] and therefore reduce the effectiveness of leprosy care and control activities [4,13]. In an attempt to hide their disease and prevent discrimination, stigmatized individuals often delay seeking treatment until they develop permanent, visible disabilities [8]. When people delay seeking treatment, transmission of the disease is prolonged, which hinders the treatment and prevention of the disease. To improve strategies for early case detection it is essential to improve the knowledge of leprosy and reduce stigma.

There are many factors that contribute to the stigma of leprosy, including fear of transmission and contagion, the visible manifestations such as deformity and disability in persons affected and religious and cultural beliefs regarding causes and treatment of leprosy [12,13]. Knowledge about leprosy plays a crucial role in stigma [14–18]. Local (mis)beliefs, such as the beliefs that all leprosy patients end up with disabilities, that leprosy is not curable or results in death or that imply that the person affected has done wrong and brought the disease upon himself all contribute to stigma [13,14,18–23]. Research showed that personal characteristics such as gender [15,16,22,24], occupation [16,22,24–26], years of education [15–17,22,25], age [15,25,27,28] and living area [15,24,27,29,30] are associated with community stigma against persons affected by leprosy.

Although knowledge, attitudes and practices regarding leprosy have been studied extensively and different determinants of knowledge and community stigma have been identified, we lack consensus about determinants of stigma and insight in how we can change negative perceptions and stigmatising local beliefs about leprosy. Insight in the dynamics, differences and commonalities in knowledge, attitudes and practices between leprosy endemic countries can help us to better target interventions to improve the knowledge and perception of leprosy, and thus reduce stigma. This study aims to examine the differences and commonalities in and determinants of knowledge, attitudes, practices and fears regarding leprosy in leprosy endemic districts in India and Indonesia.

Methods

Study design

The study used a cross-sectional research design with a mixed methods approach. Interviewer-administered questionnaires included demographic characteristics and knowledge and attitudes of people towards (persons affected by) leprosy. In-depth information was obtained using semi-structured interviews and focus group discussions.

Study site

The study was conducted in Fatehpur and Chandauli district, Uttar Pradesh, India and in Pamekasan and Pasuruan regencies (including Pasuruan city) in East Java, Indonesia. India and Indonesia are among the three most endemic countries for leprosy. India and Indonesia account for 92% of the South-East Asian region's case load and for almost 66% of the global disease burden of leprosy [31]. In March 2017, the prevalence of leprosy was 0.89 per 10.000 population in Fatehpur and 0.66 per 10,000 population in Chandauli district [32]. The prevalence of leprosy was 2.27 per 10,000 population in Pamekasan regency, 1.07 per 10,000 population in Pasuruan regency and 0.55 per 10,000 population in Pasuruan city in December 2018 respectively (data obtained from the local Provincial Health Offices).

Study population and sample

Four groups of people were included in the study: (1) persons affected by leprosy; (2) close contacts of persons affected; (3) community members; and (4) health care workers. We aimed to include a random sample of at least 100 persons of each target group per country for the interview-administered questionnaires. In addition, in each country we aimed to have semi-structured interviews with six persons from each participant group and one focus group discussion per participant group. These participants were to be a subset of those in the quantitative sample. More information about the sample size calculation can be found elsewhere [18].

Eligibility criteria

Participants needed to be inhabitants of one of the districts included in the study. Only persons affected diagnosed with leprosy within the last five years were included. Closest contacts had to have had intensive contact with the person affected for at least 20 hours per week for at least three months in the 12 months before the person affected was diagnosed. Close contacts included household contacts, family members, neighbours and other social contacts. Only those community members living in the same village or neighbourhood as the person affected by leprosy were asked to participate. Health care workers included professionals and volunteers, and persons with and without specific responsibilities for leprosy treatment services. Persons below the age of 16 and persons unwilling or unable to give informed consent were excluded. Close contacts, community members and health care workers were also excluded if they were or had ever been diagnosed with leprosy. Participants who were listed as close contact could not participate as community member also.

Sampling methods

Participants for the interview-administered questionnaires were selected as follows:

1. The persons affected were selected by stratified systematic sampling with a random start from a list of leprosy patients registered at the primary health care centre.
2. In India, close contacts were selected by convenience sampling. We realized in hindsight that convenience sampling was not the best approach and therefore applied random sampling of close contacts when initiating the data collection in Indonesia. In Indonesia, all persons affected included in the study were each asked to name their 20 closest contacts whose names were written down on pieces of paper. These pieces were put in a cup, and one

was randomly drawn. If this person was not available or did not give consent, another name was randomly drawn. In both countries one close contact per person affected was included.

3. The community members were selected by convenience sampling from among those living in the same village or neighbourhood as the person affected by leprosy. We tried to select the community members as randomly as possible by selecting one or two community members per person affected from within a radius of 500 meters of the house of the person affected.
4. Health care workers were selected based on convenience sampling from among those present and available at the primary health care centres in the district. Half of the health care workers included in this study had received training about leprosy and had specific responsibilities for leprosy treatment services.

The participants for the qualitative interviews were a subset of those in the quantitative sample. Participants in the interviews were selected using purposive sampling to ensure adequate representation of age, sex and villages.

Data collection

Three measures were used in this study: a knowledge, attitudes and practices (KAP) measure, the Explanatory Model Interview Catalogue Community Stigma Scale (EMIC-CSS) and the Social Distance Scale (SDS). Demographic information was also obtained from all participants. The EMIC-CSS and SDS were not administered to persons affected, since they assess community stigma.

The up-to 17-item (depending on the participant type) KAP measure was used to assess the knowledge, attitudes and practices of persons affected, close contacts, community members and health care workers regarding leprosy. Based on seven items on the KAP measure considering knowledge of leprosy a total knowledge score is calculated. A total score of eight could be obtained on the KAP adding up the correct answers even if incorrect answers were present. We defined ‘poor knowledge’ as a score of two or less out of eight on the KAP measure (≤25% correct), ‘moderate knowledge’ as a score between two and six (25-75% correct) and ‘adequate knowledge’ as a score of six or more (≥75% correct). The KAP measure has been used in several leprosy studies in Nepal, India, Indonesia and Brazil between 2012 and 2017 [18,33–35].

The 15-item EMIC-CSS was used to measure attitudes and behaviour towards persons affected by leprosy. The total score ranges from zero (no negative attitudes) to 30 (most negative attitudes). The EMIC-CSS has been validated among community members of persons affected by leprosy in India and Indonesia [36,37].

The 7-item SDS was used to assess the social distance the interviewee wants to keep towards persons affected by leprosy as a proxy for their attitudes. The SDS total score ranges from zero (no negative attitudes) to 21 (most negative attitudes). The SDS has been validated among community members of persons affected by leprosy in Indonesia [37]. The SDS has been translated to Hindi using forward and backward translation and partially validated (item interpretability, floor and ceiling effects and internal consistency) showing adequate validity, among community members of persons affected by leprosy in Uttar Pradesh, India [33].

Cross-sectional data on attitudes and perceptions of the participants towards (persons affected by) leprosy were also obtained using semi-structured in-depth interviews and focus group discussions. All participants were interviewed by a trained local interviewer at or near their home. The interview guide was pilot tested in each district before use, resulting in minor revisions to the guide. Participants of the pilot interviews were not included in the final sample. The in-depth interviews and focus group discussions were audio recorded, transcribed verbatim and translated into English. Data were collected in different intervals between March 2017 and December 2018.

Data analysis

Quantitative data analyses were performed in SPSS version 24. Simple descriptive methods were used to generate a demographic profile of the study sample. Stepwise multivariate regression with backward elimination was done to investigate the contribution of potential determinants (age, gender, participant type, education, occupation, knowing someone affected by leprosy and district) to the outcomes of interest (knowledge, stigma and social distance). Dependent variables that originally consisted of multiple categories were recoded into binary dummy variables. Independent variables were considered for entry into the multivariable logistic regression model if they had a *p*-value of ≤ 0.2 obtained in univariate analysis. Variables with *p*-values of ≥ 0.05 were eliminated one-by-one from the multivariate model until all variables that remained in the model were statistically significant ($p<0.05$). Bootstrapped stepwise multivariate regression with backward elimination, to correct for non-normality, was done for dependent variables that were distributed non-normally.

For each dependent variable four models were made: one using the whole database (four districts, all participant types), one for health workers only, one for persons affected only and one for the general population (close contacts and community members). Separate models for health workers were made because we considered them a more heterogenous group and a different group than the general population in terms of having completed (higher) education, knowledge (training) about leprosy and occupation. Separate models for persons affected were made because of their personal experience with leprosy and because they likely received a briefing or information about their condition. We hypothesized that participant type correlated with dependent and independent variables in the model. To control for confounding of participant type, and to ensure that the effects of participant type would be removed from the final results, we decided to always keep all participant types in the model, when analysing the whole dataset.

The recordings of interviews were transcribed, translated to English and analysed using open, inductive coding and content analysis. Similar phrases with recurring themes were clustered together in tables, to identify connections. Qualitative data analyses were performed in Nvivo version 12, Microsoft Word and Microsoft Excel. All records were anonymised before analysis.

Ethical considerations

Ethical approval for this study was obtained as part of a larger research project: the Post-Exposure Prophylaxis project (PEP++ project). Ethical approval was obtained from the Institutional Ethics Committee in India and from Airlangga University in Indonesia. All participants were fully informed about the nature, objectives and procedures of the study, their rights and of confidentiality of the data prior to data collection. Written consent for participation in the study was obtained from each participant. For minors (participants below 18 years of age), verbal informed consent was obtained from the minor and written informed consent was obtained from one of the minor’s legal representatives, e.g. a parent or guardian.

Results

Demographic information

A total of 2344 participants were included. A little over half (n=1277, 54%) of the participants were from Indonesia. The average age of the participants was 40.5 years. Approximately half of the participants were female (41% in India (n=433), 51% in Indonesia (n=654)). Four groups of people were included in the study: 19% of the participants were affected by leprosy (n=438), 19% were close contacts of persons affected (n=449), 54% were community members (n=1256) and 9% health care workers (n=201). An overview of all participant characteristics can be found in Table 1.

Table 1. Socio-demographic characteristics and leprosy-related stigma and knowledge levels.

	India (*n* = 1067)[a]	**Indonesia (*n* = 1277)**[b]	**Total (*n* = 2344)**
Average age (range; SD)	40.4 (16-90; 15.6)	40.5 (16-95; 13.0)	40.5 (16-95; 14.2)
Female, *n* (%)	433 (40.6)	654 (51.2)	1087 (46.4)
Participant type, *n* (%)			
Person affected by leprosy	200 (18.7)	238 (18.5)	438 (18.7)
Close contact of person affected	211 (19.8)	238 (18.5)	449 (19.2)
Community member	556 (52.1)	700 (55.0)	1256 (53.6)
Health care worker	100 (9.4)	101 (8.0)	201 (8.6)
No education, *n* (%)	279 (26.1)	264 (20.7)	543 (23.2)
Religion, *n* (%)			
Islam	89 (8.3)	567 (45.1)	1365 (28.4)
Hinduism	970 (90.9)	0 (0.0)	970 (41.4)
Unknown	8 (0.7)	700 (54.8)	708 (30.2)
Occupation, *n* (%)			
Paid work	349 (32.7)	250 (19.6)	599 (25.6)
Self-employed	347 (32.5)	615 (48.2)	962 (41.0)
Retired or unemployed	82 (7.7)	192 (15.0)	274 (11.7)
Other (such as non-paid work, student)	289 (27.1)	220 (17.2)	509 (21.7)
Questionnaire scores, mean			
KAP measure (range 0-8)[c]	3.9	3.2	3.5
EMIC-CSS (range 0-30)[d]	15.9	15.5	15.7
SDS (range 0-21)[d]	6.6	8.6	7.7

[a] 446 participants from Fatehpur and 621 participants from Chandauli district.

[b] 639 participants from Pamekasan and 638 participants from Pasuruan regency.

[c] In the presence of incorrect answers. Participants could give multiple answers to some of the KAP questions.

[d] The EMIC-CSS and SDS were administered to close contacts, community members and health workers.

In addition, a total of 110 participants (52 in India, 58 in Indonesia) were interviewed in-depth (average age 39 years, 45% female) and in India 60 participants were included in seven focus group discussions (average age 40 years, gender not recorded). These 170 participants were a subset of those who had completed the questionnaires. An overview of all participant in the interviews can be found as supporting information file (S1 Table).

Differences and commonalities in knowledge, attitudes and practices regarding leprosy

Differences and commonalities in leprosy knowledge and beliefs

The questions related to knowledge that were answered correctly most and least frequently in both countries were the same. An overview of the number of correct responses given per participant group per country, per knowledge question of the KAP measure can be found as supporting information files (S2 Table and S1 Text). Mode of transmission, cause, early symptoms and whether leprosy is contagious after treatment or not was least known among all participant groups. In both countries treatment and treatability of leprosy was best known (>74% correct). In addition, in both countries health workers had significantly better knowledge than the other participants (independent samples t-test, $p<0.001$). When comparing overall scores between participant groups, merging the data from India and Indonesia, we found that persons affected had significantly higher knowledge scores than close contacts and community members, and that health workers had significantly higher knowledge scores than all participant groups (independent samples t-test, $p<0.001$).

Persons affected, contacts and community members from India had significantly higher mean knowledge scores (independent samples t-test, $p<0.05$) than the same participant groups in Indonesia. This considered e.g. questions about treatment and treatability (91-93% correct in India versus 75-78% in Indonesia), prevention of disabilities (69% correct in India versus 50% Indonesia) and contagiousness after treatment (43% correct in India versus 23% in Indonesia). Participants from Indonesia had slightly better knowledge about mode of transmission (12% correct in Indonesia versus 6% in India) and cause of leprosy (17% correct in Indonesia versus 13% in India). While health workers from Indonesia had significantly better ($p=0.001$) knowledge than health workers from India on almost all aspects, their knowledge about whether leprosy is contagious or not after treatment was low (9% correct in Indonesia versus 59% in India).

We found that 12% of the participants in India and 10% of the participants in Indonesia had adequate knowledge of leprosy. In addition, 76% of the participants in India and 58% of the participants in Indonesia had moderate knowledge and 14% of the participants in India and 32% of the participants in Indonesia had poor knowledge of leprosy.

Both on the KAP measure and during the in-depth interviews and focus group discussions, participants often used multiple explanations and held different beliefs per knowledge topic. This is illustrated by the following quote:

> *"…[Being affected by leprosy] may be because of wearing wet clothes or some kind of allergy in my blood. It may also be that it had happened to some friend and I got infected while playing because it can spread through the touch…"*
>
> – Person affected, male, India, in-depth interview

In addition, in both countries participants believed that certain types of food or drinks, for example seafood or unhealthy food, could cause leprosy. While some participants in both countries believed that an unclean environment could cause leprosy, this belief was more prominent in India. At the same time, some participants in both countries believed leprosy was hereditary, but this was a much more prominent belief in Indonesia. Some participants believed in supernatural causes of leprosy, especially in Chandauli in India (karma and evil spirits) and Pasuruan in Indonesia (black magic, God's will).

There were several local beliefs around the cause and mode of transmission of leprosy in Indonesia. Participants believed that leprosy is caused by 'impure blood' caused by '*karuwat* sin', which creates 'bad flesh'. Bad flesh is also one of the names used for leprosy in Indonesia. *Karuwat* is when a man and a woman have sexual intercourse while the woman has her period and conceive a child. It is believed that the child will be affected by leprosy. One participant explained:

> *"…To my knowledge, women in their menstrual cycle are not permitted to have sexual relations in any religion. So the majority of the community here believe that to be the cause. Bacteria is the cause, and it is believed that [babies] born carrying contaminated bacteria end up having leprosy…"*
>
> - Close contact, male, Indonesia, in-depth interview

In addition, some participants in Indonesia believed that leprosy is in the family for seven generations. Another less common belief in Indonesia was that one can get leprosy from stepping on the grave of a person affected by leprosy. One participant explained:

> *"…When [a friend and I] were in junior high school [we] were playing on the graveyard, cemetery, and [we] stepped on hot soil. Here hot means not hot literally (…) it is like contagious (…) the graveyard is from people with leprosy, so they get infected [by stepping on that land] (…) The one that stepped on it is the one that untreatable. But the other friend got it from hereditary…"*
>
> – Community member, male, Indonesia, in-depth interview

In addition, some participants in Indonesia believed there were two types of leprosy: 'lepra', the skin condition and '*kusta*', a more severe and feared form with visible impairments. Many participants in India thought leprosy transmits by touch and linked this to 'untouchability' and sometimes to being religiously unclean. One participant said:

> *"…Some people maintain distance, they don't eat or drink together because of untouchability…"*
>
> - Community member, female, India, in-depth interview

Differences and commonalities in leprosy-related stigma

The mean EMIC-CSS (stigma) score was 15.9 in India and 15.5 in Indonesia. In both countries, community members had the highest mean EMIC-CSS score (17.4 India, 17.0 Indonesia), followed by health workers (15.0 India, 13.7 Indonesia) and close contacts (12.4 India, 11.8 Indonesia). Differences between the countries in mean EMIC-CSS scores per participant group were not significant (independent samples t-test, $p>0.05$, Table 2). However, when comparing overall scores between participant groups, merging the data from India and Indonesia, we found that close contacts had significantly lower and community members significantly higher mean EMIC-CSS scores (independent samples t-test, $p<0.001$).

Table 2. Differences in total scores on the KAP (range 0-8), EMIC-CSS (range 0-30) and SDS (range 0-21) per participant group, per country.

Instrument and participant type		Mean India (*n*=1067)	Mean Indonesia (*n*=1277)	p-value[b]
KAP[a]	Persons affected by leprosy	3.9	3.4	.000
	Close contacts	3.5	3.0	.001
	Community members	3.8	2.8	.000
	Close contacts and community members[c]	3.7	2.9	.000
	Health workers	5.6	6.2	.001
	All groups	3.9	3.2	.000
EMIC-CSS	Close contacts	12.4	11.8	.356
	Community members	17.4	17.0	.268
	Close contacts and community members	16.0	15.6	.289
	Health workers	15.0	13.7	.217
	All groups	15.9	15.5	.188
SDS	Close contacts	6.7	8.6	.000
	Community members	7.2	9.1	.000
	Close contacts and community members	7.0	9.0	.000
	Health workers	3.4	5.3	.002
	All groups	6.6	8.6	.000

[a] In the presence of incorrect answers. Participants could give multiple answers to some of the KAP questions.

[b] p-value of the difference in total questionnaire score between India and Indonesia in the corresponding column. Tested using an independent samples t-test (significance, 2-tailed, equal variances were assumed if Levene's test for equality of variances had a p-value of >0.05).

[c] This is a merged group that combines the data of close contacts and community members.

The EMIC-CSS questions reflecting negative attitudes that were frequently endorsed (indicating most stigma) were similar between the two countries. These questions relate to marriage, avoiding persons affected, shame and disclosure. An overview of all responses to the EMIC-CSS can be found as supporting information file (S1 Fig).

The overall mean SDS score was 6.6 in India and 8.6 in Indonesia ($p<0.05$), indicating more stigma in Indonesia. In both countries community members had the highest mean SDS scores (7.2 India, 9.1 Indonesia), followed by close contacts (6.7 India, 8.6 Indonesia) and health workers (3.4 India, 5.3 Indonesia), see Table 2. Health workers had significantly lower SDS scores than close contacts and community members (independent samples t-test, $p<0.001$). In both countries, the questions indicating most stigma were the same and relate to having a person affected by leprosy as caretaker of one's children and to having one's children marry a person affected by leprosy (>45% negative responses in both countries). An overview of all responses to the SDS can be found as supporting information file (S1 Fig).

In the in-depth interviews and focus group discussions, many participants indicated that community members have a negative attitude towards persons affected by leprosy and avoid or exclude persons affected by leprosy. The main explanation in both countries was fear getting infected by the disease. One participant explained:

> *"…I feel sorry for them. One, because they are alienated from their community. Two, because rarely ever would anyone talk to them or involve them or allow them to raise their own children. But what to do? As a community, if someone has leprosy, we fear for our own health, fear of infection. It is horrifying…"*
> - Close contact, female, Indonesia, in-depth interview

Especially in India, some participants had a very negative perception of persons affected. One participant said:

> *"…Most leprosy patients are dirty and poor. So, if they don't treat their leprosy it will go on, go on, go on. It gets worse. If it gets worse, working is not possible. No job for them. No one would want to work with leprosy patients, no one would like to have leprosy patients employed, no one would buy from leprosy patients. So, if leprosy is visible, they can't do work."*
> – Community member, female, India, in-depth interview

Another participant said:

> *"…People do not eat with leprosy patient nor touch them. And his living place is also separated. They refuse to visit some places and says don't touch me, or else I will also have disease. Because of this he becomes sad and suffers from inferiority complex. Sometimes he also tried to commit suicide…"*
> - Community member, gender unknown, India, focus group discussion

On the other hand, some participants said that there was no discrimination in the community. In addition, most participants in the in-depth interviews said that in general health workers have a good attitude towards persons affected by leprosy.

In Indonesia some participants stressed that when leprosy is not visible, the community will treat these individuals normally. It became clear in the interviews that in Indonesia the terminology around leprosy is sensitive. Some health workers indicated it's better not to tell someone they have leprosy and had therefore adopted different names:

> *"...In the Madurese context we should not mention the word leprosy; instead we refer to it as skin condition that is treatable. This is so that the patients do not evade treatment. If we do [say that it is leprosy] then patients definitely will not come back..."*
>
> – Health worker, male, Indonesia, in-depth interview

Some participants indicated that they used different names for '*kusta' (*leprosy), such as '*daging jubek*' or '*budukan'* (bad flesh) or *'gatal gatal'* (itchy). These terms are only used to indicate leprosy. In India participants indicated that they were reluctant to touch persons affected by leprosy.

Determinants of leprosy knowledge and leprosy-related stigma

Determinants of knowledge of leprosy

Multivariate analysis showed that being illiterate, not having had any (formal) education, only completing primary education, not knowing anyone affected by leprosy and living in Pamekasan or Pasuruan district (Indonesia) were all associated with lower levels of knowledge of leprosy (Table 3). Multivariate regression models per participant group could explain 11-22% of the variability of knowledge of leprosy, see Table 3. The determinants of knowledge of leprosy per participant group related to age, gender, knowing someone affected by leprosy, education and area of residence.

Table 3. Correlations between level of knowledge (KAP score, in the presence of incorrect answers) about leprosy, community stigma (EMIC-CSS), social distance (SDS) and the other variables in the dataset per participant group. Full models can be found as supporting information file (S1 Text).

	Determinants of lower levels of knowledge (KAP measure)	**R-squared**	**Determinants of higher levels of stigma (EMIC-CSS)**	**R-squared**	**Determinants of higher levels of social distance (SDS)**	**R-squared**
Whole dataset	• Illiterate, no (formal) education or only completed primary education • Does not know someone affected by leprosy • From Pamekasan or Pasuruan district	0.286	• From Chandauli, Fatehpur or Pamekasan district	0.096	• Female gender • Not having completed higher education • Lower knowledge about leprosy • From Fatehpur, Pamekasan or Pasuruan district	0.103
Persons affected	• Older age • Female gender • Not completed any (formal) education • From Pamekasan district	0.120	-	-	-	-
Contacts and community	• Illiterate, no (formal) education or only completed primary education • Does not know someone affected by leprosy • From Pasuruan or Pamekasan district	0.107	• From Chandauli, Fatehpur or Pamekasan district	0.103	• Female gender • Not having completed higher education • Lower knowledge about leprosy • From Fatehpur, Pamekasan or Pasuruan district	0.069
Health workers	• Female gender • Does not know someone affected by leprosy • From Pamekasan, Fatehpur or Chandauli district	0.237	• Younger age	0.032	• Older age	0.033

Determinants of leprosy-related stigma

Multivariate analysis showed that living in Chandauli, Fatehpur or Pamekasan district was associated with higher levels of stigma towards persons affected by leprosy (Table 3). This model explained 10% of the variability of stigma. Models per participant group can be found in Table 3. The models per participant group showed that age was the only determinant of leprosy community stigma for health workers, and Chandauli, Fatehpur or Pamekasan district the only determinants of stigma in contacts and community members.

Female gender, not having completed higher education, low knowledge about leprosy, and living in Chandauli, Fatehpur or Pasuruan district were associated with higher levels of social distance towards persons affected by leprosy. This model explained 10% of the variability of social distance. Multivariate regression models per participant group can be found in Table 3. These models explained 3-7% of the variability of social distance towards persons affected by leprosy.

Discussion

We found both in India and Indonesia that knowledge about leprosy was poor, while community stigma towards (persons affected by) leprosy was high. There were differences in levels of knowledge and stigma between participant groups: knowledge was better among health workers and stigma was higher among community members. The levels of knowledge were similar in both countries, but the explanations given, the 'local beliefs' and 'misconceptions', differed for some topics. Our findings identified three main drivers of stigma: (1) poor knowledge and misconceptions about leprosy, (2) local beliefs, and (3) fear of contagion. We will now discuss these findings in more detail by drivers of stigma and by participant group.

Poor knowledge and misconceptions

Lower levels of knowledge of leprosy were associated with higher levels of social distance, a proxy for fear and stigma in the community. Lacking knowledge about leprosy is more often found to be associated with negative attitudes towards persons affected by leprosy [14–18]. Misconceptions such as that leprosy transmits by touch, a prominent belief among participants from India in the present study, increase stigma. These misconceptions are often linked to fear of the disease and fear of transmission [13,20,38,39]. To reduce stigma these misconceptions need to be addressed and challenged and knowledge needs to be increased. This is also crucial to improve strategies for early case detection, since lack of knowledge of leprosy is a major contributing factor to late diagnosis [7].

Local beliefs

Interestingly, even though the questions related to knowledge that were answered correctly most and least frequently were the same in both countries in the present study, the local beliefs, especially considering the cause and mode of transmission, varied by area of residence. This confirms findings from other studies that showed that (socio)cultural beliefs about leprosy can increase stigma [13,14,18–23]. We found several local beliefs that can be addressed, such as the belief that leprosy is in the family for seven generations, that a cause of leprosy is that a woman conceives while having sexual intercourse during her period (Indonesia), that leprosy has a supernatural cause and that persons affected by leprosy are untouchable (India).

Some studies have suggested that these beliefs are influenced by religious beliefs and religious teachings about leprosy [13,19,20,40]. We hypothesize that the local beliefs in the present study have to some extent also been influenced by religion and religious practices. For example in Indonesia, where almost all participants were Muslim, local beliefs regarding the cause of leprosy revolved around stepping on a grave (people are buried in Islam and Christianity but cremated in Hinduism) and sexual intercourse with menstruating women (explicitly mentioned as prohibited in the Quran). In India, where almost all participants were Hindu, local beliefs revolved around karma (a fundamental concept of Hinduism), untouchables/untouchability (the former name for a member of low-caste Hindu groups) and being religiously unclean (untouchables are considered religiously unclean).

Fear of contagion

A third important driver of stigma found in the present study was that people were afraid of getting infected with the disease. This is something found in other studies also [13,20,38,39] and something that should receive specific attention when designing leprosy campaigns.

Persons affected by leprosy

We found that persons affected had significantly better knowledge of leprosy than close contacts and community members. This is similar to findings from a study in India, that found that persons affected by leprosy had higher knowledge scores than their family members [17]. Persons affected likely have better knowledge because of their personal experience with the disease and because they have often received information about their condition from health workers when they were diagnosed. However, knowledge about leprosy was low in the present study. Similar to our findings, several other studies in India reported low or inadequate levels of knowledge of leprosy among persons affected [41–43]. Low levels of leprosy knowledge may contribute to non-compliance to treatment and need to be addressed [44].

Close contacts of persons affected and community members

The present study found that community members had the highest stigma levels of all participant groups. This may be explained by their poor knowledge about leprosy, something that has been associated with higher levels of stigma towards persons affected by leprosy in other studies also [14–17]. The image that community members have of persons affected by leprosy is likely not based on knowledge from personal contact, but on incorrect information and negative beliefs.

The present study reported mean stigma scores (EMIC-CSS) ranging from 11.8 (contacts) to 17.4 (community members), which is above the cut-off score for perceived stigmatisation of 8, as proposed by Sermrittirong and colleagues [45]. This confirms findings in Indonesia, Brazil, Thailand, Nepal, Nigeria and New Zealand (mean or median EMIC-CSS scores ranging from 12 to 18) [14,16,22,25,28,45–48]. .

Desired social distance towards persons affected by leprosy, how close one is willing to be towards an affected person in a given situation, is an indicator of the fear and attitude of the respondent themselves and a proxy for fear and stigma in the community. In Indonesia, social distance was assessed among community members in two studies using the SDS [37,46]. The mean SDS scores of 9.3 and 10.5 found in the SARI Project [37,46] are very similar to the score of 9.1 we found among community members in Indonesia, indicating a similar desire for social distance in the present study. These results indicate the desire of community members to keep a distance from persons affected by leprosy. Interestingly, while close contacts had a much lower mean perceived community stigma (EMIC-CSS) score, their mean social distance (SDS) score was about the same as that of community members. We expect that the difference between stigma and social distance scores of close contacts can be explained by the way the questions are asked. In the EMIC-CSS respondents are asked how ‘others’ feel or behave, while in the SDS respondents are asked how they themselves would feel relating to the person portrayed in a vignette. Thus, the SDS assesses personal attitudes and fears and the EMIC-CSS perceived attitudes and behaviour of others.

Several determinants of stigma have been identified in other studies, including knowledge of leprosy [14–18], (cultural) beliefs [13,14,18–23], female gender [15,16,22,24], occupation [16,22,24–26], fewer years of education [15–17,22,25], older age [15,25,27,28], knowing a person affected [28], religious beliefs [13,19,20,40] and living area [15,24,27,29,30]. We included almost all of these determinants, except for living area and religion, and found that together they explained very little of the variability in level of stigma (7% on the EMIC-CSS and 10% on the SDS).

We expect that 'local beliefs' and local explanations play an important role in knowledge and stigma and that these explanations vary by area of residence. Furthermore, some studies have found additional determinants of stigma, such as having seen a leprosy patient [21], regulations regarding leprosy [20], exposure to leprosy health promotion messages [13,19], marital status [16,22], economic status [24], ethnicity [14,16,28], distance of residence from the hospital [14] and migrant status [28]. We recommend including these variables in future studies, to get a better understanding of the underlying mechanisms behind stigma and knowledge. In addition, we think it would be helpful to explore the influence of religion better, not just taking beliefs, but also level of religious faith and dedication into account.

Health workers

We found that in both countries health workers had the highest leprosy knowledge levels. This is likely due to their (para)medical training, while some had also received leprosy training. Having had more training and/or more years of service has been associated with better knowledge about leprosy among health workers in other studies also [49–53]. In addition, all health workers had completed higher education, which was associated with higher levels of knowledge about leprosy in all four participant groups in our study. Even though health workers had higher levels of knowledge than the other participant groups, many still lacked knowledge, for example about mode of transmission and contagiousness of leprosy. Standard training on leprosy for all health workers and regular refresher courses could likely improve this. Interestingly, the topics on which health workers lacked knowledge (e.g. mode of transmission and contagiousness after treatment) and had adequate knowledge (e.g. treatment) was reflected in the knowledge about leprosy among the other participant groups. This likely shows that adequate knowledge about a particular topic among health workers enables them to pass on correct information to persons affected, contacts and community members. In addition, we believe it reflects the messages that have been used in past government education campaigns. These findings underline the importance of ensuring that health workers have correct knowledge about leprosy.

The present study also found that health workers had high mean stigma scores of 13.7 (Indonesia) and 15.0 (India) on the EMIC-CSS. Determinants of stigma for health workers only included age, this only explained 3% of the variability in level of stigma. Determinants of stigma among health workers should be explored further. Because health workers are in a respected position in the community, their attitudes and behaviour can influence how others perceive leprosy. Health workers are therefore an important group to target with stigma reduction interventions.

Interventions to improve the knowledge and perception of leprosy

Our findings indicate the need for effective interventions to positively influence the perception of leprosy and improve knowledge of leprosy. We believe our findings of local differences in knowledge gaps, misconceptions, beliefs and fears indicate that interventions should be culture-specific and contextualised [54,55]. This is expected to be much more effective to increase positive attitudes and acceptance of persons affected by leprosy than generic messages [40]. We believe our knowledge findings indicate that certain topics should be prioritized in health education in both countries: cause, mode of transmission, early symptoms and contagiousness of leprosy. These findings also show that some messages may be important as such, but do not have to be prioritized at the moment: knowledge about treatability of leprosy was good in both India and Indonesia. This is likely a reflection of the messages in past government education campaigns. While knowledge gaps can be addressed by information, attitudes, beliefs and fears require an additional approach. Changing knowledge and perceptions is best done as a combination of health education and behavioural change interventions [56,57].

Health education should target the general community, who had the highest stigma levels in both countries. This may be done by targeting key influencers and authority figures in the community, for example village leaders, who could influence others in the community and allow for the information to filter down.

Strengths and limitations

One of the limitations of this study is its cross-sectional design, which prevented us from making more definitive causal inferences. Another limitation of this study is the difference in sampling methods of close contact selection in India and

Indonesia. Close contacts in India were selected by convenience sampling, while close contacts in Indonesia were selected by random sampling. Despite differences in recruitment methods, the patterns of results showed parallels. Study strengths include the mixed-method approach that allowed for triangulation of the data.

Conclusion

Our study revealed poor knowledge regarding leprosy in India and Indonesia, especially regarding cause, mode of transmission, early symptoms and contagiousness of leprosy. Knowledge about treatment and treatability was good. Stigma levels were high in both countries and were driven by poor knowledge and misconceptions about leprosy, local beliefs, and fear of contagion. These findings show the importance of investigating the perceptions regarding leprosy in the communities targeted for educational interventions. Local misconceptions and beliefs, especially around the cause and mode of transmission of leprosy, differed in the two countries. Contextualised health education and behaviour change interventions are required to improve knowledge, reduce misconceptions and positively influence the perception of leprosy. Interventions should address specific knowledge gaps, beliefs and fears.

The determinants of leprosy knowledge and stigma explained only a small proportion of the variability in level of knowledge and stigma. Future studies should attempt to find additional determinants to get a better understanding of the underlying mechanisms behind stigma and knowledge and find ways to improve interventions. We also recommend that future studies explore the role of religion, religiosity and area of residence in local beliefs.

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References

1. Link BG, Phelan JC. Conceptualizing stigma. Annu Rev Sociol 2001;27:363–85.
2. Weiss MG, Ramakrishna J, Somma D. Health-related stigma: rethinking concepts and interventions. Psychol Health Med 2006;11:277–87.
3. Litt E, Baker MC, Molyneux D. Neglected tropical diseases and mental health: a perspective on comorbidity. Trends Parasitol 2012;28:195–201.
4. Van Brakel WH. Measuring health-related stigma—a literature review. Psychol Health Med 2006;11:307–34.
5. Staring ABP, Van der Gaag M, Van den Berge M, Duivenvoorden HJ, Mulder CL. Stigma moderates the associations of insight with depressed mood, low self-esteem, and low quality of life in patients with schizophrenia spectrum disorders. Schizophr Res 2009;115:363–9.
6. Tsutsumi A, Izutsu T, Islam AM, Maksuda AN, Kato H, Wakai S. The quality of life, mental health, and perceived stigma of leprosy patients in Bangladesh. Soc Sci Med 2007;64:2443–53.
7. Lastória JC, Abreu MAMM de. Leprosy: review of the epidemiological, clinical, and etiopathogenic aspects-part 1. An Bras Dermatol 2014;89:205–18.
8. Suzuki K, Akama T, Kawashima A, Yoshihara A, Yotsu RR, Ishii N. Current status of leprosy: epidemiology, basic science and clinical perspectives. J Dermatol 2012;39:121–9.
9. Navon L. Beggars, metaphors, and stigma: a missing link in the social history of leprosy. Soc Hist Med 1998;11:89–105.
10. Heijnders ML. The dynamics of stigma in leprosy. Int J Lepr 2004;72:437–47.
11. Heijnders ML. Experiencing leprosy: perceiving and coping with leprosy and its treatment. A qualitative study conducted in Nepal. Lepr Rev 2004;75:327–37.
12. Rafferty J. Curing the stigma of leprosy. Lepr Rev 2005;76:119–26.
13. Sermrittirong S, Van Brakel W. Stigma in leprosy: concepts, causes and determinants. Lepr Rev 2014;85:36–47.
14. Adhikari B, Shrestha K, Kaehler N, Raut S, Chapman SR. Community attitudes towards leprosy affected persons in Pokhara municipality of western Nepal. J Nepal Health Res Counc 2014.
15. O'Donoghue J, Ishengoma A, Masao H, Mbega M. Evaluation of a sustained 7-year health education campaign on leprosy in Rufiji District, Tanzania. Lepr Rev 1998;69:57–74.
16. Singh R, Singh B, Mahato S. Community knowledge, attitude, and perceived stigma of leprosy amongst community members living in Dhanusha and Parsa districts of Southern Central Nepal. PLoS Negl Trop Dis 2019;13:e0007075.
17. Seshadri D, Khaitan BK, Khanna N, Sagar R. The tangled web: a study of knowledge and attitude towards leprosy from a tertiary care hospital in India. Indian J Lepr 2014:27–41.
18. van't Noordende AT, Korfage I, Lisam S, Arif MA, Kumar A, van Brakel WH. The role of perceptions and knowledge of leprosy in the elimination of leprosy: A baseline study in Fatehpur district, northern India. PLoS Negl Trop Dis 2019;13:e0007302.
19. Ebenso B, Newell J, Emmel N, Adeyemi G, Ola B. Changing stigmatisation of leprosy: an exploratory, qualitative life course study in Western Nigeria. BMJ Glob Heal 2019;4:e001250.
20. Sottie CA, Darkey J. Living with stigma: Voices from the Cured Lepers' village in Ghana. Soc Work Health Care 2019;58:151–65.

21. Tabah EN, Nsagha DS, Bissek A-CZ-K, Njamnshi TN, Njih IN-N, Pluschke G, et al. Community knowledge, perceptions and attitudes regarding leprosy in rural Cameroon: The case of Ekondotiti and Mbonge health districts in the South-west Region. PLoS Negl Trop Dis 2018;12:e0006233.
22. Ibikunle PO, Nwokeji SC. Assessment of stigma among people living with Hansen's disease in south-east Nigeria. Lepr Rev 2017;88:43–57.
23. Tekle-Haimanot R, Forsgren L, Gebre-Mariam A, Abebe M, Holmgren G, Heijbel J, et al. Attitudes of rural people in central Ethiopia towards leprosy and a brief comparison with observations on epilepsy. Lepr Rev 1992;63:157–68.
24. Raju MS, Reddy J V. Community attitude to divorce in leprosy. Indian J Lepr 1995;67:389–403.
25. Kaehler N, Adhikar B, Raut S, Marahatta SB, Chapman RS. Perceived stigma towards leprosy among community members living close to Nonsomboon leprosy Colony in Thailand. PLoS One 2015;10:e0129086.
26. Premkumar R, Kumar KS, Dave SL. Understanding the attitude of multidisciplinary teams working in leprosy. Lepr Rev 1994;65:66–77.
27. Saha G, Mandal NK, Dutta RN. Current Perceptions and Practices (KAP) about Leprosy among Leprosy Patients: A Comparative Study between High Prevalent & Low Prevalent Districts of West Bengal. Indian J Lepr 2015;87:1–16.
28. Faatoese A, Sikaleti S, Priest P, Chambers S. Knowledge and attitudes to leprosy of Pacific People living in New Zealand. Lepr Rev 2016;87:368–77.
29. Kanodia SK, Dixit AM, Shukla SR, Seth AK, Balothia V, Gupta R. A study on knowledge, beliefs and attitude towards leprosy in students of Jaipur, Rajasthan. Indian J Lepr 2012;84:277–85.
30. Nsagha DS, Bissek A-CZK, Nsagha SM, Njunda AL, Assob JCN, Tabah EN, et al. Social stigma as an epidemiological determinant for leprosy elimination in Cameroon. J Public Health Africa 2011;2.
31. World Health Organization. Weekly Epidemiological Record, 30 August 2019. Global leprosy update, 2018: moving towards a leprosy-free world. vol. 94. 2019.
32. National Leprosy Eradication Programme. District wise annual new case detection and prevalence as on March 2017. 2017.
33. Ballering A V, Peters RMH, Waltz MM, Arif MA, Mishra CP, BRAKEL W, et al. Community stigma and desired social distance towards people affected by leprosy in Chandauli District, India. Lepr Rev 2019;90:418–32.
34. Mieras L, Singh MK, Manglani PR, Arif MM, Banstola NL, Pandey B, et al. A single-dose of rifampicin to prevent leprosy; quantitative analysis of impact on perception, attitudes and behaviour of persons affected, contacts and community members towards leprosy in India, Nepal and Indonesia. Lepr Rev 2020:Accepted for publication.
35. Peters R, Mieras L, Subedi M, Apte H, Koesbardiati T, Banstola NL, et al. A single dose of rifampicin to prevent leprosy: qualitative analysis of perceptions of persons affected, contacts, community members and health professionals towards chemoprophylaxis and the impact on their attitudes in India, Nepal and Indonesia. Lepr Rev 2018;89:335–52.
36. Rensen C, Bandyopadhyay S, Gopal PK, Van Brakel WH. Measuring leprosy-related stigma–a pilot study to validate a toolkit of instruments. Disabil Rehabil 2011;33:711–9.
37. Peters RMH, Van Brakel WH, Zweekhorst MBM, Damayanti R, Bunders JFG. The cultural validation of two scales to assess social stigma in leprosy. PLoS Negl Trop Dis 2014;8:e3274.

38. Hofstraat K, van Brakel WH. Social stigma towards neglected tropical diseases: a systematic review. Int Health 2016;8:i53–70.
39. de Stigter DH, Geus LD, Heynders ML. Leprosy: between acceptance and segregation. Community behaviour towards persons affected by leprosy in eastern Nepal. Lepr Rev 2000;71:492–8.
40. Brown W. Can social marketing approaches change community attitudes towards leprosy? Lepr Rev 2006;77:89–98.
41. Grewal I, Negi Y, Kishore J, Adhish S V. Knowledge and attitude about Leprosy in Delhi in post elimination phase. Indian J Lepr 2013;85.
42. Barkataki P, Kumar S, Rao PS. Knowledge of and attitudes to leprosy among patients and community members: a comparative study in Uttar Pradesh, India. Lepr Rev 2006;77:62–8.
43. Mankar MJ, Joshi SM, Velankar DH, Mhatre RK, Nalgundwar AN. A comparative study of the quality of life, knowledge, attitude and belief about leprosy disease among leprosy patients and community members in Shantivan Leprosy Rehabilitation centre, Nere, Maharashtra, India. J Glob Infect Dis 2011;3:378.
44. Girão RJS, Soares NLR, Pinheiro JV, da Paz Oliveira G, De Carvalho SMF, De Abreu LC, et al. Leprosy treatment dropout: A sistematic review. Int Arch Med 2013;6:34.
45. Sermrittirong S, Van Brakel WH, Kraipui N, Traithip S, Bunders-Aelen JF. Comparing the perception of community members towards leprosy and tuberculosis stigmatization. Lepr Rev 2015;86:54–61.
46. Peters RMH, Zweekhorst MBM, Bunders JFG, van Brakel WH. A cluster-randomized controlled intervention study to assess the effect of a contact intervention in reducing leprosy-related stigma in Indonesia. PLoS Negl Trop Dis 2015;9:e0004003.
47. Van Brakel WH, Sihombing B, Djarir H, Beise K, Kusumawardhani L, Yulihane R, et al. Disability in people affected by leprosy: the role of impairment, activity, social participation, stigma and discrimination. Glob Health Action 2012;5:18394.
48. Oliveira HX. Adaptação transcultural das escalas de estigma Explanatory Model Interview Catalogue (EMIC) na perspectiva de pessoas acometidas pela hanseníase e da comunidade para o contexto brasileiro. Universidade Federal do Ceará, 2018.
49. Jain M, Sharma A, Jain V, Virjee K, Singh S. Knowledge and Attitude about Leprosy among Indian Dental Students in Faridabad. J Clin Diagnostic Res JCDR 2016;10:ZC48.
50. Giri PA, Phalke DB, Aarif SM. A study of knowledge, attitude and practices regarding leprosy among undergraduates and interns of a medical college and hospital from rural India. Indian J Lepr 2011;83:75–80.
51. Kar S, Ahmad S, Pal R. Current knowledge attitudes, and practices of healthcare providers about leprosy in Assam, India. J Glob Infect Dis 2010;2:212.
52. Asnake MK, Ahmed M, Genebo T, Mekebib B. Knowledge and attitude of health professionals in relation to the integration of leprosy control into the general health service in Ethiopia. Int J Lepr Other Mycobact Dis 2000;68:299.
53. Rao P V., Rao SL, Vijayakrishnan B, Chaudhary AB, Peril S, Reddy BP, et al. Knowledge, attitude and practices about leprosy among medical officers of Hyderabad urban district of Andhra Pradesh. Indian J Lepr 2007;79:27–43.
54. Opala J, Boillot F. Leprosy among the Limba: illness and healing in the context of world view. Soc Sci Med 1996;42:3–19.

55. Chen PC, Sim HC. The development of culture-specific health education packages to increase case-finding of leprosy in Sarawak. Southeast Asian J Trop Med Public Health 1986;17:427–32.

56. Seligman HK, Wallace AS, DeWalt DA, Schillinger D, Arnold CL, Shilliday BB, et al. Facilitating behavior change with low-literacy patient education materials. Am J Health Behav 2007;31:S69–78.

57. Noar SM, Benac CN, Harris MS. Does tailoring matter? Meta-analytic review of tailored print health behavior change interventions. Psychol Bull 2007;133:673.

CHAPTER 4

Towards a cross-neglected tropical disease perception study toolkit: a prototype toolkit developed in the field of leprosy

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Abstract

Objectives

A negative perception of leprosy and other neglected tropical diseases (NTDs) is a problem that is seen globally. It results in discrimination, social exclusion and widespread mental health problems. There is a need for a standardised toolkit to assess the different aspects of perception of leprosy or other NTDs, including essential knowledge of these conditions.

Methods

We developed the Perception Study Toolkit (PST). This toolkit consists of four measures, a Communication Needs Assessment questionnaire, Knowledge Attitudes and Practices measures, the EMIC community stigma scale and the Social Distance Scale. It also comprises qualitative methods to investigate perception: the way people see leprosy, what they know about leprosy and their attitudes, beliefs and reported behaviour towards persons affected by leprosy. The PST is a toolkit and comprises separate instruments that assess different aspects of perception, it is also possible to use only one or a few of the instruments of the PST.

Results

This is not applicable because this is not a study.

Conclusions

The PST can help identify specific beliefs, knowledge gaps, misconceptions and fears to inform community education and behaviour change interventions and can be used to monitor and evaluate such interventions. Using a standard toolkit like the PST would enable assessment of the perception of leprosy or other NTDs that would allow comparison across projects and countries including monitoring of changes over time.

Introduction

Leprosy is an infectious neglected tropical disease (NTD) that primarily affects the peripheral nerves and skin. If left untreated or detected late, leprosy may lead to severe visible and permanent disabilities (1,2). Persons affected by leprosy often experience stigma and discrimination, especially if they have visible impairments due to their condition (3,4).

Health-related stigma refers to a negative social response to a disease. Stigma exists 'when elements of labeling, stereotyping, separation, status loss, and discrimination co-occur in a power situation that allows the components of stigma to unfold' (5). Discrimination is enacted stigma, it is acting on negative attitudes (5,6). Stigma does not always have to result in discrimination (e.g. people who have negative feelings or attitudes towards a person or group, but who do not act on this). Discrimination may also be caused or aggravated by other factors (e.g. fear, racism, sexism) (6).

Stigma and discrimination affect and disrupt many areas of a person's life, such as family and social life, social participation and mental wellbeing (4,7–10). Disabilities due to the condition may cause similar effects. Leprosy causes disability and is severely stigmatised. As a result, persons affected by leprosy may suffer depression, anxiety, suicide (attempts), mental distress, low self-esteem and low quality of life (11). Because of disability, stigma and poor mental health, they often experience social participation restrictions such as barriers to employment and social life, education or marriage (12). This may impose a social and economic burden on already marginalized families (9,13). Family members and friends may also experience stigma and discrimination (11). In addition, a negative perception of leprosy makes people reluctant to seek treatment, which may result in a delay in diagnosis or cure, increasing the risk of impairments that otherwise would have been prevented (2). Stigma is also a barrier to case finding and treatment adherence, and therefore timeliness and effectiveness of treatment (7,14,15).

Perception is how individuals or groups "see" an object, person, event or institution (16–18). Perception is a broad concept; it may refer to how an individual or group "sees" others (social perception) (16,17) but also refers to a person's interpretation and understanding of a disease and its potential consequences (disease or illness perception) (18). Perception comprises knowledge, beliefs, attitudes and emotions, which in turn influence and are influenced by personal factors (e.g. personality, experience) and environmental factors (e.g. culture, religion) (16,17). . Perceptions about leprosy, such as knowledge, attitudes and cultural beliefs, play an important role in stigma and early case finding (14,19–28). For example, people's knowledge influences their awareness that signs and symptoms are due to leprosy which, if awareness is lacking, hampers early reporting of leprosy (29–31). In addition, community attitudes towards persons affected by leprosy are strongly influenced by local misconceptions, cultural and religious beliefs and fears that may be linked to this. For example, in some cultures leprosy is linked to supernatural causes, witchcraft, sins or immoral behaviour, or believed to be hereditary (19,22,26,28,29). These misconceptions and beliefs are often different in different cultures and countries. Understanding these, as well as local knowledge gaps, is crucial to understand explanatory models, attitudes and behaviour (29,32).

Efforts to address stigma and discrimination have focused on a number of areas including the stigmatised person, the stigmatising context and policy and systems (33,34). Positive outcomes in terms of reduced stigma have been reported for interventions like peer counselling (35–37), direct contact interventions such as community meetings, and indirect contact such as participatory videos and comics (37–40). For interventions to be effective, i.e. to increase positive attitudes and acceptance of persons affected by leprosy, it is important that they are culture-specific and address local beliefs and knowledge gaps (32). Only by understanding and addressing these local beliefs and perceptions, can we design effective public messages about leprosy and other interventions. Much leprosy-related suffering can be prevented by positively influencing the perception of leprosy and by reducing or eliminating stigma.

Many knowledge, attitudes and practices (KAP) surveys and stigma studies have been conducted. These studies have used a variety of tools: in-depth interviews and/or focus group discussions, validated questionnaires or questionnaires developed specifically for the study. At least 33 studies have developed their own questionnaire to assess knowledge about leprosy (24,41–44) or knowledge, attitudes and practices about leprosy (20,22,27,28,45–68). These questionnaires are often not described well or not included in the articles. The field of leprosy would benefit from having more standardised tools, also for assessing the perception of leprosy to be able to measure progress and compare between projects.

Recently, 'leprosy perception studies' were conducted in endemic districts in Brazil, India, Nepal and Indonesia (26,69–72). The purpose of these studies was to investigate the perceptions regarding leprosy, i.e., the way people see leprosy, what they know about leprosy and their attitudes, beliefs, fears and reported behaviour towards persons affected by leprosy. Information of these studies is used to develop context-specific community education and behaviour change interventions that are implemented to raise awareness about leprosy and to improve knowledge, attitudes and behaviour towards (persons affected by) leprosy in the context of the PEP++ project, a cluster-randomised trial to test an enhanced regimen for post-exposure prophylaxis against leprosy. The objective of this paper is to describe the Perception Study Toolkit (PST), which was used in the PEP++ project, to implement 'perception studies' for the assessment and monitoring of knowledge, attitudes and practices regarding leprosy. We propose that the PST could form the basis for developing a standard toolkit to assess perception of leprosy and, with minor adaptations, other NTDs that would enable comparison across projects, countries and would allow monitoring of changes over time. We are not aware of an alternative toolkit to measure perception that is used in the field of NTDs.

Material and methods

Purpose of the Perception Study Toolkit

The purpose of the PST is to investigate the perceptions regarding leprosy: the way people see leprosy, what they know about leprosy and their attitudes, beliefs, fears and reported behaviour towards persons affected by leprosy. With minor adaptations, the PST can also be used for other NTDs. It should be noted that the PST is a toolkit and not one instrument with different subscales (it is not intended to calculate an overall score). The PST comprises separate instruments that assess different aspects of perception, it is also possible to use only one or a few of the instruments of the PST, depending on the purpose of the study.

Content of the Perception Study Toolkit

The PST comprises questions to assess knowledge regarding key aspects of leprosy, such as what patients were told about their diagnosis, how they would prefer to call the disease, what people believe to be early symptoms and causes of leprosy, how it transmitted, whether it can be treated, etc. Perceived attitudes, behaviours and fears regarding leprosy in the community are explored and measured using the EMIC Community Stigma Scale (EMIC-CSS) and the Social Distance Scale (SDS). In addition, semi-structured interviews and focus group discussions are held to obtain an in-depth perspective to complement and help interpret the quantitative data. Demographic data are also obtained from all participants. Each instrument is briefly described below. An overview can be found in Table 1.

Table 1. Instruments included in the Perception Study Toolkit.

Tool	Type of tool	Purpose of tool
Demographic information form	Form	To collect basic participant information such as name, age, gender, address, occupation and education
KAP Questionnaire	Questionnaire	To assesses knowledge, attitudes and beliefs of individuals regarding leprosy
Explanatory Model Interview Catalogue Community Stigma Scale (EMIC-CSS)	Scale	To assess perceived attitudes and behaviour towards affected persons of community members in general
Social Distance Scale (SDS)	Scale	To assess how close a contact or relationship a respondent is willing to have to a person affected by leprosy as a proxy for their attitudes
Communication Needs Assessment (CNA) questionnaire	Questionnaire	To assess what means of communication people are using, what means they are familiar with and what means they find most acceptable
Semi-structured interviews	In-depth interview	To establish an in-depth conversation and understanding regarding perceptions towards leprosy
Focus group discussions (FGDs)	Focus group discussion	To provide a broad and diverse spectrum of opinions and ideas on knowledge and perceptions towards leprosy.

Demographic information

A demographic information form can be used to collect basic participant information such as name, age, gender, address, occupation and education. It is important to collect data on factors that may influence perception of leprosy or for practical reasons such as follow-up or dissemination of results.

KAP Questionnaire

The 17-item KAP questionnaire assesses knowledge, attitudes and beliefs of individuals regarding leprosy. The questionnaire consists of yes/no/don’t know questions and multiple answer questions. Topics include perception regarding cause and treatment of the disease, emotions after hearing the diagnosis and the emotions and attitudes of family, friends and neighbours towards leprosy. All questions are open ended, it is therefore important not to read out the answer options. Eight items on the KAP questionnaire can be used to assess knowledge of leprosy, to calculate a total knowledge score. On some of the questions, multiple answers are possible. A total score of nine can be obtained on the KAP when considering answers correct if the correct answers were given regardless of any incorrect answers. ‘Adequate knowledge’ is defined as a score of six or more out of nine on the KAP (>67% correct), ‘moderate knowledge’ as a score between 3 and 6 (33-67% correct) and ‘poor knowledge’ as a score of three or less (<33% correct) on the KAP questionnaire. The KAP questionnaire has been used in several leprosy studies in India, Nepal, Indonesia and Brazil between 2012 and 2019, reports of four of these studies have been published to date (26,69,70,72).

EMIC Community stigma scale

The 15-item Explanatory Model Interview Catalogue Community Stigma Scale (EMIC-CSS) measures perceived attitudes and behaviour towards affected persons of community members in general. The EMIC-CSS covers areas of life that may be affected by stigma, such as concealment, avoidance, pity, shame, being made fun of, respect and marriage (prospects). During the interview, the participant is asked to respond to 15 questions, offering four response options that are valued with different scores: yes (2), possibly (1), no (0) and don't know (0). The EMIC-CSS total score ranges from zero (no negative attitudes) to 30 (most negative attitudes). The EMIC-CSS has been validated among community members of persons affected by leprosy in Brazil, India, Nepal and Indonesia (24,73–75).

Social Distance Scale

The 7-item Social Distance Scale (SDS) measures how close a contact or relationship a respondent is willing to have to a person affected by leprosy as a proxy for their attitudes. The interviewee will start the SDS by reading the gender-specific vignette, a short description or word picture of a person with leprosy (separate for men and women). The vignette is followed by seven questions concerning the person in the vignette. The participant can respond to the questions by choosing one of the four options which are valued with different scores, i.e. definitely willing (0), probably willing (1), probably not willing (2) or definitely not willing (3). The SDS total score ranges from zero (no negative attitudes or fear) to 21 (most negative attitudes/fear). The SDS has been validated among community members of persons affected by leprosy in Indonesia (74). The SDS has not been formally validated for use with persons affected by leprosy in India, but has been translated to Hindi, and partially validated and used in studies in Uttar Pradesh and Dadra & Nagar Haveli, India (69,70).

Communication needs assessment

The 7-item Communication Needs Assessment (CNA) questionnaire is used to assess what means of communication people are using, what means they are familiar with and what means they find most acceptable. The results are used to inform the choice of media when designing education and behaviour change interventions. All questions on the CNA are open ended. Since questions are context-specific, for example 'what TV programmes do you usually watch?', the answer options should be adjusted according to make them appropriate for the context they are being used in. The CNA is only used when the PTS is used to develop community education and behaviour change interventions, as input/ inventory of communication preference.

Semi-structured interviews

The aim of the semi-structured interviews is to establish an in-depth conversation regarding perceptions towards leprosy. The interview guide provides the interviewer with a clear layout of the interview and, depending on type of respondent, consists of 10-15 open questions.

Focus group discussions

Focus group discussions provide a broad and diverse spectrum of opinions and ideas on knowledge, sources of knowledge and reasons behinds certain views and perceptions and allows checking of views expressed by individuals or ranking of e.g. perceived frequency of opinions about components of perception or causative factors. During the focus group, the facilitator will use a question and topic guide as well as a timetable to conduct the discussion smoothly.

Experiences with its use

The PST has been used in several studies in Brazil, India, Nepal and Indonesia already (26,69–72). The instruments can be used to investigate (gaps in) knowledge, specific fears, attitude and practices and to compare scores between participant groups, countries and in different points in time. Some examples of the use of the PST are provided below.

Investigate and assess perceptions regarding leprosy

Ballering and colleagues (69) found that community members in Chandauli district in India had a more tolerant and accepting attitude when it comes to renting out a room to, being a colleague of or living next to a person cured from leprosy. At the same time and confirming findings from elsewhere, a majority of respondents were reluctant to have a person affected by leprosy as a caretaker of their children, or have a person affected by leprosy marry one of their children (69). The authors also report total scores on the KAP measure, SDS and EMIC-CSS (69).

Interaction between knowledge, beliefs, attitudes and fears

The PST has been used in a study in Fatehpur district, Uttar Pradesh, India, to examine the interaction between knowledge, beliefs, attitudes and fears (26). Using multivariate regression analysis, the authors found that health workers, participants who knew someone affected by leprosy, men, and participants with better knowledge of leprosy had more positive attitudes towards persons affected by leprosy (26). The authors also determined the factors that had an independent effect on knowledge and community stigma. These findings were later used as input for developing context-specific community education and behaviour change interventions (not yet published).

Added benefit of using a mixed methods approach

The study in Fatehpur district, India (26) also illustrates the complementary nature of the in-depth interviews and focus group discussions. While the EMIC and SDS scales revealed negative attitudes and stigma towards persons affected by leprosy, the interviews provided in-depth insights in the reasons for exclusion. Avoidance of persons affected by leprosy was often linked to the belief that leprosy transmits by touch and (fear of) transmission (26). In addition, a study that used the PST in India, Nepal and Indonesia emphasized '(…) the importance of collecting both quantitative and qualitative data on a given topic, since results from the latter are often not generalisable' (70).

Evaluation of an intervention

Mieras and colleagues (70) used the PST to assess the impact of the Leprosy Post-Exposure Prophylaxis (LPEP) project approximately one year after the start of its implementation in India, Nepal and Indonesia. They included persons affected by leprosy, close contacts and community members in each country before and after the start of the intervention. Their study provided insight in changes in KAP measure, EMIC-CSS and SDS scores in each of these groups in the three countries. They found a significant increase in knowledge of leprosy (from 4% correct to 29% correct answers on the KAP measure) and a decrease in SDS and EMIC-CSS stigma scores in community members in Nepal after the intervention (70).

The implementation protocol of the PST, that comprises recommendations for using the toolkit in the field, can be found as additional information file 2.

Discussion

This paper describes the Perception Study Toolkit. The toolkit consists of qualitative and quantitative (mixed) methods to allow for triangulation and provide rich data. This allows for a comprehensive understanding of the perception of leprosy. By using a standard toolkit to assess perception of leprosy, data can be compared across projects and countries, and progress can be monitored over time. We are not aware of an alternative toolkit to measure perception that is used in the field of NTDs.

We found 33 knowledge, attitudes and practices (KAP) surveys and stigma studies that developed their own questionnaire to assess knowledge about leprosy (24,41–44) or knowledge, attitudes and practices about leprosy (20,22,27,28,45–68). These questionnaires were often not described well and were not included in the articles. The KAP questionnaire used in the PST Toolkit covers eight main topics: early symptoms, cause, mode of transmission, treatment, prevention, curability, contagiousness when on treatment and prevention of disabilities. The questions about curability and prevention have been added after consultation with leprosy experts, these two questions have not yet been pilot tested. We think the field of leprosy would benefit from having more standardised tools assessing the perception of leprosy and believe the KAP questionnaire could be such a standardised tool. We called the KAP questionnaire a questionnaire, because it has not been formally validated as a scale. This should be done in a future study, but we would first like to offer it to readers of this article for further improvement. Readers are invited to suggest further improvements to the KAP questionnaire.

Standardized tools used in other KAP or stigma studies are the EMIC-CSS (23,24,28,37,41,44,76) and de SDS (23,37). The EMIC-CSS and SDS are also included in the PST. Since the EMIC-CSS questions are phrased in a more general way (for example ‘would leprosy be a problem for a person to get married?’) and the SDS questions are directed to the responded personally (for example ‘how [would *you* feel about] about having one of your children marry someone like [name of person affected]?’), using both the EMIC-CSS and the SDS allows for the exploration of community and personal attitudes towards persons affected by leprosy. Interestingly, some studies using KAP or stigma surveys among persons affected by leprosy have also included tools to assess perceived or internalised stigma or social participation. The Participation Scale (23,77–79), which measures social participation, an outcome directly affected by stigma, and the EMIC stigma scale for affected persons (42,78,79) have most frequently been used, followed by the Internalized Stigma of Mental Illness scale (23,79), the Jacoby stigma scale (78), Discrimination Assessment Form (78) and the Leprosy Dehabilitation Scale (80). Even though the KAP questionnaire in the PST contains five questions about stigma for persons affected, we believe the PST would benefit from adding a tool to assess internalised and/or perceived stigma. The EMIC affected persons (EMIC-AP) (81) would allow for comparison with the EMIC-CSS, given the similarities in the questions asked in the EMIC-AP and the EMIC-CSS. An alternative would be the Anticipated stigma sub-scale (4 items) and Internalised stigma sub-scale (6 items) of the SARI Stigma Scale (82).

Besides assessing and monitoring knowledge, attitudes and practices regarding leprosy, the toolkit can also be used to develop culture-specific messages for health education, and to evaluate the impact of interventions. Even though low knowledge of leprosy has been associated with high levels of stigma (20,24,26–28), high levels of knowledge alone do not lead to more positive attitudes and behaviour towards persons affected by leprosy (83). Understanding the cultural belief systems in countries where leprosy is endemic is essential to understand and challenge stigma (84). Interviews with the target group to understand where behaviour comes from are crucial when developing interventions to change behaviour (85). A protocol that is often used to develop behaviour change interventions is intervention mapping. Intervention mapping is a six-step protocol that describes the path from problem identification to problem mitigation or problem solving (86). The PST covers step one to three of intervention mapping: a needs assessment or problem analysis and identifying which beliefs should be targeted (KAP, EMIC-CSS, SDS and interviews) and exploring media that fit the context of the target group (CNA). To our knowledge, 'communication needs assessments' are not often done in the leprosy field, when designing interventions for behaviour change (87). Step four to six of intervention mapping deal with drafting materials and designing implementation and evaluation plans (86).

Determining whether changes pre-and post-intervention can be attributed to the intervention is challenging. We have listed several strategies to best deal with this in our recommendations for implementation (additional file 2). These strategies include appropriate timing of assessments, including a control group, cluster randomisation, adding specific questions about involvement in interventions in the follow-up assessment and implementing interventions step-wise if multiple interventions are implemented. The amount of time that should have elapsed since the intervention before it is evaluated depends on the type of intervention that is evaluated. Given that long-term effects are often desired, at least one year after the intervention implementation was completed would seem a reasonable period.

Readers are invited to suggest further improvements to the toolkit. Recommendations for the implementation of the toolkit in the field can be found as additional file (S2).

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References

1. Lastória JC, Abreu MAMM de. Leprosy: review of the epidemiological, clinical, and etiopathogenic aspects-part 1. An Bras Dermatol. 2014;89(2):205–18.
2. Suzuki K, Akama T, Kawashima A, Yoshihara A, Yotsu RR, Ishii N. Current status of leprosy: epidemiology, basic science and clinical perspectives. J Dermatol. 2012;39(2):121–9.
3. Navon L. Beggars, metaphors, and stigma: a missing link in the social history of leprosy. Soc Hist Med. 1998;11(1):89–105.
4. Adhikari B, Kaehler N, Raut S, Marahatta SB, Ggyanwali K. Risk factors of stigma related to leprosy-A systematic review. J Manmohan Meml Inst Heal Sci. 2013;1(2):3–11.
5. Link BG, Phelan JC. Conceptualizing stigma. Annu Rev Sociol. 2001;27(1):363–85.
6. Deacon H. Understanding HIV/AIDS stigma: A theoretical and methodological analysis. HSRC press; 2005.
7. Van Brakel WH. Measuring health-related stigma—a literature review. Psychol Health Med. 2006;11(3):307–34.
8. Heijnders ML. The dynamics of stigma in leprosy. Int J Lepr. 2004;72:437–47.
9. van't Noordende AT, Aycheh MW, Schippers A. The impact of leprosy, podoconiosis and lymphatic filariasis on family quality of life: A qualitative study in Northwest Ethiopia. PLoS Negl Trop Dis. 2020;14(3):e0008173.
10. van't Noordende AT, van Brakel WH, Banstola N, Dhakal KP. The impact of leprosy on marital relationships and sexual health among married women in eastern Nepal. J Trop Med. 2016;2016.
11. Somar PMW, Waltz MM, van Brakel WH. The impact of leprosy on the mental wellbeing of leprosy-affected persons and their family members–a systematic review. Glob Ment Heal. 2020;7.
12. Hofstraat K, van Brakel WH. Social stigma towards neglected tropical diseases: a systematic review. Int Health. 2016;8(suppl_1):i53–70.
13. Awofeso N. Stigma and socio-economic reintegration of leprosy sufferers in Nigeria. Acta Leprol. 1996;10(2):89–91.
14. Sermrittirong S, Van Brakel W. Stigma in leprosy: concepts, causes and determinants. Lepr Rev. 2014;85:36–47.
15. Heijnders ML. Experiencing leprosy: perceiving and coping with leprosy and its treatment. A qualitative study conducted in Nepal. Lepr Rev. 2004;75(4):327–37.
16. Pickens J. Attitudes and perceptions. Organ Behav Heal care. 2005;4(7).
17. Hiebert PG. Transforming worldviews: An anthropological understanding of how people change. Baker Academic; 2008.
18. Broadbent E, Wilkes C, Koschwanez H, Weinman J, Norton S, Petrie KJ. A systematic review and meta-analysis of the Brief Illness Perception Questionnaire. Psychol Health. 2015;30(11):1361–85.
19. Ebenso B, Newell J, Emmel N, Adeyemi G, Ola B. Changing stigmatisation of leprosy: an exploratory, qualitative life course study in Western Nigeria. BMJ Glob Heal. 2019;4(2):e001250.
20. Seshadri D, Khaitan BK, Khanna N, Sagar R. The tangled web: a study of knowledge and attitude towards leprosy from a tertiary care hospital in India. Indian J Lepr. 2014;27–41.
21. Sottie CA, Darkey J. Living with stigma: Voices from the Cured Lepers' village in Ghana. Soc Work Health Care. 2019;58(2):151–65.

22. Tabah EN, Nsagha DS, Bissek A-CZ-K, Njamnshi TN, Njih IN-N, Pluschke G, et al. Community knowledge, perceptions and attitudes regarding leprosy in rural Cameroon: The case of Ekondotiti and Mbonge health districts in the South-west Region. PLoS Negl Trop Dis. 2018;12(2):e0006233.
23. Ibikunle PO, Nwokeji SC. Assessment of stigma among people living with Hansen's disease in south-east Nigeria. Lepr Rev. 2017;88:43–57.
24. Adhikari B, Shrestha K, Kaehler N, Raut S, Chapman SR. Community attitudes towards leprosy affected persons in Pokhara municipality of western Nepal. J Nepal Health Res Counc. 2014;
25. Tekle-Haimanot R, Forsgren L, Gebre-Mariam A, Abebe M, Holmgren G, Heijbel J, et al. Attitudes of rural people in central Ethiopia towards leprosy and a brief comparison with observations on epilepsy. Lepr Rev. 1992;63(2):157–68.
26. van't Noordende AT, Korfage I, Lisam S, Arif MA, Kumar A, van Brakel WH. The role of perceptions and knowledge of leprosy in the elimination of leprosy: A baseline study in Fatehpur district, northern India. PLoS Negl Trop Dis. 2019;13(4):e0007302.
27. van den Broek J, O'Donoghue J, Ishengoma A, Masao H, Mbega M. Evaluation of a sustained 7-year health education campaign on leprosy in Rufiji District, Tanzania. Lepr Rev. 1998;69(1):57–74.
28. Singh R, Singh B, Mahato S. Community knowledge, attitude, and perceived stigma of leprosy amongst community members living in Dhanusha and Parsa districts of Southern Central Nepal. PLoS Negl Trop Dis. 2019;13(1):e0007075.
29. Opala J, Boillot F. Leprosy among the Limba: illness and healing in the context of world view. Soc Sci Med. 1996;42(1):3–19.
30. Da Silva Souza C, Bacha JT. Delayed diagnosis of leprosy and the potential role of educational activities in Brazil. Lepr Rev. 2003;74:249–58.
31. Robertson LM, Nicholls PG, Butlin R. Delay in presentation and start of treatment in leprosy: experience in an out-patient clinic in Nepal. Lepr Rev. 2000;71(5):511–6.
32. Brown W. Can social marketing approaches change community attitudes towards leprosy? Lepr Rev. 2006;77(2):89–98.
33. Cook JE, Purdie-Vaughns V, Meyer IH, Busch JTA. Intervening within and across levels: A multilevel approach to stigma and public health. Soc Sci Med. 2014;103:101–9.
34. Van't Noordende AT, Kuipers P, Pereira ZBD. Strengthening personal and family resilience: a literature review for the leprosy context. Lepr Rev. 2019;90(1):88–104.
35. Lusli M, Peters RMH, Zweekhorst MB, Van Brakel WH, Seda FS. Lay and peer counsellors to reduce leprosy-related stigma—lessons learnt in Cirebon, Indonesia. Lepr Rev. 2015;86:37–53.
36. Lusli M, Peters R, van Brakel W, Zweekhorst M, Iancu S, Bunders J, et al. The impact of a rights-based counselling intervention to reduce stigma in people affected by leprosy in Indonesia. PLoS Negl Trop Dis. 2016;10(12):e0005088.
37. Dadun D, Van Brakel WH, Peters RMH, Lusli M, Zweekhorst M, Bunders JGF, et al. Impact of socio-economic development, contact and peer counselling on stigma against persons affected by leprosy in Cirebon, Indonesia–a randomised controlled trial. Lepr Rev. 2017;88:2–22.
38. Peters RMH, Zweekhorst MBM, Bunders JFG, van Brakel WH. A cluster-randomized controlled intervention study to assess the effect of a contact intervention in reducing leprosy-related stigma in Indonesia. PLoS Negl Trop Dis. 2015;9(10):e0004003.

39. Peters R, Lusli M, Zweekhorst M, Miranda-Galarza B, van Brakel W, Irwanto, et al. Learning from a leprosy project in Indonesia: making mindsets explicit for stigma reduction. Dev Pract. 2015;25(8):1105–19.
40. Peters RMH, Zweekhorst MBM, van Brakel WH, Bunders JFG, Irwanto. 'People like me don't make things like that': Participatory video as a method for reducing leprosy-related stigma. Glob Public Health. 2016;11(5–6):666–82.
41. Kaehler N, Adhikar B, Raut S, Marahatta SB, Chapman RS. Perceived stigma towards leprosy among community members living close to Nonsomboon leprosy Colony in Thailand. PLoS One. 2015;10(6):e0129086.
42. Adhikari B, Kaehler N, Chapman RS, Raut S, Roche P. Factors affecting perceived stigma in leprosy affected persons in western Nepal. PLoS Negl Trop Dis. 2014;8(6):e2940.
43. Kar S, Ahmad S, Pal R. Current knowledge attitudes, and practices of healthcare providers about leprosy in Assam, India. J Glob Infect Dis. 2010;2(3):212.
44. Faatoese A, Sikaleti S, Priest P, Chambers S. Knowledge and attitudes to leprosy of Pacific People living in New Zealand. Lepr Rev. 2016;87:368–77.
45. Rao P V., Rao SL, Vijayakrishnan B, Chaudhary AB, Peril S, Reddy BP, et al. Knowledge, attitude and practices about leprosy among medical officers of Hyderabad urban district of Andhra Pradesh. Indian J Lepr. 2007;79(1):27–43.
46. ØSTBYE T. Knowledge, attitudes and practices relating to leprosy among public health care providers in Colombo, Sri Lanka. Lepr Rev. 2017;88:75–84.
47. Grewal I, Negi Y, Kishore J, Adhish S V. Knowledge and attitude about Leprosy in Delhi in post elimination phase. Indian J Lepr. 2013;85(93).
48. Graciano-Machuca O, Velarde-de la Cruz EE, Ramírez-Dueñas MG, Alvarado-Navarro A. University students' knowledge and attitudes towards leprosy. J Infect Dev Ctries. 2013;7(09):658–64.
49. John AS, Rao OS. Awareness and attitudes towards leprosy in urban slums of Kolkata, India. Indian J Lepr. 2009;81(3):135–40.
50. Bajaj DR, Matlani BL, Soomro FR, Iqbal MP. Knowledge, attitude and practices regarding leprosy among general practitioners at Hyderabad. J Coll Physicians Surg Pak. 2009;19(4): 215–8.
51. Chen S-M, Zhang L, Liu D-C, Liu H-X. Assessment of knowledge and skills in early diagnosis of leprosy and attitudes towards leprosy amongst doctors working in dermatological services, Shandong Province, People's Republic of China. Lepr Rev. 2004;75(4):348–56.
52. Briden A, Maguire E. An assessment of knowledge and attitudes towards leprosy/Hansen's disease amongst healthcare workers in Guyana. Lepr Rev. 2003;74(2):154–62.
53. Touko A, Kemmegne J, Nyiama T. Perception of lepers by non-lepers in an urban center in Cameroon. Sante. 1996;6(5):269–74.
54. Gittens C. Attitudes towards leprosy in the outpatient population of dermatology clinics in Trinidad. Lepr Rev. 1992;63(2):151–6.
55. Crook N, Ramasubban R, Samy A, Singh B. An educational approach to leprosy control: an evaluation of knowledge, attitudes and practice in two poor localities in Bombay, India. Lepr Rev. 1991;62(4):395–401.

56. Gerochi LN. Knowledge, beliefs and attitudes on leprosy in Iloilo City Proper, Philippines. Southeast Asian J Trop Med Public Health. 1986;17(3):433–6.
57. Stephen T, Selvaraj I, Parameswari PJ. Assessment of knowledge, attitude and practice about leprosy among patients and their families in a rural community in Tamil Nadu. Indian J Lepr. 2014;86(1):7–14.
58. FESO N. Appraisal of the knowledge and attitude of Nigerian nurses toward leprosy. Publ Q Br Lepr Reli Assoc ISSN 0305-7518. :169.
59. Danturty I, Manchala S, Jairaj S. Community based study of knowledge, attitude and practices towards leprosy in an urban slum in Secunderabad, Telangana. JMSCR. 2016;49(11):13527–31.
60. Asnake MK, Ahmed M, Genebo T, Mekebib B. Knowledge and attitude of health professionals in relation to the integration of leprosy control into the general health service in Ethiopia. Int J Lepr other Mycobact Dis. 2000;68(3):299.
61. Urgesa K, Bobosha K, Seyoum B, Geda B, Weldegebreal F, Mihret A, et al. Knowledge of and Attitude Toward Leprosy in a Leprosy Endemic District, Eastern Ethiopia: A Community-Based Study. Risk Manag Healthc Policy. 2020;13:1069.
62. Tesema AA, Beriso M. Assessment of knowledge and attitude of community on leprosy patients in Kuyera Town, West Arsi Zone, Oromia Region Southeast Ethiopia. Hered Genet. 2015;4(4):156.
63. Jain M, Sharma A, Jain V, Virjee K, Singh S. Knowledge and Attitude about Leprosy among Indian Dental Students in Faridabad. J Clin diagnostic Res JCDR. 2016;10(3):ZC48.
64. Mankar MJ, Joshi SM, Velankar DH, Mhatre RK, Nalgundwar AN. A comparative study of the quality of life, knowledge, attitude and belief about leprosy disease among leprosy patients and community members in Shantivan Leprosy Rehabilitation centre, Nere, Maharashtra, India. J Glob Infect Dis. 2011;3(4):378.
65. Barkataki P, Kumar S, Rao PS. Knowledge of and attitudes to leprosy among patients and community members: a comparative study in Uttar Pradesh, India. Lepr Rev. 2006;77:62–8.
66. Kumar R, Singhasivanon P, Sherchand JB, Mahaisavariya P, Kaewkungwal J, Peerapakorn S, et al. Gender difference in socio-epidemiological factors for leprosy in the most hyper-endemic district of Nepal. Nepal Med Coll J NMCJ. 2004;6(2):98–105.
67. Croft RP, Croft RA. Knowledge, attitude and practice regarding leprosy and tuberculosis in Bangladesh. Lepr Rev. 1999;70(1):34–42.
68. Saha G, Mandal NK, Dutta RN. Current Perceptions and Practices (KAP) about Leprosy among Leprosy Patients: A Comparative Study between High Prevalent & Low Prevalent Districts of West Bengal. Indian J Lepr. 2015;87(1):1–16.
69. Ballering A V, Peters RMH, Waltz MM, Arif MA, Mishra CP, van Brakel WH, et al. Community stigma and desired social distance towards people affected by leprosy in Chandauli District, India. Lepr Rev. 2019;90(4):418–32.
70. Mieras L, Singh MK, Manglani PR, Arif MM, Banstola NL, Pandey B, et al. A single-dose of rifampicin to prevent leprosy; quantitative analysis of impact on perception, attitudes and behaviour of persons affected, contacts and community members towards leprosy in India, Nepal and Indonesia. Lepr Rev. 2020;91:314–27.
71. Peters R, Mieras L, Subedi M, Apte H, Koesbardiati T, Banstola NL, et al. A single dose of rifampicin to prevent leprosy: qualitative analysis of perceptions of persons affected, contacts, community members and health professionals towards chemoprophylaxis and the impact on their attitudes in India, Nepal and Indonesia. Lepr Rev. 2018;89(4):335–52.

72. van't Noordende AT, Lisam S, Ruthindartri P, Sadiq A, Singh V, Arifin M, et al. Leprosy perceptions and knowledge in endemic districts in India and Indonesia: differences and commonalities. PLoS Negl Trop Dis. 2021;15(1):e0009031.
73. Rensen C, Bandyopadhyay S, Gopal PK, Van Brakel WH. Measuring leprosy-related stigma–a pilot study to validate a toolkit of instruments. Disabil Rehabil. 2011;33(9):711–9.
74. Peters RMH, Van Brakel WH, Zweekhorst MBM, Damayanti R, Bunders JFG. The cultural validation of two scales to assess social stigma in leprosy. PLoS Negl Trop Dis. 2014;8(11):e3274.
75. Oliveira HX. Adaptação transcultural das escalas de estigma Explanatory Model Interview Catalogue (EMIC) na perspectiva de pessoas acometidas pela hanseníase e da comunidade para o contexto brasileiro [Internet]. Universidade Federal do Ceará; 2018. Available from: http://www.repositorio.ufc.br/bitstream/riufc/39006/1/2018_dis_hxoliveira.pdf
76. Sermrittirong S, Van Brakel WH, Kraipui N, Traithip S, Bunders-Aelen JF. Comparing the perception of community members towards leprosy and tuberculosis stigmatization. Lepr Rev. 2015;86(1):54–61.
77. Sinha A, Kushwaha AS, Kotwal A, Sanghi S, Verma AK. 4 Stigma in leprosy: Miles to go! Indian J Lepr. 2010;82(3):137.
78. Van Brakel WH, Sihombing B, Djarir H, Beise K, Kusumawardhani L, Yulihane R, et al. Disability in people affected by leprosy: the role of impairment, activity, social participation, stigma and discrimination. Glob Health Action. 2012;5(1):18394.
79. Stevelink SAM, Van Brakel WH, Augustine V. Stigma and social participation in Southern India: Differences and commonalities among persons affected by leprosy and persons living with HIV/AIDS. Psychol Health Med. 2011;16(6):695–707.
80. Roosta N, Black DS, Rea TH. A comparison of stigma among patients with leprosy in rural Tanzania and urban United States: a role for public health in dermatology. Int J Dermatol. 2013;52(4):432–40.
81. Weiss MG, Doongaji DR, Siddhartha S, Wypij D, Pathare S, Bhatawdekar M, et al. The explanatory model interview catalogue (EMIC). Framew. 1997;
82. Dadun, Peters RMH, Van Brakel W, Lusli M, Damayanti R, Bunders JFG, et al. Cultural validation of a new instrument to measure leprosy-related stigma: the SARI Stigma Scale. Lepr Rev. 2017;88:23–42.
83. Wong ML. Guest editorial: designing programmes to address stigma in leprosy: issues and challenges. Asia Pacific Disabil Rehabil J. 2004;15:3–12.
84. Cross H. Interventions to address the stigma associated with leprosy: a perspective on the issues. Psychol Health Med. 2006;11(3):367–73.
85. Montano DE, Kasprzyk D. Theory of reasoned action, theory of planned behavior, and the integrated behavioral model. Heal Behav Theory, Res Pract. 2015;70(4):231.
86. Bartholomew LK. Intervention Mapping: A Process for Developing Theory-and Evidence-Based Health Education Programs. Heal Educ Behav. 1998;25(5):545–63.
87. Lynch P. A new face for an old disease: some reflections on the role of the media in Nepal's first National Leprosy Elimination Campaign. Lepr Rev. 2000;71(1):62–70.

PART 2

The impact of leprosy on marital and family life

CHAPTER 5

The impact of leprosy on marital relationships and sexual health among married women in Eastern Nepal

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Abstract

Background

Leprosy is one of the most stigmatized diseases known today. The stigma surrounding leprosy can be a major burden and affects many dimensions of a person's life, including intimate relationships. We aimed to investigate the experiences of women affected by leprosy regarding marital life and sexuality, comparing these to the experiences of women with other physical disabilities and to those of able-bodied women in South-East Nepal.

Methods

This study used a qualitative approach and a cross-sectional, nonrandom survey design. Thirty women underwent in-depth interviews about their marital and sexual relationship by means of a semi-structured interview guide. These thirty women included ten women affected by leprosy, ten women with other physical disabilities, and ten able-bodied women living in South-East Nepal.

Results

We found that many women faced violence and abuse in their marriages. However, women affected by leprosy appeared to face more problems with regard to their marital and sexual relationships than women with physical disabilities and able-bodied women. Some of these related to the fear of leprosy.

Conclusions

Further research is recommended to investigate the extent of this problem and ways to ameliorate the situation of the affected women. Education and counselling at diagnosis may help prevent many of the problems reported.

Introduction

Leprosy is one of the most stigmatized diseases known today [1–4]. Leprosy patients may face the effects of stigma and different forms of discrimination, such as rejection, abuse, divorce, or loss of employment, leading to reduced self-esteem and loss of respect from their communities [4– 6]. Interpersonal relationships, social status, mobility, and dignity suffer [5, 6] and may cause anxiety, depression, emotional stress, isolation, and suicide or attempted suicide. Multiple studies found women to be more affected by leprosy and its stigma than men [3, 4, 6–9]. Stigma may aggravate existing inequalities due to age, gender, and social class [10]. Often, the social and psychological complications due to leprosy remain even after the medical treatment is finished. The psychosocial consequences a person has to bear after being diagnosed by leprosy are often heavier than the physical consequences that may occur [11, 12].

An important example of a social complication of leprosy is the effect of leprosy on marital relationships. A qualitative study on the psychological needs of men and women with leprosy in South Africa found that one-third of leprosy patients had been abandoned by their spouses [13]. Try found that stigma of leprosy has an effect on marriage. She states that “it is clear that in Maithili and Nepali culture, it is undesirable to marry someone who has been or is affected by leprosy” and “the prevalence of visual signs of leprosy is important [this time in] affecting the opinion of prospective partners in arranged marriages” [7]. Adhikari and colleagues, who did a cross-sectional study in Nepal among community members unaffected by leprosy, found that 48% of the community members thought that people affected by leprosy would encounter marital problems [12]. However, not much is known about the nature of the effects of leprosy on ongoing marriages and even less is known about the effects on sexual relationships and perceptions of sexuality and reproductive health.

In Nepal, as in most cultures in the Global South, marriage is considered very important [14]. Definitions of marriage may vary among Nepal’s 60 ethnic groups [15]. According to Lamichhane and colleagues, however, overall “women are expected to play a subordinate, submissive, and more conservative gender role in marital relationships especially in rural areas” [16]. In particular in rural areas, early marriage is quite common [15, 17–20]. Even though there is a slow shift from arranged marriages to love marriages, arranged marriages are still predominant in Nepal and individual choices are subordinate to relationships and agreements between families [16–18, 21]. In case of arranged marriages, especially when they happen at a young age, premarital romantic relationships often do not happen. For the majority of Nepali women, the onset of sexual activity occurs within marriage [18]. Closely linked to the common practice of arranged marriages, marriage in Nepal occurs at a rather young age, with a median age at first marriage of 16.5 years for women born in the late 1970s [17].

Sexuality is closely linked to marriage and has long been avoided as a study topic because of the taboo associated with discussing sexuality. For this reason, the topic was difficult to address [22]. To our knowledge, no research has been done on the effects of leprosy on sexual relationships and perceptions of sexual health. However, a number of studies have investigated the effects of disability on sexual relationships. For people with a disability, sexuality is often not recognized as a legitimate form of pleasure and an expression of love [22]. McCabe and Taleporos, who studied predominantly people with a spinal cord injury, cerebral palsy, and acquired brain injury, found that having a physical disability leads to increased negative feelings, including a belief of being less sexually attractive than people without a disability and the feeling that people with a disability are limited in expressing their sexuality [23]. The latter are generally less satisfied with their romantic relationships than their able-bodied peers [24, 25]. People with a disability are sometimes viewed as asexual [22, 26, 27]. It is often inaccurately thought that people with disabilities lack the desire, ability, and capacity to be sexually active [27, 28]. According to Nosek and colleagues, having a mobility-related disability limits the opportunity for sexual activity. They state that “women with disabilities reported significantly lower levels of sexual activity, sexual response, and satisfaction with their sex lives” [25]. A lack of privacy, dependence on others for care, and inaccessibility to homes and meeting places also make it more difficult for people with a disability to maintain sexual relationships [29].

Infectious diseases still constitute a significant proportion of the total disease burden in Nepal. Leprosy is one of the neglected tropical diseases endemic in the country. In 2014, 3,046 new cases were registered [30]. The distribution of leprosy is not equal in Nepal. The Terai districts account for over 83% of cases [31]. In addition to those currently on treatment, many thousands of people live with residual leprosy-related disabilities, many of which are aggravated by social stigma which is still very strong.

The scanty evidence that is available indicates that leprosy may severely affect relationships, to the extent that even divorce is not uncommon in marriages in which one spouse develops leprosy [5, 13]. Given the fear of contagion that surrounds leprosy, it is likely that divorce is only the tip of the iceberg and that many problems in marital and sexual relationships go unnoticed. This study aimed to investigate the experiences of women affected by leprosy regarding marital life and sexuality, comparing these to the experiences of women with other physical disabilities and to those of able-bodied women in South-East Nepal. This study focused on women, because women are often more severely affected by leprosy and its stigma than men [3, 4, 6–9].

Methods

Study design

This study used a cross-sectional, nonrandom survey design with a qualitative approach.

Study site

This study was conducted in the Eastern Terai region of Nepal.

Study population

Three groups of people were included in the study:

1. Women affected by leprosy, with and without visible impairments.
2. Non-leprosy-affected women with visible impairments.
3. Able-bodied, healthy women.

The first group consisted of women who had completed their leprosy treatment and women who still received treatment. Of the women who had a disability resulting from leprosy, the disabilities were graded using the grading system of the WHO, which grades impairments in eyes, hands, and feet [32]. Each hand, foot, and eye (left and right) is assessed and graded on its own. Either the maximum grade or the sum of the six grades is used as indicator of the severity of impairment. In this study, the women affected by leprosy had to have at least a grade 1 impairment. Both women with grade 1 and women with grade 2 impairments were included, to explore both problems resulting from physical appearance and problems resulting from the diagnosis of leprosy itself.

The second group consisted of women with a visible physical impairment. Included were women with an impairment obvious to the community based on appearance and/or those with limited functioning. Examples of visible physical impairments include people with neurological impairments requiring mobility aids and people with severe burn scars. Both women with congenital impairments and women who acquired an impairment later in life were included.

The third group served as “control” group. Able-bodied, healthy women were interviewed, to try and distinguish issues resulting from cultural practices, and lack of knowledge or awareness, from those caused by disability or leprosy.

Study sample

In total, 10 women affected by leprosy, 10 women with a visible physical impairment, and 10 able-bodied women were included. Of the women affected by leprosy, 6 had a grade 1 impairment and 4 had a grade 2 impairment. All participants lived in the Eastern Terai region of Nepal, in Jhapa, Morang, Saptari, or Sunsari districts. If women indicated a need for support or counselling, they were referred to the Biratnagar Leprosy Referral Centre.

Sampling methods

Because we wanted to interview women with specific characteristics, participants were selected using purposive sampling. All women included in the study had to be married and had to be between the ages of 18 and 50. Excluded were women with a mental illness that interfered with their ability to undergo an in-depth interview, widowed women, and women whose husbands did not know they have or have had leprosy. The participants were contacted through the Netherlands Leprosy Relief (NLR) network in the Eastern Development Region of Nepal and through local health posts. Local health posts in the study area were visited to check whether there were leprosy-affected women that met the inclusion criteria. The records of the NLR-supported referral clinic in Biratnagar were also checked. Both the women with disabilities and the able-bodied women were selected based on their similarity to the leprosy-affected women, mainly in age and living area. Women with disabilities were identified through local Disabled People's Organisations (DPOs).

Data collection

Cross-sectional data were obtained from in-depth interviews. Participants underwent in-depth interviews about their marital and sexual relationship by means of semi structured interviews. Data were collected between March and June 2014. The interview guide used consisted of four themes: sense of self, marital relationship, knowledge and awareness of sexual and reproductive health, and sexual relationship. The interview guide was developed based on a literature review and on discussions with leprosy specialists. It was translated in Nepali. The translation was thoroughly checked by translating the instrument back into English. Some questions were translated multiple times using different interpreters, to ensure that the meaning of the original English version was retained.

Taking into account the sensitivity of the topic, participants were interviewed by a local, married female interpreter in their home, or at a private, safe space near their home. The interpreter had several years of experience working for NLR in the area of research. The interpreter had experience with and knowledge of working with people affected by leprosy and qualitative data collection. To minimise interobserver variation, all interviews were conducted by the same inter-viewer. The interview guide and the interview technique were tested by means of pilot interviews. Prior to the pilot interviews, the interpreter received an interview training of several days in which she did role plays and was provided with feedback by the researcher. The researcher herself was not present during the interviews to prevent discomfort on the side of the interviewee. Interviews of 30–70 minutes were conducted in the local language and were audio recorded.

Data analysis

The recordings were translated, transcribed, and analysed using open coding and content analysis. The interviews were transcribed in English by the interpreter and discussed with the researcher to help put issues in perspective and context. The software programme "MAXQDA" was used to assist in analysing the data. All information in the transcripts was coded by the lead author (AvtN). Open, inductive coding was done in MAXQDA, where similar phrases with recurring themes were coded. All codes with supporting quotes were then clustered together in different tables, ordered by subquestion to get an overview of responses and to identify connection between codes and themes.

Ethical considerations

Prior to the in-depth interviews, participants were fully informed about the nature and objective of the study and of confidentiality of the data. Written consent for participation in the study was obtained from each participant. Ethical approval was sought and obtained from the Nepal Health and Research Council.

Results

Characteristics of the study sample

Thirty women were included. The mean age was 35 years for the women affected by leprosy (range: 22–50 years), 36 for the women with physical impairments (range: 24–50 years), and 36 for the control group (range: 24–50 years). Assessment of impairments using the WHO's grading system for disabilities resulting from leprosy classified six women as grade 1 and four as grade 2. Three women still received treatment for leprosy, whereas seven women were released from treatment.

All the women (n = 30) who participated in this study were still with their husbands. Ethnicity was categorized into four groups: (i) Brahmin/Chettri, (ii) Dalit, (iii) Tribal, and Other. Brahmin/Chettri accounted for the biggest group (n = 16). The majority of women were Hindu (n = 26) and lived in rural areas (n = 21). Most marriages were arranged (n = 20). Love marriages occurred mostly among the women with physical disabilities, with five women having a love marriage. Except for one participant who had upper back problems, all participants with a visible physical disability had impairments related to their feet and legs, observable in walking. Except for three women, all had at least one child. Three women, one in each group, were visibly pregnant at the time of the interviews. More than half of the women (n = 17) indicated they did not work outside the home. No demographic data was collected about the husbands of the women included in the study.

Additional information concerning leprosy-affected women

For five women affected by leprosy, all contacts, husband, neighbours, and relatives, knew they had leprosy, for two, only the husband knew, and for three, only a few people who were very close knew they were affected by leprosy. It seemed that the cause of their disease was not always well known. Four out of ten women did not seem to know the real cause of their disease. They also had misconceptions about the transmission of the disease. Once their treatment was finished, these misconceptions no longer applied. One woman told us,

> ...*Before there were problems, I did not give them [family] food which I had taken. I was worried that it would transfer to them*....
>
> (Woman affected by leprosy, age 33)

Another woman said,

> ...*My husband is afraid that it transmits through respiration, so he does not want to tongue kiss for seven months*....
>
> (Woman affected by leprosy, age 22)

In the quotes presented, women mostly referred to their situation *after* contracting leprosy.

Results regarding marriage, sexual relationships, and sex education applicable to all women

Most of the thirty women who were interviewed (n = 26/30) indicated that being married is important in their community and to themselves also. Sexual relationship, an important part of marriage, seemed to be of mixed importance. Six women from the control group indicated that sex is an important part of their relationship. For the women with a disability, five women considered it important, five did not. Six of the women affected by leprosy said they did not find sex important. Except for two women, all women indicated that sex is important to their husbands. Some women (n = 4/30) thought that their opinion on whether sex is important is not really of importance. According to them, what *they* think is important does not always matter. They are supposed to be ready whenever their husbands are ready, as the following quotes illustrate:

> ...*Yes, it is also important for me, but our importance has no value. We cannot express our feelings even with our husband*....
>
> (Woman affected by leprosy, age 26)

> ...*It is not necessary how important it is for us because whenever our husband is ready we should be ready*....
>
> (Woman affected by leprosy, age 50)

> ...*It is not so important for me because of my condition, but I used to be ready for my husband and I do not mind, after all he is my husband*....
>
> (Woman with physical disability, age 48)

Most women (n = 24/30) did not receive sexual education in school or through a health post, or at least not before marriage. Only six women said they received sexual education in school, mostly mentioning grade eight, nine, or ten. All of the women who mentioned they had had sexual education had received higher or secondary education. Other women mentioned they received some form of sexual education through the health post or from a neighbour or relative. Many women (n = 15) mentioned receiving sexual education through TV or radio.

Factors affecting the marital relationship of women

Positive factors

Several factors may influence the marital relationship of the women interviewed. Factors reported to have a positive effect on their marital relationship were love, harmony, and understanding each other, money or property, and sex. Two women said:

> ...*I think sex is the ultimate factor for a couple to stay happy*....
> (Woman with physical disability, age 43)

> ...*The most important is trust, love and understanding*....
> (Woman affected by leprosy, age 32)

Negative factors

Factors reported to have a negative influence on marriage are the (alcohol) drinking habit of the husband, a negative attitude of family members or others towards the woman, misunderstanding between husband and wife, and an unsupportive husband. Furthermore, two women with leprosy indicated that they felt that people were more distant since they knew they are affected by leprosy. Some examples are given below:

...*I used to stay with my father and mother in law in their house. My sister in law was very rude, she used to come to the house as she was married and tell unnecessary things to my husband. My husband used to listen to her and be angry with me*....
(Woman from control group, age 32)

...*When I was diagnosed with leprosy I felt that my husband's behaviour had changed, he did not share anything with me and he pretended to be busy with work. But actually he was trying to be far away from me*.... (
Woman affected by leprosy, age 33)

...*Before there were problems, when my father and mother-in-law knew about my disease, they hesitated to talk to me and come near me*....
(Woman affected by leprosy, age 33)

However, the alcohol problem of the husband, if present, seemed to be the biggest problem. This was mentioned by all three groups, but was most frequently experienced by the women affected by leprosy. Twelve out of the thirty women had a husband with an alcohol problem. Of these women, three women were in the control group, four women had a disability, and five women were affected by leprosy.

When asked about the possibility of the husband of someone with either a disability or leprosy marrying and taking on a second wife, six women said they had never heard of this, 17 women said they had heard of this but had not experienced this themselves, four women indicated that only certain types of people do this, and two women affected by leprosy said they had experienced this themselves. The husband of one of these women had taken a second wife, because of her leprosy. Another husband sent his wife away to her parents' house and then had several affairs. Some women with a disability pointed out the difference between men and women when it comes to remaining faithful to their marriage. One woman said, about taking on a second wife,

> ...*If the husband has a disability then they marry a common woman or if he becomes disabled after marriage, then his wife stays with him, but if the woman is disabled then she cannot marry a common man and if she becomes disabled then the husband brings another wife*....
>
> (Woman with physical disability, age 32)

> ...*I feel that, if the problem that my husband has had had happened to me, my husband would have brought a second wife. But I am a woman so I cannot do so and I love him. Sometimes I used to be angry with myself for my condition*....
>
> (Woman with physical disability, age 32)

Factors affecting the sexual relationship of married women in Nepal

Positive factors

There are positive and negative factors that influence the sexual relationship of women. Factors that may have a positive influence include loving each other and being emotionally engaged, understanding each other, obeying the husband and/or giving priority to him, and the husband not drinking alcohol. Two women said,

> ...*If sexual intercourse is a mutual understanding then we can get pleasure*....
>
> (Woman from control group, age 32)

> ...*Love helps for the good sexual relationship with my husband*....
>
> (Woman affected by leprosy, age 35)

Negative factors

Factors that may have a negative influence on the sexual relationship of married women include the alcohol problem of the husband, being forced by the husband to have sex, disagreeing with the husband, and reduced interest in having sex on the side of the wife. Only 11 women indicated not having any sexual problems. The husband drinking alcohol and, with that, sexual abuse were most often mentioned. One woman said about this,

> ...*He has the bad habit of drinking alcohol. He wants every time when he is drunk. I feel so irritated but what can I do, we think of our husband as God and we should obey him*....
>
> (Woman affected by leprosy, age 26)

About being sexually abused, two women said,

> ...*When I do not want to have sexual intercourse, my husband forces me. He scolds me "I used to earn money, bring food for you all but you do not want [to have sex], then get out of the house!" Sometimes he raised a hand on me. So I have to be near and close and have sex with him*....
>
> (Woman affected by leprosy, age 32)

> ...*He never asks about my health and forces me to have intercourse. I feel like I am a doll to him*....
>
> (Woman with physical disability, age 33)

Of the women facing sexual abuse, three women were in the control group, three women had a disability, and five were affected by leprosy. The only two Muslim women included in this study both faced sexual abuse. Furthermore, three of the four women who had a leprosy-related grade 2 impairment were being sexually abused by their husbands. In addition, of the women who had a husband with an alcohol problem (n = 12), all but one faced sexual abuse by their husbands, as illustrated below:

> ...*There were problems, he used to drink alcohol and come near me and force me to have sexual intercourse*....
>
> (Woman affected by leprosy, age 41)

> ...*When he drinks alcohol and comes I feel irritated and he forces me to have sexual intercourse with him*....
>
> (Woman from control group, age 32)

Sexual abuse and alcohol abuse seemed to go hand-in-hand with violence. Many (n = 7/10) women were either beaten or threatened to be beaten if they do not obey. This also became clear when talking about what happens if they do not agree with their husbands:

> ...*If I refuse him he scolds me and raises a hand on me*....
>
> (Woman with physical disability, age 33)

> ...*If the husband likes to have sex, then we have to give, if not he certainly beats me*....
>
> (Woman from control group, age 50)

> ...*I have to give him everything he wants even when I am not feeling well, because he gets angry if I refuse to give. He warns me that he'll have sexual pleasure with another girl if I cannot give pleasure*....
>
> (Woman from control group, age 28)

Five women affected by leprosy, of whom three also faced sexual abuse, faced additional problems while receiving treatment or when they were first diagnosed. These problems disappeared later on and were not experienced at the time of the interviews. They included having no intercourse at all due to fear of transmission of the disease, experiencing more distance and sometimes sleeping in separate beds while taking medicine:

> ...*At first when he knew that I was affected by leprosy he did not sleep with me. He used to scold me for no reason. Once he came to the Biratnagar clinic with me, he asked the doctor about the sexual relationship. He was told that it does not transfer to him so he started having sexual intercourse with me again*....
>
> (Woman affected by leprosy, age 32)

One woman did not want to talk about the problems she had before,

> ...*Before I had very bad problems, but now there is no effect, I do not want to remember the past and talk about that*....
>
> (Woman affected by leprosy, age 35)

Discussion

We found that many women experience marital problems and/or sexual abuse, regardless of their leprosy or disability status. In addition, we found clear indications that leprosy may influence the marital and sexual relationship of married women in various ways. This included significant problems during treatment, which is often a full year, such as having no intercourse at all due to fear of contagion, experiencing more distance from close others, and husband and wife sleeping in separate beds. Others were being abandoned or sexually abused by the husband even after treatment. These problems may be due to the negative attitudes surrounding the diagnosis of leprosy. Other studies found stigma to have negative consequences for persons affected by leprosy, leading to discrimination [4–6, 33], problems in interpersonal relationships, and problems with social status [5, 6].

Women affected by leprosy seem to face most problems when first diagnosed or while receiving treatment. The above problems may have been caused or aggravated by the fact that almost half of the women did not seem to know the cause of their disease and how leprosy is transmitted. This may have been true for their spouses also. Thilakavathi and colleagues, who conducted in-depth interviews with 72 leprosy-affected men and women, of whom 48 were married, found that a few participants did not sleep in the same room as their spouses, but they did not elaborate on this [34]. They also found that most of their interviewees lacked basic knowledge on the transmission and cause of leprosy.

Leprosy-affected women are sometimes abandoned by their husbands. This happened to two women with grade 2 impairments due to leprosy in our study. Qualitative evidence suggests that women are more likely to be deserted by their spouses than men, but conclusive evidence is not yet available [7]. Research in South-East Nepal found that, of the nine men and ten women interviewed, three husbands had left their leprosy-affected wife and one wife had left her leprosy-affected husband [7]. The reason for the spouse leaving was the other spouse's diagnosis of leprosy. The separation occurred a few months after diagnosis. These findings are similar to those in our study. A study in South Africa showed that "of 23 married subjects, 9 men and 7 women had been deserted by their marriage partners because of leprosy" [13]. We cannot tell whether the frequency of divorce found by Scott [13], which is much higher than in our study, is due to sampling error, bias in the samples, or actual cultural differences, since the present study was only designed to explore the impact of leprosy, not to determine the prevalence of marital problems or divorce due to leprosy. Our findings indicate that divorce is only the tip of the iceberg of marital problems that may be due to leprosy or other causes.

An important finding is the high frequency of alcohol abuse among the husbands of the participants. This occurred in all three groups and therefore points to a more structural phenomenon in society. A larger survey using random sampling will have to confirm whether the greater frequency of alcohol abuse among the husbands of leprosy-affected women compared to the other women included in this study is real, or a result of sampling error. Jhingan and colleagues looked at alcohol dependence in Dharan, in Eastern Nepal, and found the prevalence of alcohol dependence to be 25.8% [35]. They found dependence to increase with age, peaking with 41% in the age group 45–54, compared to 10.7% in the 15–24 age group. Alcohol dependence was more than twice as common in men as in women. The findings of the present study support the findings of Jhingan and colleagues [35]. The severity of alcohol abuse in the present study is not known. Several studies found the risk of sexual abuse and violence towards women to increase when husbands are drunk or are alcoholics [36–40]. These studies were also conducted in developing countries and highlight the important role of alcohol use in sexual abuse [36–40]. The present study seems to confirm the relationship between alcohol abuse and sexual abuse: most of the women who had a husband with an alcohol problem experienced sexual abuse and all husbands who sexually abused their wives reportedly had an alcohol problem. We did not find any literature on the relationship between alcohol abuse and sexual abuse when the spouse has an impairment.

Sexual abuse by the husband occurred in all groups, but the frequency was higher among women affected by leprosy. Again, this may be due to sampling error and/or bias in subject selection. A larger follow-up study using random sampling will need to clarify this. Another study assessed the occurrence of violence against young married women aged 15–24 years in rural Nepal [16]. As many as 53% reported having experienced some form of violence in their lifetime and 46% reported experiencing sexual violence. No or little interspousal communication and low autonomy of women were associated with violence against women [16]. Other studies that investigated sexual violence against young married women found a similar prevalence (49%) [18, 41]. Lamichhane and colleagues associated women's lower status in family and society with violence against women, particularly young women in rural Nepal [16]. Also Pradhananga and Shrestha [42] and Puri and colleagues [18] stress the low status of women in Nepal. Deepak and colleagues, who looked at violence and sexual violence against persons with disabilities in India, found that 14% of their 146 participants reported experiences of sexual violence during the previous 12 months [43]. The presence of visible impairments among leprosy patients and its influence on acceptance by others have been highlighted by other studies [44, 45]. Kopparty, who looked at coping strategies of 500 families who had a leprosy-affected family member with and without disfigurement, found that "the proportion of families having patients with deformities facing problems was ten times higher (57%) than those having patients with no deformities (5.7%)" [45]. It is therefore not unlikely that women with visible impairments would experience more discrimination and abuse than leprosy-affected women without visible impairments. Their position in society may be low, possibly aggravated by alcohol abuse of the husband. This in turn may lead to sexual abuse. Furthermore, Puri and colleagues found that 8 out of 15 women who refused to have intercourse with their husbands were beaten [41]. Being beaten or threatened with violence when not obeying their husbands was also reported by women in the present study.

Our study showed that a husband often has power over his wife and that wives are expected to obey their husband or otherwise may be expected to be punished. Feelings were often not shared, because women felt their feelings and desires were not valued. These findings fit with the description of prevailing attitudes towards women described by Regmi and colleagues [46]. They stated that, in Nepal, "unequal power relations and lack of autonomy characterise the situation of married young women in many settings, the autonomy of married young women is particularly constrained" and "gender norms stress male entitlement to sex, even if forced within marriage." Certain social roles are expected, and most of the women's roles revolve around the household [7].

An important finding was that most women had not received sexual education, or at least not before marriage. The few women who had sexual education before marriage received this sometime between grades 8 and 10 in school. Regmi and colleagues reported that "there are major gaps in receiving information, services, and skills on sexual and reproductive health issues" [46]. The Government of Nepal has introduced sexual and reproductive health education in public schools for grades six to ten and in university curricula from 1998 onwards [46]. Regmi and colleagues assert that, in reality, young people do not have good access to appropriate information on sexual and reproductive health issues. This fits with our finding that several women who were in their twenties and who had secondary education or more indicated that they did not receive sexual education, despite the fact that a sexual and reproductive health education programme had already been introduced when they were in school.

The current findings show that knowledge about leprosy and the relation between leprosy and marriage and sexual health should be addressed preventively whenever someone is diagnosed with leprosy. If at all possible, the spouse and possibly the in-laws of any newly diagnosed married patient should be included in such counselling efforts. Materials addressing these issues should be developed and should be made available for distribution in primary health centres and other health facilities where persons affected by leprosy are diagnosed and treated.

However, it was evident in our study that within-marriage violence and sexual abuse of women, aggravated by alcohol abuse of the husband, occurred in all groups regardless of health or disability status. Sexual and reproductive health, freedom from violence, and freedom from discrimination are fundamental human rights that were systematically violated in the lives of many of the women interviewed. Therefore, interventions to improve sexual health and safety of married women should be designed, tested, and implemented as a matter of urgency.

Limitations of study

The first limitation is the nonrandom sampling and small study size, as mentioned above. This, together with the specific geographic location of the present study, means that the results of the study cannot be generalized to the whole study population or beyond. Furthermore, due to time constraints, the interviews only included women. If men would have been included also, a more complete insight of the impact of leprosy on marital and sexual relationships could have been given. Another limitation was the use of an interpreter. Translating the interviews from Nepali to English may have introduced some mistakes or misinterpretations, since not all words could be translated literally.

Further Research

The current study indicates that leprosy may influence the marital and sexual relationship of women in Nepal in several ways. Additional research is needed to gain more insight in the underlying reasons. Knowing more about the factors that influence marital and sexual relationship may help patients as well as health workers deal with marital and sexual problems and may enhance their ability to anticipate and prevent problems. It will also inform policy and interventions to reduce within-marriage sexual abuse and violence. Raising awareness of health workers concerning the risk of marital problems following a diagnosis of leprosy is essential. Simple educational materials with facts about leprosy, explaining the absence of risk of transmission once someone is being treated, would help health workers discuss these very important issues with their patients, preferably together with their spouses. On a wider scale, interventions to improve sexual health and safety of married women should be implemented as a matter of urgency. Implementation research should investigate the appropriate format and manner in which this can be done. A larger study with a random sample is needed to determine the extent of the problems identified and the added risk of being leprosy-affected or having a disability.

Conclusions

Many women in our study experienced marital problems and/or sexual abuse, regardless of their leprosy or disability status. Fundamental human rights such as the rights to sexual and reproductive health, freedom from violence, and freedom from discrimination are systematically violated in the lives of many of the women interviewed.

Women affected by leprosy faced additional problems, related to fear of the disease, negative attitudes, and discrimination on account of leprosy.

Knowledge on the cause and transmission of leprosy was still lacking among leprosy-affected women and their community members. Appropriate preventive and educational measures should be designed and tested to address these issues. Lastly, women appear to have insufficient access to sexual education, despite programmes on sexual health education in schools and elsewhere. This should be addressed by the appropriate authorities.

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References

1. H. Cross and R. Choudhary, “STEP: an intervention to address the issue of stigma related to leprosy in southern Nepal,” *Leprosy Review*, vol. 76, no. 4, pp. 316–324, 2005.
2. H. Kaur and W. Van Brakel, “Dehabilitation of leprosy affected people-a study on leprosy affected beggars,” *Leprosy Review*, vol. 73, no. 4, pp. 346–355, 2002.
3. M. J. Mankar, S. M. Joshi, D. H. Velankar, R. K. Mhatre, and A. N. Nalgundwar, “A comparative study of the quality of life, knowledge, attitude and belief about leprosy disease among leprosy patients and community members in Shantivan Leprosy Rehabilitation centre, Nere, Maharashtra, India,” *Journal of Global Infectious Diseases*, vol. 3, no. 4, pp. 378–382, 2011.
4. J. Rafferty, “Curing the stigma of leprosy,”*Leprosy Review*, vol. 76, no. 2, pp. 119–126, 2005.
5. W. H. Van Brakel, “Measuring leprosy stigma-a preliminary review of the leprosy literature,” *International Journal of Leprosy and Other Mycobacterial Diseases*, vol. 71, no. 3, pp. 190–197, 2003.
6. G. P. Singh, “Psychosocial aspects of Hansen’s disease (leprosy),” *Indian Dermatol Online*, vol. 3, no. 3, pp. 166–170, 2012.
7. L. Try, “Gendered experiences: marriage and the stigma of leprosy,” *Asia Pacific Disability Rehabilitation Journal*, vol. 17, no. 2, pp. 45–46, 2006.
8. C. M. Varkevisser, P. Lever, O. Alubo et al., “Gender and leprosy: case studies in Indonesia, Nigeria, Nepal and Brazil,” *Leprosy Review*, vol. 80, no. 1, pp. 65–76, 2009.
9. C. Vlassof f, S. Khot, and S. Rao, “Double jeopardy: women and leprosy in India,” *World Health Statistics Quarterly*, vol. 49, no. 2, pp. 120–126, 1996.
10. M. L. Heijnders, “The dynamics of stigma in leprosy,” *The Inter-national Journal of Leprosy and Other Mycobacterial Diseases*, vol. 72, no. 4, pp. 437–447, 2004.
11. B. Adhikari, N. Kaehler, S. Raut, K. Gyanwali, and S. R. Chapman, “Stigma in leprosy: a qualitative study of Leprosy affected patients at green pastures Hospital, western region of Nepal,” *Journal of Health Research*, vol. 27, no. 5, pp. 295–300, 2013.
12. B. Adhikari, K. Shrestha, N. Kaehler, S. Raut, and S. R. Chap-man, “Community attitudes towards leprosy affected persons in Pokhara municipality of western Nepal,” *Journal of Nepal Health Research Council*, vol. 11, no. 25, pp. 264–268, 2013.
13. J. Scott, “The psychosocial needs of leprosy patients,” *Leprosy Review*, vol. 71, no. 4, pp. 486–491, 2000.
14. ESCAP, *Hidden Sisters: Women and Girls with Disabilities in the Asia-Pacific Region*, Social Development Division, United Nations ESCAP, 1997.
15. M. K. Choe, S. Thapa, and V. Mishra, “Early marriage and early motherhood in Nepal,” *Journal of Biosocial Science*, vol. 37, no. 2, pp. 143–162, 2005.
16. P. Lamichhane, M. Puri, J. Tamang, and B. Dulal, “Women’s status and violence against young married women in rural Nepal,” *BMC Women’s Health*, vol. 11, no. 1, article 19, 2011.
17. M. Caltabiano and M. Castiglioni, “Changing family formation in Nepal: marriage, cohabitation and first sexual intercourse,” *International Family Planning Perspectives*, vol. 34, no. 1, pp. 30– 39, 2008.
18. M. Puri, I. Shah, and J. Tamang, “Exploring the nature and reasons for sexual violence within marriage among young women in Nepal,” *Journal of Interpersonal Violence*, vol. 25, no. 10, pp. 1873–1892, 2010.

19. S. Singh, "Adolescent childbearing in developing countries: a global review," *Studies in Family Planning*, vol. 29, no. 2, pp. 117– 136, 1998.
20. S. Singh and R. Samara, "Early marriage among women in developing countries," *International Family Planning Perspec-tives*, vol. 22, no. 4, pp. 148–175, 1996.
21. D. J. Ghimire, W. G. Axinn, S. T. Yabiku, and A. Thornton, "Social change, premarital nonfamily experience, and spouse choice in an arranged marriage society," *American Journal of Sociology*, vol. 111, no. 4, pp. 1181–1218, 2006.
22. S. Esmail, K. Darry, A. Walter, and H. Knupp, "Attitudes and perceptions towards disability and sexuality," *Disability and Rehabilitation*, vol. 32, no. 14, pp. 1148–1155, 2010.
23. M. P. McCabe and G. Taleporos, "Sexual esteem, sexual sat-isfaction, and sexual behavior among people with physical disability," *Archives of Sexual Behavior*, vol. 32, no. 4, pp. 359– 369, 2003.
24. A. L. Janus, "Disability and the transition to adulthood," *Social Forces*, vol. 88, no. 1, pp. 99–120, 2009.
25. M. A. Nosek, C. Howland, D. H. Rintala, M. E. Young, and G. F. Chanpong, "National study of women with physical disabilities: final report," *Sexuality and Disability*, vol. 19, no. 1, pp. 5–40, 2001.
26. V. Kallianes and P. Rubenfeld, "Disabled women and reproduc-tive rights," *Disability and Society*, vol. 12, no. 2, pp. 203–221, 1997.
27. M. S. Milligan and A. H. Neufeldt, "The myth of asexuality: a survey of social and empirical evidence," *Sexuality and Disability*, vol. 19, no. 2, pp. 91–109, 2001.
28. M. S. Tepper, "Sexuality and disability: the missing discourse of pleasure," *Sexuality and Disability*, vol. 18, no. 4, pp. 283–290, 2000.
29. G. Taleporos and M. P. McCabe, "Relationships, sexuality and adjustment among people with physical disability," *Sexual and Relationship Therapy*, vol. 18, no. 1, pp. 25–43, 2003.
30. World Health Organization, "Weekly epidemiological record," *World Health Organization*, vol. 90, no. 36, pp. 461–476, 2015, http://www.who.int/wer/2015/wer9036/en/.
31. A. K. Jha, "Leprosy: before and after elimination from Nepal," *Kathmandu University Medical Journal*, vol. 10, no. 37, pp. 1–2, 2012.
32. World Health Organization, *A Guide to Leprosy Control*, World Health Organization, Geneva, Switzerland, 2nd edition, 1988, http://whqlibdoc.who.int/publications/1988/9241542233 %28 part1%29.pdf.
33. I. Schuller, W. H. Van Brakel, I. Van Der Vliet et al., "T he way women experience disabilities and especially disabilities related to leprosy in rural areas in South Sulawesi, Indonesia," *Asia Pacific Disability Rehabilitation Journal*, vol. 21, no. 1, pp. 60–70, 2010.
34. S. Thilakavathi, P. Manickam, and S. M. Mehendale, "Awareness, social acceptance and community views on leprosy and its relevance for leprosy control, Tamil Nadu," *Indian Journal of Leprosy*, vol. 84, no. 3, pp. 233–240, 2012.
35. H. P. Jhingan, P. Shyangwa, A. Sharma, K. M. R. Prasad, and S. K. Khandelwal, "Prevalence of alcohol dependence in a town in Nepal as assessed by the CAGE questionnaire," *Addiction*, vol. 98, no. 3, pp. 339–343, 2003.
36. K. L. Hof fman, D. H. Demo, and J. N. Edwards, "Physical wife abuse in a non-western society: an integrated theoretical approach," *Journal of Marriage and the Family*, vol. 56, no. 1, pp. 131–146, 1994.

37. M. A. Koenig, S. Ahmed, M. B. Hossain, and A. B. M. K. Alam Mozumder, “Women’s status and domestic violence in rural Bangladesh: individual- and community-level effects,” *Demography*, vol. 40, no. 2, pp. 269–288, 2003.
38. M. A. Koenig, T. Lutalo, F. Zhao et al., “Coercive sex in rural Uganda: prevalence and associated risk factors,” *Social Science and Medicine*, vol. 58, no. 4, pp. 787–798, 2004.
39. W. L. Paris, T. Wang, E. O. Laumann, S. Pan, and Y. Luo, “Intimate partner violence in China: national prevalence, risk factors and associated health problems,” *International Family Planning Perspectives*, vol. 30, no. 4, pp. 174–181, 2004.
40. V. Rao, “Wife-beating in rural South India: a qualitative and econometric analysis,” *Social Science and Medicine*, vol. 44, no. 8, pp. 1169–1179, 1997.
41. M. Puri, J. Tamang, and I. Shah, “Suffering in silence: con-sequences of sexual violence within marriage among young women in Nepal,” *BMC Public Health*, vol. 11, no. 1, article 29, 2011.
42. R. B. Pradhananga and P. Shrestha, *Domestic Violence against Women in Nepal: Concept, History and Existing Laws*, 2006, http://www.childtrafficking.com/Docs/domestic violence 0607.pdf.
43. S. Deepak, J. Kumar, B. Santosh et al., “Violence against persons with disabilities in Bidar District, India,” *Disability, CBR & Inclusive Development*, vol. 25, no. 2, pp. 35–53, 2014.
44. A. J. Schwenk, C. Turcu, I. V. Burkov et al., “Problems and coping strategies of families having patients with and without deformities,” *Mathematics Magazine*, vol. 67, no. 2, pp. 145–152, 1994.
45. S. N. M. Kopparty, “Acceptance in the family: a study of deformed and non−deformed leprosy patients in Tamil Nadu, India,” *International Journal of Sociology and Social Policy*, vol. 18, no. 7-8, pp. 83–101, 1998.
46. P. R. Regmi, P. Simkhada, and E. R. Van Teijlingen, “Sexual and reproductive health status among young people in Nepal: opportunities and barriers for sexual health education and services utilization,” *Kathmandu University Medical Journal*, vol. 6, no. 22, pp. 248–256, 2008.

CHAPTER 6

The impact of leprosy, podoconiosis and lymphatic filariasis on family quality of life: a qualitative study in Northwest Ethiopia

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Abstract

Background

Several studies have shown that leprosy, podoconiosis and lymphatic filariasis impact individual quality of life. In contrast, family quality of life has not received as much attention despite evidence that families are also affected. This is especially relevant given the crucial role of the family in most societies around the world. This study looks at the impact of leprosy, podoconiosis and lymphatic filariasis on family quality of life.

Methodology

The study used a cross-sectional design with a qualitative approach. Both semi-structured interviews and focus group discussions were conducted. Participants, persons affected and their family members, were selected by purposive sampling. Data were collected between August and November 2017 in Awi zone, Northwest Ethiopia and analysed by three independent researchers using open, inductive coding and content analysis.

Results

A total of 86 participants were included in this study: 56 participants in the in-depth interviews and 30 participants in the focus group discussions. We found that participation restrictions, reduced productivity and marginalisation were common. In addition, discrimination in the communities occurred often, often extending to family members of persons affected. Divorce and difficulties in finding a spouse were common for persons affected and their family members. Many persons affected reported mental health problems. While most people got social and physical support from their families, there were a few exceptions. In particular, persons with younger children seemed to lack social support. Having to provide for their affected family member sometimes caused stress, school dropouts and an additional workload. Financial problems and loss of livelihood were reported by almost all participants.

Conclusion

This study revealed that leprosy, lymphatic filariasis and podoconiosis have an effect on several dimensions of family quality of life. Many problems reported related to stigma and poverty.

Introduction

Leprosy and lymphatic filariasis are both communicable diseases. Leprosy is caused by *Mycobacterium leprae* and is transmitted by prolonged close contact between untreated leprosy patients and susceptible and genetically predisposed individuals [1,2]. Lymphatic filariasis, sometimes called ‘elephantiasis’, is caused by three nematode worms - *Wuchereria bancrofti, Brugia malayi* and *Brugia timori*. The parasites that cause lymphatic filariasis are transmitted by mosquitoes [3]. Podoconiosis, or non-filarial elephantiasis, is a non-communicable disease that is believed to be caused by chronic exposure to mineral particles in red clay volcanic soil that penetrate the skin and induce an inflammatory reaction in the lymphatic system [4]. Podoconiosis predominantly affects individuals who live and work barefoot on red clay soil [4–6].

Leprosy primarily affects the peripheral nerves and skin. Nerve damage may cause impairments to their sensory, motor and autonomic functions. This may manifest itself in loss of sensation, impairments to the eyes and shortened or deformed fingers and toes [1,2]. Podoconiosis and lymphatic filariasis are both characterised by lymphoedema of the limbs, leading to enlarged legs, male genitals and female breasts [7–9]. Podoconiosis is clinically distinguished from lymphatic filariasis through commonly being ascending and bilateral but asymmetric, while lymphatic filariasis is often unilateral [7,10]. All three diseases may cause both temporary and permanent long-term impairments [1,2,8,9].

Leprosy, lymphatic filariasis and podoconiosis can have a significant social impact. Persons affected often face stigma, discrimination and social participation restrictions such as isolation, barriers to employment, education or marriage [11–20]. Living with a person affected by leprosy, podoconiosis and lymphatic filariasis can have profound effects on all family members. Often family members also experience stigma on account of having an affected person in the family [21–24]. Living with an affected person can impact many aspects of family life, such as family income and the chance of finding a marriage partner for a son or daughter [15–17,25].

Several studies showed leprosy, podoconiosis and lymphatic filariasis to have an impact on quality of life of the person affected [26–31]. In contrast, the quality of life of family members has not received much attention, despite evidence that families are also affected and play a crucial role in most societies around the world [32]. Individual quality of life broadly encompasses an individual’s perception of the ‘goodness’ of multiple aspects of their life, such as mental, physical, role, environment and social functioning [33]. Family quality of life, a natural extension of individual quality of life, is not focused on individuals but rather on all family members in the family unit [34].

Social relationships and social support play a key role in a person's health and mental wellbeing. This is especially true for persons with physical disabilities, who due to restrictions in social participation, often have fewer opportunities for positive exchange within their close social environment [35]. High quality relationships can in turn strengthen mental wellbeing [35].

There have been several studies into the quality of life of persons affected by leprosy, podoconiosis and lymphatic filariasis [26–31,36]. However, to date no studies have been conducted on the impact of these conditions on family quality of life. The current, qualitative, study aimed to investigate how families with a family member affected by leprosy, podoconiosis and lymphatic filariasis experience their family life, what factors influence family quality of life and how having a family member with leprosy, podoconiosis and lymphatic filariasis-related disabilities impacts family quality of life, in the Awi zone, Ethiopia. This study is part of a larger project that aims to develop a family-based approach to support prevention and self-management of leprosy, podoconiosis and lymphatic filariasis-related disabilities in the Ethiopian context.

Methods

Study objectives

For most families, to achieve good family good quality of life all family members have to be "healthy, have a safe place to live, have a stable income, enjoy their lives together, have opportunities to learn and improve, benefit from their community supports and resources, and experience fulfilling social relationships with others" [37]. Family relations play an important role in the perceptions of individuals, and drives their behaviour, which is important for all families [38], including those with members affected by leprosy, podoconiosis and lymphatic filariasis.

The objective of this study was to explore the quality of life of families with a family member affected by leprosy, podoconiosis and lymphatic filariasis. Based on this exploration, the study aimed to develop a family-based approach to support prevention and self-management of leprosy, podoconiosis and lymphatic filariasis-related disabilities in the Ethiopian context. The ultimate aim of this approach is to improve the lives of the families involved in the study.

Study design and study site

This study used a cross-sectional, non-random survey design with a qualitative approach. Both semi-structured interviews and focus group discussions were conducted. In-depth interviews and focus group discussions were chosen because no tools to assess family quality of life have been validated in Ethiopia to date. In addition, interviews allow pursuing in-depth information about a particular topic. The study was conducted in the Awi zone, one of the eleven zones in the Amhara region. The Awi zone is located in the Northwest of Ethiopia. The study included three woredas (districts): Zigem, Guagusa Shikudad (Injibara town), and Fagita Lekoma (Addis Kidam town). Leprosy, lymphatic filariasis and podoconiosis are all endemic in the Awi Zone [39–41].

Study population and sample

Six groups of people were included in the study: (1) persons affected by leprosy; (2) persons affected by lymphatic filariasis; (3) persons affected by podoconiosis; (4) family members of persons affected by leprosy; (5) family members of persons affected by lymphatic filariasis; and (6) family members of persons affected by podoconiosis. Throughout this manuscript, we will sometimes refer to groups one to three as “persons affected” and groups four to six as “family members”.

We aimed to have semi-structured interviews with at least 10 persons from each participant group. We also aimed to conduct one focus group discussion per participant group with at least five participants in each group.

Eligibility criteria

Participants had to live in one of the three districts included in the study. The persons affected had to be diagnosed with leprosy, lymphatic filariasis or podoconiosis and had to have visible impairments due to their condition. Family members had to live in the same household as the persons affected. Persons unwilling or unable to give informed consent, persons younger than 16 years of age and persons affected whose family members did not know of their condition were excluded.

Sampling methods

Because we wanted to interview participants with specific characteristics, participants were selected using convenience sampling. Local health posts and organisations of persons affected in the study area were visited to check whether there were persons affected that met the inclusion criteria. A list was prepared of all eligible persons in the study area. From this list, participants were visited in their home and included based on their availability. Family members were selected by the persons affected from among those living in the same household, based on their availability. One family member per person affected was included.

Data collection

Data were collected between August and November 2017. Cross-sectional data on participants' daily life, family (quality of) life and what it is like to have an 'affected' family member were obtained using semi-structured in-depth interviews and focus group discussions. Interviews were conducted by four local health extension workers who spoke both Amharic and Agew languages. The interviewers were trained in leprosy, podoconiosis, lymphatic filariasis and interviewing techniques prior to data collection. The interview guides were pilot tested before data collection, minor revisions to the interview guide were made based on the pilot interviews. These participants were not included in the final sample. The interviews were conducted either in participants' homes or, if they were members of a patient organisation, in a private space near the patient organisation. The in-depth interviews lasted on average 40 minutes, the focus group discussion lasted on average 90 minutes. The in-depth interviews and focus group discussions were audio recorded. A district coordinator monitored the entire process.

Data analysis

The recordings of qualitative data were transcribed, translated to English and coded. A unique identifying number was given to all participants in advance, so sensitive personal data of participants were removed before analysis. Three rounds of coding were done: the first two rounds, conducted by two independent researchers, comprised of open, inductive coding and content analysis. Similar phrases with recurring themes that were derived from the data were coded. In the third analysis round, conducted by a third researcher, relevant domains were selected based on the themes identified in the first two rounds. All data were then analysed again and clustered together in different tables, ordered by theme: physical, psychological aspects and mental wellbeing, level of independence, environment, and social support and family relations. Microsoft Word, Microsoft Excel and Open Code 4.03 software were used to analyse the data.

Ethical considerations

Ethical approval for this study was obtained as part of a larger project that aims to develop a family-based approach to support prevention of disabilities in the Ethiopian context. Ethical approval was obtained from the Debre Markos University Health Science College Research Review Committee. Permission to conduct the study in the region was also gained from the Awi zone (district) Health Desk office. The literacy rate is low in Ethiopia, only 51.8% of people over 15 years old are literate. (*http://uis.unesco.org/en/country/et*). The literacy rate is even lower in our study sample, formed by persons with visible impairments and their family members predominantly living in rural areas. Therefore, all participants were verbally informed about the nature and objective of the study and of confidentiality of the data prior to data collection. All participants were allowed to ask questions and were given sufficient time to consider whether or not they wanted to participate in the research. Enrolment was voluntary, verbal consent from each participant was obtained prior to data collection. Parental consent was obtained in addition to the child's own consent for the two participants who were below 18 years old.

Results

Demographic information

A total of 86 participants were included in this study. Fifty-six participants were included in the in-depth interviews: 14 persons affected by podoconiosis, 12 persons affected by leprosy, 12 persons affected by lymphatic filariasis, one person affected by both leprosy and lymphatic filariasis and 17 family members of persons affected. Family members were children (n=8), spouses (n=4), parents (n=2), sibling (n=2) or grandparents (n=1). Most family members (n=11) were family members of a person affected by podoconiosis instead of leprosy (n=3) or lymphatic filariasis (n=3). The average family size was 5 people, ranging from 1 to 10 (standard deviation 2.5).

The average age of all participants was 43 years (range 17-73). Persons affected by leprosy (51 years) and podoconiosis (48 years) were, on average, older than persons affected by lymphatic filariasis (39 years) and the group of family members (36 years). Many participants were farmers (n=16) or worked in daily labour (n=16). Three persons affected and two family members were students. Almost two-third of the participants had no formal education (n=36).

Four focus group discussions were conducted to supplement the in-depth interviews. Thirty people were included in the focus group discussions: ten persons affected by leprosy, six children/grandchildren of persons affected by leprosy, eight persons affected by podoconiosis and six children of persons affected by podoconiosis. The average age was 52 (range 25-80) for the persons affected and 20 (range 16-27) for the family members. An overview of the number of participants in the interviews can be found in Table 1. All persons affected included in the focus group discussions were uneducated (n=18/18), while most family members had completed primary or secondary education (n=11/12). Table 2 provides an overview of the demographic information of the participants.

Table 1. Overview of the number of participants (*n*=86) included in the in-depth interviews and focus group discussion.

	Persons affected		**Family members of persons affected**	
	Interview	**Focus group discussion**	**Interview**	**Focus group discussion**
Podoconiosis	14	8	11	6
Lymphatic filariasis	12	-	3	
Leprosy	12	10	3	6
Leprosy and lymphatic filariasis	1	-	-	-
Total	39	18	17	12

Table 2. Socio-demographic characteristics of the participants (*n*=86).

	In-depth interviews (*n*=56)	**Focus group discussions (*n*=30)**
Average age (range)	43 (17-73)	39 (16-80)
Female, *n* (%)	32 (57%)	16 (53%)
Participant type		
Person affected podoconiosis, *n* (%)	14 (25%)	8 (27%)
Person affected by leprosy, *n* (%)	12 (21%)	10 (33%)
Person affected by LF, *n* (%)	12 (21%)	-
Person affected by leprosy and LF, *n* (%)	1 (2%)	-
Family member, *n* (%)	17 (30%)	12 (40%)
Family size, mean (range, SD)	5 (1-10, 2.5)	5 (2-7, 1.5)
No education, *n* (%)	36 (64%)	19 (63%)
Occupation		
Farmer, *n* (%)	16 (29%)	15 (50%)
Daily labour, *n* (%)	16 (29%)	8 (27%)
Student, *n* (%)	5 (9%)	-
Other, *n* (%)	19 (34%)	7 (23%)

Physical: symptoms, cause and self-care

Persons affected by podoconiosis and lymphatic filariasis reported symptoms like itching, rashes and swelling – mostly of the legs. The majority of the persons affected by podoconiosis and lymphatic filariasis (n=20/26; 11 persons affected by podoconiosis and 9 persons affected by lymphatic filariasis) reported that they frequently experienced pain because of their condition. Persons affected by leprosy described their symptoms as itching, a loss of sensation and/or not feeling pain, wounds and inability to use their hands. Some participants (n=11/39) said their symptoms increased over time.

A person affected by leprosy explained:

> *"...First it started when I swam in the river with a scabies-like rash on my whole body and it was itching, finally the wound started from my foot and spread to my whole body, then a feeling of senseless, finally it eats my fingers and I lose my fingers..."*
>
> (Man affected by leprosy, age 60)

Most participants in the in-depth interviews, persons affected and their family members, believed the disease was either God's will (n=26/56) or punishment (n=2/56), hereditary (n=17/56) or caused by something else (n=9/56). Some participants (n=7/39) indicated that the disease is normally hereditary, but not in their case because they didn't have any relatives who were affected by the disease. Most participants in the focus group discussions believed the disease was 'from God', hereditary, due to sins, or because of walking barefoot. One participant explained:

> *"...My father and I assumed that the disease would be transmitted to my children but the reality is not that because my children are still not affected now..."*
>
> (Man affected by leprosy, age 64, focus group discussion)

A number of participants believed traditional medicine or holy water would cure their condition. Almost a quarter of the persons affected (n=9/39) explained they went to the holy waters to try and get a cure for their condition or some relief from the pain. Some participants said they (had) used traditional medicine (n=6/39; 3 persons affected by podoconiosis, two lymphatic filariasis, one leprosy). The majority of the participants said they regularly practiced self-care. Two participants indicated they were not able to practice self-care because they felt weak. One participant explained:

> *"...I wash my leg using soap and water and I wear my shoes. My family takes me to health centre and holy water..."*
>
> (Woman affected by lymphatic filariasis, age 25)

Psychological aspects and mental wellbeing

The majority of the persons affected (n=21/39; ten persons affected by lymphatic filariasis, five podoconiosis, five leprosy, one leprosy and lymphatic filariasis) described a mental health issue. Some participants said that they felt inferior compared to their friends or community members (n=7/39; four persons affected by leprosy, two lymphatic filariasis, one podoconiosis). A few participants affected by podoconiosis or lymphatic filariasis said that they (n=3/26) or their affected family member (n=2/26) sometimes felt sad or depressed. Three other participants, women affected by lymphatic filariasis, indicated that they felt like they had no opportunities.

Other psychological challenges described by the participants include worrying (n=3/39), low self-esteem (n=2/39), being ashamed (n=2/39), feeling hopeless (n=1/39), feeling deserted (n=1/39) and sleeping problems (n=1/39). In addition, one person affected by leprosy from the focus group discussion said he used to have suicidal thoughts.

Level of independence: day-to-day life, work and resources

Over three quarters of the participants reported on their ability to move around and do their day-to-day activities (n=30/39). One third of the participants described some (n=8/30) or severe (n=11/30) activity limitations. Participants said they had difficulty moving, for example they were unable to walk long distances, unable to move and/or had a low energy level. Some participants affected by podoconiosis or lymphatic filariasis (n=5/30) indicated that they only experienced limitations when they were in pain. There were also participants who did not have any problems moving around or in their day-to-day activities (n=6/30):

> *"…I can walk, move and take care of myself…"*
>
> (Man affected by lymphatic filariasis, age 40)

Almost one in five persons affected said they were still able to work as they did before (n=7/39), five persons affected did not answer and almost three quarters of the persons affected said they were not able to work as before (n=27/39; 12 podoconiosis, nine leprosy, five lymphatic filariasis, one leprosy and lymphatic filariasis). In addition, not being able to work in the same capacity was mentioned in the focus group discussions by a number of participants (n=9/30; three persons affected by leprosy, six family members). The participants who indicated they were not able to work as before said this was the case because of their condition, pain, disability or because working on the land had become too hard. Some participants had taken on other, lighter work (n=5/39).

> *"…I work in handicraft since my hand's fingers are well (…) I cannot do my previous agricultural work because of my disease. If [I'm] exposed to soil and*

> *mud my wound aggravates, that makes me poor. I thank God my hand is well…"*
>
> (Woman affected by leprosy, age 56)

Almost half of the persons affected and their family members stated they were poor, lived in poverty and/or lacked money, without being asked about their financial situation by the interviewer (n=27/56). An additional eight participants indicated they were in need of money, without mentioning that they were poor. All participants who said they went to the holy water, a costly expedition, said they were poor. Other things asked for by the participants include "being cured" or (effective) treatment (n=21/56), materials such as Vaseline and shoes (n=10/56), government support (n=7/56) and a loan (n=5/56). In addition, some participants (n=8/56) stated that they wished to move from the rural to the urban areas, because there is less mud in the towns. One participant said:

> *"…I became economically dependent on my family due to my disease (…) I wish I was cured either by holy water or drugs so that [I'm] able to work effectively. My great obstacle is poverty. I cannot afford soap, food and transport for my treatment…"*
>
> (Man affected by podoconiosis, age 35)

Similar results were found in the focus group discussions. In addition, participants from the focus group discussion explained that living further away from the town also brings additional costs for transportation.

Environment: attitudes and social participation

Discrimination in the communities was common. The majority of the participants in the in-depth interviews (n=27/39; 11 podoconiosis, ten leprosy, five lymphatic filariasis, one leprosy and lymphatic filariasis) and almost half of the persons affected in the focus group discussions (n=8/18) said they were discriminated against by their community members. Many of these participants (n=20/27 in the in-depth interviews and n=7/8 in the FGD) were also insulted, e.g. they were called 'leper', 'lost finger' or 'swollen leg'. Two participants explained:

> *"…Some of the community members see him as inferior…"*
>
> (Wife of person affected by podoconiosis, age 45)

And

> *"...Many of my neighbours used to say we cannot enter their house, they separate me from coffee [ceremonies] too. Sometimes when I said hello to kids their parents were not happy, some of them warned me not to touch them. That was the worst time during my illness (...) When people discussed my disease and prevented [me] from social life my wife asked me to divorce..."*
>
> (Man affected by leprosy, age 45)

Some participants said they had no, bad or limited social contact with their neighbours and/or community members (n=6/39; three podoconiosis, two leprosy, one lymphatic filariasis). There were also participants who had a good relationship with the community (n=7/39; four podoconiosis, two lymphatic filariasis, one leprosy). One participant said:

> *"...Like me my children also live in good relationship with the community..."*
>
> (Woman affected by podoconiosis, age 70)

Almost half of the family members of persons affected in the in-depth interviews (n=8/17) and all the family members of persons affected by leprosy in the focus group discussions (n=6/6) also experienced discrimination. They were either insulted or discriminated against (n=7/17) or had to leave school because of discrimination or because they had to work to help provide for the family (n=4/17). Two persons affected said they had trouble finding a husband or wife for their children because of their condition. Two participants explained:

> *"...We have a good relation with the majority of the community but some individuals abuse us by saying 'lost finger son' and the like..."*
>
> (Son of person affected by leprosy, age 20, focus group discussions)

And

> *"...Some people insult me and also they insult my children (...) People in the wedding made me stay outside of the tent and they did not treat me as [if] I was healthy. They are afraid of my disease and don't want it to be transmitted to them. How can I be equal with this disease, I sometimes agree with what they did (...) One of my daughters went to school, her friends insult her (...) and now she does not go to school..."*
>
> (Woman affected by podoconiosis, age 40)

About one-third of the persons affected (n=12/39) indicated that they didn't experience any social participation restrictions. In addition, one-third of the persons affected indicated that they experienced social participation restrictions due to activity limitations (n=6/39), stigma (n=3/39), activity limitations and stigma (n=2/39) or their physical condition (n=2/39). Social participation restrictions included isolation, not being invited to weddings, the houses of friends and coffee ceremonies and barriers to employment and education. Two students stated they had to stop their education because they were ill.

> *"…I go to church every morning and help my family with household work (…) I stopped my education due to frequent [acute] attacks of the disease…"*
>
> (Woman affected by lymphatic filariasis, age 18).

Social support and family relations

Over half of the participants said that family support given to the affected family member was good (n=30/56, 20 persons affected and 10 family members). One in five participants said that persons affected received some support from family members (n=12/56; eight persons affected and four family members). An additional one in five participants said no or limited support was given to affected family members (n=12/56; 11 persons affected and one family member). In four "participant pairs" of a person affected by podoconiosis and their family member, the family member thought they were giving more support than the person affected perceived. One family member explained:

> *"…We are also in fear of contracting the disease. For more than one year and eight months I wash his feet, hands, take care of his urine and feed him. Now he starts to take care of himself. We took him to the holy water and the hospital but there is no change and he is not cured (…) There is nothing suitable for the poor, we are in trouble. Both our kids and I are working day-to-day as daily labour to support the family…"*
>
> (Wife of person affected by leprosy, age 36)

Support from family members included moral or psychological support, providing money or other resources, taking over household duties such as cooking and washing clothes and helping with self-care. Participants mostly relied on their children and spouse for support, calling them their "great opportunity". One third of the persons affected were very dependent on others due to motor restrictions (n=3/39) or because they had no or very limited social support (n=10/39; four persons affected by podoconiosis, four lymphatic filariasis, two leprosy). The participants who indicated they had no or very limited social support were living alone, had young children, were very poor with a big family to support or divorced. One participant explained:

> ***"...***_There is no support from my family (...) My wife is weak and cannot give me support and my children are small..."_
>
> (Man affected by lymphatic filariasis, age 40)

In the focus group discussions, the participants explained that young children can't help their families when they are at school and that it is often difficult to afford sending children to school. For this reason, many children of affected families drop out of school early. At the same time, having to provide for their affected family member also impacts the family members, causing stress and additional workload. One participant explained:

> *"...When he was healthy he supported us but because of the disease we are forced to help him rather than getting help from our father. We left our education in order to support the family by doing daily work..."*
>
> (Daughter of person affected by leprosy, age 18, focus group discussion)

Some participants explained that they not only needed support, but also had to give (financial) support their family members (n=4/39). Poverty was a challenge for many families.

One in five participants were divorced because of their condition (n=6/39) or continuously asked by their partner for divorce (n=2/39). One participant explained:

> *"...My wife repeatedly asks me to be divorce and even she was lost for more than two weeks. Then I begged the elders and priests in the town for her to come back. Especially her relatives forced her to leave me..."*
>
> (Man affected by leprosy, age 45)

Divorce seemed to occur more often among persons affected by leprosy (n=5/8): three persons affected by leprosy were divorced and the spouses of two persons affected by leprosy asked for divorce. Divorce seemed to have a negative impact on mental wellbeing. One participant said:

> *"...[I] divorced with my husband (...) He married another wife and had two additional children (...) It was the worst situation in the last times to live with the community but now it is improved. I feel ashamed when people [include] me in social interactions..."*
>
> (Woman affected by leprosy, age 56)

Many persons affected by leprosy said they were part of a leprosy association that gave them a lot of support (n=7/12). Two participants received a lot of support from their neighbours. One participant explained:

> *"...My home renter told me to leave his house since I cannot pay on time. I was forced to leave his house with my children. However, my neighbours pay my rent (...) my neighbours lend me money for holy water and other expenses (...) The people around me helped me what they can ..."*
>
> (Man affected by leprosy, age 38)

Discussion

This study revealed that leprosy, lymphatic filariasis and podoconiosis have an impact on several dimensions of family quality of life. That leprosy, lymphatic filariasis and podoconiosis impact individual quality of life is supported by several other studies[26–31,36]. Two quantitative studies that have been conducted on the impact of podoconiosis on individual quality of life in Ethiopia found that podoconiosis has a negative effect on individual quality of life [29,42]. The present study is the first study into family quality of life of these conditions.

We found that persons affected often experience pain due to their condition. Some participants had activity limitations, such as not being able to walk long distances and an inability to move at all. In one-third of the persons affected in our study, activity limitations and stigma led to social participation restrictions such as isolation, not being invited to coffee ceremonies and barriers to employment and education. These findings are similar to other studies conducted in Africa. In studies among persons affected by leprosy in Nigeria and Mozambique [43–45], persons affected by lymphatic filariasis in Nigeria, Ghana and Malawi [46–50], and persons affected by podoconiosis in Ethiopia [51,52], those interviewed also reported (severe) social participation restrictions. This was often linked to stigmatisation of persons affected [43–47,50,52]. In addition, in a cross-sectional survey study among 233 community members of persons affected by leprosy in Cameroon, only one-third of the participants approved of participation of persons affected by leprosy [53].

The present study also found that discrimination in the communities was common, often extending to family members of persons affected. Persons affected and their family members were sometimes socially excluded and insulted by their community members and divorce and difficulties in finding a spouse were not uncommon. Similar findings were found in a study on women with disabilities in Ethiopia, who experienced societal denial of marriage and motherhood [54]. Several studies among persons affected by leprosy, lymphatic filariasis and podoconiosis found high levels of stigma - for example among persons affected by podoconiosis in Ethiopia [17,20,29,52,55–59]. Some of these studies attributed the high levels of stigma to beliefs about the disease's causation – the belief that podoconiosis is hereditary [55,59]. This belief and the fear of costs of treatment and of disability in turn had a negative influence on marriage prospects and marital stability, also for family members of persons affected [55,59]. High levels of stigma were also found among persons affected by lymphatic filariasis in Ghana [15,50] and Nigeria [46,47] and persons affected by leprosy in Ghana [60,61], Tanzania [62] and Nigeria [43,45,63,64]. Some studies in Africa found that persons affected by leprosy and lymphatic filariasis are also stigmatized by their family members [50,61,62]. This was not found in the nuclear family in the present study.

Some studies found stigma to deteriorate the economic situation of persons affected [45,46,50]. This was found in the present study also, as almost all participants reported financial problems and loss of livelihood. In addition, in our study, almost three-quarters of the persons affected said they were not able to work as before because of their condition. A recent literature review into the extent, similarities and differences of social stigma in neglected tropical diseases found evidence that reduced work opportunities are common among persons affected by neglected tropical diseases such as leprosy, lymphatic filariasis and podoconiosis [11]. In our study, most participants who indicated they were unable to work said they were physically unable to work because of their condition. We think that in the present study, physical impairments that hamper daily functioning in productive activities, high occurrence of divorce and hence loss of social support, large families to support and high costs for (alternative) treatment exacerbated financial problems of participants.

We found that persons with younger children seemed to lack social support. Participants explained that young children can’t help their families when they are at school and that they can’t always afford to send their children to school. For this reason and because of stigma, persons affected and children of affected families drop out of school early. In addition, having to provide for their affected family member also impacted the family members, causing stress and an additional workload. These findings suggest that providing care to affected family members may result in physical, emotional, financial and social burdens that can diminish their (family) quality of life. This is supported by studies in other fields [65–68]. However, in the present study we found that most people receive social and physical support from their families. This is reflected by other studies on family quality of life where families that include a member with disabilities reported positive aspects on their family life, such as problem solving and family sense of coherence [37]. Results from worldwide research on family quality of life show that positive family relationships are a common strength of most families where one or more members have a disability [69], where negative feelings are likely to be attributed to societal norms that are imposed on families [70].

We found that half of the persons affected sometimes experienced negative affect such as feeling sad, ashamed, worried or hopeless. This is consistent with what is already known about the psychological effects of stigmatized conditions [71,72]. In the present study, some persons affected lacked social support or were dependent on others. Two literature reviews found that social relationships play a key role in mental well-being in persons with disabilities [35] and in quality of life of people with mental health problems [73]. In addition, strengthening social support can increase a person's feeling of belonging. Connection and belonging are important to quality of life [73,74]. Some studies even suggest that people are fundamentally motivated by a need to belong [74]. This suggests that strengthening social support and quality relationships may improve mental wellbeing of persons affected by leprosy, lymphatic filariasis and podoconiosis.

Lastly, we found that the quality of life dimensions that were affected (the domains physical, psychological aspects, independence, environment, and social support and family relations) were similar among the three conditions. This finding is supported by a literature review on health-related stigma, that found that the consequences of stigma affect the quality of life of persons affected and that the areas of life affected by stigma are similar in different conditions and different cultural settings [72].

A limitation of this study is the non-random sampling and the small study size per participant group. In addition, the study focused on one geographic location only. This means that the results of the study cannot be generalized to the whole study population or beyond. Another limitation of the study is that we did not register the frequency of 'acute attacks' in persons affected by lymphatic filariasis and podoconiosis. We also did not register severity of disability and the occurrence of reactions in persons affected by leprosy. Acute attacks and leprosy reactions may affect the quality of life of persons affected. However, we collected data on participants' experiences of pain.

Taking the above limitations into account, the results of our study offer insights into the impact of leprosy, lymphatic filariasis and podoconiosis on family quality of life in Awi zone, Ethiopia. The results of this study have a number of implications for leprosy, lymphatic filariasis and podoconiosis treatment and after care programmes. Many of the problems reported in the present study related to impairments, stigma and a lack of finances. A family-based approach that addresses self-care and social and economic aspects may improve individual and family quality of life. Micro-credit loans and vocational training may reduce stigma by protecting persons affected against loss of social value [63] while strengthening social support and quality relationships may improve mental wellbeing of persons affected and their family members [73,74]. Efforts to improve quality of life of persons affected and their family members should give priority to those who are living alone, have young children, or do not have a partner.

Conclusion

This study revealed that leprosy, lymphatic filariasis and podoconiosis have an effect on several dimensions of family quality of life. Physically, persons affected often experience pain. Psychologically, persons affected experienced negative affect, for example feeling depressed, inferior, deserted, ashamed, worried and/or hopeless. Socially, participation restrictions, reduced productivity and marginalisation were common. Discrimination of persons affected and their family members occurred often. Divorce and difficulties in finding a spouse, especially for persons affected by leprosy, were not uncommon, even extending to their family members. Persons with younger children seemed to lack social support. Having to provide for their affected family member sometimes caused stress, school dropouts and an additional workload. Financial problems and loss of livelihood were reported by almost all participants.

We found that the areas of life that were affected were similar among the three conditions. This indicates that programmes focusing on treatment and after care of persons affected should follow a holistic approach that addresses the physical, psychological, social and environmental impact of the disease and focus on the entire family. A family-based approach that addresses self-care and social and economic aspects may improve individual and family quality of life. Efforts to improve quality of life of persons affected and their family members should give priority to those who are living alone, have young children, or do not have a partner.

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References

1. Lastória JC, Abreu MAMM de. Leprosy: review of the epidemiological, clinical, and etiopathogenic aspects-part 1. An Bras Dermatol 2014;89:205–18.
2. Suzuki K, Akama T, Kawashima A, Yoshihara A, Yotsu RR, Ishii N. Current status of leprosy: epidemiology, basic science and clinical perspectives. J Dermatol 2012;39:121–9.
3. Babu S, Nutman TB. Immunology of lymphatic filariasis. Parasite Immunol 2014;36:338–46.
4. Davey G. Podoconiosis, non-filarial elephantiasis, and lymphology. Lymphology 2010;43:168.
5. Fuller LC. Podoconiosis: endemic nonfilarial elephantiasis. Curr Opin Infect Dis 2005;18:119–22.
6. Davey G. Podoconiosis: let Ethiopia lead the way. Ethiop J Heal Dev 2008;22:1–2.
7. World Health Organization. Lymphatic filariasis. Podoconiosis: endemic non-filarial elephantiasis. 2019. https://www.who.int/lymphatic_filariasis/epidemiology/podoconiosis/en/ (accessed May 20, 2019).
8. Yimer M, Hailu T, Mulu W, Abera B. Epidemiology of elephantiasis with special emphasis on podoconiosis in Ethiopia: a literature review. J Vector Borne Dis 2015;52:111.
9. Lyons OTA, Modarai B. Lymphoedema. Surg 2013;31:218–23.
10. Ministry of Health Federal Democratic Republic of Ethiopia. Lymphatic filariasis and podoconiosis morbidity management and disability prevention guidelines. Addis Ababa, Ethiop Minist Heal 2016.
11. Hofstraat K, van Brakel WH. Social stigma towards neglected tropical diseases: a systematic review. Int Health 2016;8:i53–70.
12. Rafferty J. Curing the stigma of leprosy. Lepr Rev 2005;76:119–26.
13. Singh GP. Psychosocial aspects of Hansen's disease (leprosy). Indian Dermatol Online J 2012;3:166.
14. van Brakel WH. Measuring leprosy stigma-a preliminary review of the leprosy literature1. Int J Lepr Other Mycobact Dis 2003;71:190.
15. Gyapong M, Gyapong JO, Adjei S, Vlassoff C, Weiss M. Filariasis in northern Ghana: some cultural beliefs and practices and their implications for disease control. Soc Sci Med 1996;43:235–42.
16. Perera M, Whitehead M, Molyneux D, Weerasooriya M, Gunatilleke G. Neglected patients with a neglected disease? A qualitative study of lymphatic filariasis. PLoS Negl Trop Dis 2007;1:e128.
17. Tora A, Davey G, Tadele G. A qualitative study on stigma and coping strategies of patients with podoconiosis in Wolaita zone, Southern Ethiopia. Int Health 2011;3:176–81.
18. Molla YB, Tomczyk S, Amberbir T, Tamiru A, Davey G. Podoconiosis in East and west gojam zones, northern ethiopia. PLoS Negl Trop Dis 2012;6:e1744.
19. Hotez PJ. Stigma: the stealth weapon of the NTD 2008.
20. Deribe K, Tomczyk S, Mousley E, Abreham T, Davey G. Stigma towards a neglected tropical disease: felt and enacted stigma scores among podoconiosis patients in Northern Ethiopia. BMC Public Health 2013;13:1178.
21. Cassidy T, Worrell CM, Little K, Prakash A, Patra I, Rout J, et al. Experiences of a community-based Lymphedema management program for lymphatic Filariasis in Odisha state, India: an

analysis of focus group discussions with patients, families, community members and program volunteers. PLoS Negl Trop Dis 2016;10:e0004424.

22. de Stigter DH, Geus LD, Heynders ML. Leprosy: between acceptance and segregation. Community behaviour towards persons affected by leprosy in eastern Nepal. Lepr Rev 2000;71:492–8.
23. Nicholls PG, Bakirtzief Z, Van Brakel WH, Das-Pattanaya RK, Raju MS, Norman G, et al. Risk factors for participation restriction in leprosy and development of a screening tool to identify individuals at risk. Lepr Rev 2005;76:305–15.
24. Tekola F, Bull S, Farsides B, Newport MJ, Adeyemo A, Rotimi CN, et al. Impact of social stigma on the process of obtaining informed consent for genetic research on podoconiosis: a qualitative study. BMC Med Ethics 2009;10:13.
25. Try L. Gendered experiences: marriage and the stigma of leprosy. Asia Pacific Disabil Rehabil J 2006;17:45–6.
26. Borges-de-Oliveira R, Rocha-Leite CI, Araujo-de-Freitas L, Queiroz DA, Machado PRL, Quarantini LC. Perception of social exclusion, neuropathy, and quality of life among Hansen's disease patients. Int J Psychiatry Med 2015;49:176–86.
27. Lustosa AA, Nogueira LT, Pedrosa JI dos S, Teles JBM, Campelo V. The impact of leprosy on health-related quality of life. Rev Soc Bras Med Trop 2011;44:621–6.
28. Morgan PA, Franks PJ, Moffatt CJ. Health-related quality of life with lymphoedema: a review of the literature. Int Wound J 2005;2:47–62.
29. Mousley E, Deribe K, Tamiru A, Davey G. The impact of podoconiosis on quality of life in Northern Ethiopia. Health Qual Life Outcomes 2013;11:122.
30. Tsutsumi A, Izutsu T, Islam AM, Maksuda AN, Kato H, Wakai S. The quality of life, mental health, and perceived stigma of leprosy patients in Bangladesh. Soc Sci Med 2007;64:2443–53.
31. Santos VS, Oliveira LS, Castro FDN, Gois-Santos VT, Lemos LMD, do CO Ribeiro M, et al. Functional activity limitation and quality of life of leprosy cases in an endemic area in Northeastern Brazil. PLoS Negl Trop Dis 2015;9:e0003900.
32. World Health Organization. World report on disability 2011. World Health Organization; 2011.
33. Theofilou P. Quality of Life: Definition and Measurement. Eur J Psychol 2013;9.
34. Poston D, Turnbull A, Park J, Mannan H, Marquis J, Wang M. Family quality of life: A qualitative inquiry. Ment Retard 2003;41:313–28.
35. Tough H, Siegrist J, Fekete C. Social relationships, mental health and wellbeing in physical disability: a systematic review. BMC Public Health 2017;17:414.
36. Thomas C, Narahari SR, Bose KS, Vivekananda K, Nwe S, West DP, et al. Comparison of three quality of life instruments in lymphatic filariasis: DLQI, WHODAS 2.0, and LFSQQ. PLoS Negl Trop Dis 2014;8:e2716.
37. Zuna NI, Brown I, Brown RI. Family quality of life in intellectual and developmental disabilities: A supportbased framework to enhance quality of life in other families. Qual. life Intellect. Disabil. Knowl. Appl. to other Soc. Educ. challenges, Nova Science Publishers, Inc.; 2014, p. 91–119.
38. Brown RI, Schippers A. Quality of life and family quality of life: Recent developments in research and application. J Intellect Dev Disabil 2016;41:277–8.
39. World Health Organization. Weekly Epidemiological Record, 31 August 2018. Global leprosy update, 2017: reducing the disease burden due to leprosy. vol. 93. 2018.

40. World Health Organization. Weekly Epidemiological Record, 2 November 2018. Global programme to eliminate lymphatic filariasis: progress report, 2017. vol. 93. 2018.
41. Deribe K, Cano J, Trueba ML, Newport MJ, Davey G. Global epidemiology of podoconiosis: A systematic review. PLoS Negl Trop Dis 2018;12:e0006324.
42. Henok L, Davey G. Validation of the Dermatology Life Quality Index among patients with podoconiosis in southern Ethiopia. Br J Dermatol 2008;159:903–6.
43. Ibikunle PO, Nwokeji SC. Assessment of stigma among people living with Hansen's disease in south-east Nigeria. Lepr Rev 2017;88:43–57.
44. Deepak S, Hansine PE, Braccini C. Self-care groups of leprosy-affected people in Mozambique. Lepr Rev 2013;84:283.
45. Awofeso N. Stigma and socio-economic reintegration of leprosy sufferers in Nigeria. Acta Leprol 1996;10:89–91.
46. Abdulmalik J, Nwefoh E, Obindo J, Dakwak S, Ayobola M, Umaru J, et al. Emotional difficulties and experiences of stigma among persons with lymphatic filariasis in Plateau State, Nigeria. Health Hum Rights 2018;20:27.
47. Eneanya OA, Garske T, Donnelly CA. The social, physical and economic impact of lymphedema and hydrocele: a matched cross-sectional study in rural Nigeria. BMC Infect Dis 2019;19:332.
48. Stanton MC, Yamauchi M, Mkwanda SZ, Ndhlovu P, Matipula DE, Mackenzie C, et al. Measuring the physical and economic impact of filarial lymphoedema in Chikwawa district, Malawi: a case-control study. Infect Dis Poverty 2017;6:28.
49. Martindale S, Mkwanda SZ, Smith E, Molyneux D, Stanton MC, Kelly-Hope LA. Quantifying the physical and socio-economic burden of filarial lymphoedema in Chikwawa District, Malawi. Trans R Soc Trop Med Hyg 2014;108:759–67.
50. Person B, Bartholomew LK, Gyapong M, Addiss DG, van den Borne B. Health-related stigma among women with lymphatic filariasis from the Dominican Republic and Ghana. Soc Sci Med 2009;68:30–8.
51. Bekele K, Deribe K, Amberbir T, Tadele G, Davey G, Samuel A. Burden assessment of podoconiosis in Wayu Tuka woreda, east Wollega zone, western Ethiopia: a community-based cross-sectional study. BMJ Open 2016;6:e012308.
52. Tora A, Franklin H, Deribe K, Reda AA, Davey G. Extent of podoconiosis-related stigma in Wolaita Zone, Southern Ethiopia: a cross-sectional study. Springerplus 2014;3:647.
53. Tabah EN, Nsagha DS, Bissek A-CZ-K, Njamnshi TN, Njih IN-N, Pluschke G, et al. Community knowledge, perceptions and attitudes regarding leprosy in rural Cameroon: The case of Ekondotiti and Mbonge health districts in the South-west Region. PLoS Negl Trop Dis 2018;12: e0006233.
54. Tefera B, Van Engen M, Van der Klink J, Schippers A. The grace of motherhood: disabled women contending with societal denial of intimacy, pregnancy, and motherhood in Ethiopia. Disabil Soc 2017;32:1510–33.
55. Ayode D, Tora A, Farrell D, Tadele G, Davey G, McBride CM. Dual perspectives on stigma: reports of experienced and enacted stigma by those affected and unaffected by podoconiosis. J Public Health Res 2016;5.
56. Tsegay G, Wubie M, Degu G, Tamiru A, Cooper M, Davey G. Barriers to access and re-attendance for treatment of podoconiosis: a qualitative study in northern Ethiopia. Int Health 2014;7:285–92.

57. Franklin H, Tora A, Deribe K, Reda AA, Davey G. Development of a scale to measure stigma related to podoconiosis in Southern Ethiopia. BMC Public Health 2013;13:298.
58. Tora A, Davey G, Tadele G. Factors related to discontinued clinic attendance by patients with podoconiosis in southern Ethiopia: a qualitative study. BMC Public Health 2012;12:902.
59. Ayode D, McBride CM, de Heer H, Watanabe E, Gebreyesus T, Tadele G, et al. The association of beliefs about heredity with preventive and interpersonal behaviors in communities affected by podoconiosis in rural Ethiopia. Am J Trop Med Hyg 2012;87:623–30.
60. Sottie CA, Darkey J. Living with stigma: Voices from the Cured Lepers' village in Ghana. Soc Work Health Care 2019;58:151–65.
61. Dako-Gyeke M, Asampong E, Oduro R. Stigmatisation and discrimination: Experiences of people affected by leprosy in Southern Ghana. Lepr Rev 2017;88:58–74.
62. Roosta N, Black DS, Rea TH. A comparison of stigma among patients with leprosy in rural Tanzania and urban United States: a role for public health in dermatology. Int J Dermatol 2013;52:432–40.
63. Ebenso B, Ayuba M. "Money is the vehicle of interaction": insight into social integration of people affected by leprosy in northern Nigeria. Lepr Rev 2010;81:99.
64. Ebenso B, Newell J, Emmel N, Adeyemi G, Ola B. Changing stigmatisation of leprosy: an exploratory, qualitative life course study in Western Nigeria. BMJ Glob Heal 2019;4:e001250.
65. Fitryasari R, Yusuf A, Tristiana RD, Nihayati HE. Family members' perspective of family Resilience's risk factors in taking care of schizophrenia patients. Int J Nurs Sci 2018;5:255–61.
66. Borges EL, Franceschini J, Costa LHD, Fernandes ALG, Jamnik S, Santoro IL. Family caregiver burden: the burden of caring for lung cancer patients according to the cancer stage and patient quality of life. J Bras Pneumol 2017;43:18–23.
67. Lai S-T, Tan W-Y, Wo MC-M, Lim K-S, Ahmad SB, Tan C-T. Burden in caregivers of adults with epilepsy in Asian families. Seizure 2019;71:132–9.
68. Grunfeld E, Coyle D, Whelan T, Clinch J, Reyno L, Earle CC, et al. Family caregiver burden: results of a longitudinal study of breast cancer patients and their principal caregivers. CMAJ 2004;170:1795–801.
69. Samuel PS, Rillotta F, Brown I. The development of family quality of life concepts and measures. J Intellect Disabil Res 2012;56:1–16.
70. Boelsma F, Schippers A, Dane M, Abma TA. "Special" families and their "normal" daily lives: Family quality of life and the social environment. Int J Child, Youth Fam Stud 2018;9:107–24.
71. Litt E, Baker MC, Molyneux D. Neglected tropical diseases and mental health: a perspective on comorbidity. Trends Parasitol 2012;28:195–201.
72. Van Brakel WH. Measuring health-related stigma—a literature review. Psychol Health Med 2006;11:307–34.
73. Connell J, Brazier J, O'Cathain A, Lloyd-Jones M, Paisley S. Quality of life of people with mental health problems: a synthesis of qualitative research. Health Qual Life Outcomes 2012;10:138.
74. Baumeister RF, Leary MR. The need to belong: desire for interpersonal attachments as a fundamental human motivation. Psychol Bull 1995;117:497.

PART 3

Interventions to change perception at community level and interventions to reduce the impact of leprosy at individual and family level

CHAPTER 7

Changing perception and improving knowledge of leprosy: an intervention study in Uttar Pradesh, India

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Abstract

Introduction

Since ancient times leprosy has had a negative perception, resulting in stigmatization. To improve the lives of persons affected by leprosy, these negative perceptions need to change. The aim of this study is to evaluate interventions to change perceptions and improve knowledge of leprosy.

Methodology/Principal Findings

We conducted a pre-post intervention study in Fatehpur and Chandauli districts, Uttar Pradesh, India. Based on six steps of quality intervention development (6SQuID) two interventions were designed: (1) posters that provided information about leprosy and challenged misconceptions, and (2) meetings with persons affected by leprosy, community members and influential people in the community. The effect of the interventions was evaluated in a mixed-methods design; in-depth interviews, focus group discussions, and questionnaires containing a knowledge measure (KAP), two perception measures (EMIC-CSS, SDS) and an intervention evaluation tool. 1067 participants were included in Survey 1 and 843 in Survey 2. The interventions were effective in increasing knowledge of all participant groups, and in changing community and personal attitudes of close contacts and community members (changes of 19%, 24% and 13% on the maximum KAP, EMIC-CSS and SDS scores respectively, $p<0.05$). In Survey 1, 13% of participants had adequate knowledge of leprosy versus 53% in Survey 2. Responses showed stigmatizing community attitudes in 86% (Survey 1) and 61% (Survey 2) of participants and negative personal attitudes in 37% (Survey 1) and 19% (Survey 2). The number of posters seen was associated with KAP, EMIC-CSS and SDS scores in Survey 2 ($p<0.001$). In addition, during eight post-intervention focus group discussions and 48 interviews many participants indicated that the perception of leprosy in the community had changed.

Conclusions/Significance

Contextualized posters and community meetings were effective in changing the perception of leprosy and in increasing leprosy-related knowledge. We recommend studying the long-term effect of the interventions, also on behavior.

Introduction

Perception is a broad concept, that refers to how an individual or group "sees" an object, person, event or institution [1–3]. Perception encompasses how an individual or group "sees" others (social perception), but also a person's interpretation and understanding of a disease and its potential consequences (disease perception) [1–3]. Perception comprises knowledge, beliefs, attitudes and emotions that are in turn influenced by personal factors (e.g. personality, experience) and environmental factors (e.g. culture, religion) [1,2]. These concepts are interrelated. (Negative) perception is related to stigma. However, where perception is solely cognitive, stigma includes both cognitive (e.g. knowledge, attitudes, labelling) and behavioral (e.g. discrimination, rejection, withdrawal) elements [4,5]. Perception is an important driver of stigma [6].

Leprosy is an infectious disease that has had a negative perception, resulting in stigmatization, since ancient times [7]. The main causes of leprosy-related stigma are the external manifestations of the disease (such as impairments of eyes, hands and feet), religious and cultural beliefs, fear, and a lack of knowledge [7,8]. Almost all areas of a person's life can be affected by stigma, such as employment and education opportunities, social interaction, marriage (prospects), housing and access to care [9]. These negative consequences and the fear of being stigmatized can cause chronic stress, which may negatively impact mental wellbeing and physical health [9]. In the case of health-related stigma, the fear of being stigmatized may also cause people to delay or avoid seeking treatment or care [9]. To improve the lives of persons affected by leprosy and to improve leprosy services, negative perceptions about leprosy need to be addressed.

There are several strategies and interventions to change the perception of leprosy. Many of these are similar or the same as interventions for stigma reduction. Interventions that aim to reduce stigma often address the causes of stigma, such as beliefs and attitudes that lead to labelling, stereotyping and discrimination [10]. Interventions that have reduced leprosy-related stigma include 'contact events' in which contact between persons affected by leprosy and community members is enhanced, socioeconomic rehabilitation, peer counselling, social marketing campaigns, community engagement interventions and mass media campaigns [11–16]. Crucial to changing perceptions is understanding the local context, and understanding and addressing the drivers of these perceptions [10,17,18]. Interventions should fit the audience [19]. They are more likely to be successful if culture-specific and contextualized (adapted to the local context) [20,21], addressing the main causes of leprosy: specific knowledge gaps, beliefs and fears [22].

The present study is part of a project on leprosy prevention in India, Indonesia and Brazil, the PEP++ project (https://www.trialregister.nl/trial/7022). The aim of the present study is to evaluate interventions to change perception and improve knowledge of leprosy.

Definitions

Perception comprises knowledge, beliefs, attitudes and emotions. Beliefs link an object (such as a person, group of people, disease, institution or behaviour) to an attribute. For example the belief “leprosy is dangerous”, links “leprosy” (object) to “dangerous” (attribute) [16]. Knowledge refers to theoretical or practical understanding of a subject (facts, skills or objects). Truth and belief are a prerequisite for possessing knowledge: one has a belief in something, and that belief must be true (based on observable and measurable evidence). For example, if you know that leprosy is an infectious disease, then you must believe this, and your belief must be true [17]. An attitude refers to a person's feelings toward and evaluation of an aspect of the person's world, for example an object, person, event, or towards performing specific behaviours [16,18]. It refers to “a person’s location on a dimension of affect or evaluation” and falls on a continuum from very favourable to very unfavourable [16]. Emotions are inner states such as anger, joy, fear or love. Emotions can be consciously experienced, but can also be repressed, inhibited or unconscious [21].

Methods

Ethics statement

Ethical approval for this study was obtained from the Vardhman Mahavir Institutional Ethics Committee as part of a larger research project: the PEP++ project. Written informed consent for participation was obtained from each participant prior to data collection.

Study setting

The study was conducted in two districts in Uttar Pradesh, northern India: Chandauli (population 1.95 million, 1548 villages) and Fatehpur (population 2.63 million, 1476 villages). These districts have a relatively high number of new leprosy patients annually with a new case detection rate of 5.9 per 100,000 population in both Chandauli and Fatehpur, in March 2019 (District Leprosy Office).

Study design

We applied a pre/post intervention study design. The effect of the interventions was evaluated using mixed methods.

Eligibility criteria

We included four groups as participants in the study: (1) persons diagnosed with leprosy at any time ("persons affected by leprosy"); (2) close contacts of persons affected by leprosy, these comprised household contacts, family members, neighbours and other social contacts; (3) community members; and (4) health care workers. Only individuals 16 years or older were included. Close contacts, community members and health care workers were excluded if they had ever been diagnosed with leprosy.

Interventions

The PEP++ project includes interventions that aim to change the perception of leprosy, improve knowledge of leprosy and reduce stigma, and to increase the community acceptance of preventive (chemoprophylactic) treatment. These interventions provide contextualized (adapted to the local context) information, education and communication (IEC) and are implemented before the implementation of the actual chemoprophylaxis. In doing so, we aim to increase acceptance and adherence to preventive chemoprophylactic treatment. The interventions were designed based on the six steps of quality intervention development (6SQuID) [23] using a community engagement method. 6SQuID is a pragmatic guide, based on existing frameworks for the development of interventions with a wider public health focus. The main input for the selection of the interventions (content and modes of delivery) came from: a) 'leprosy perception study' (Survey 1) of people's knowledge and perceptions of leprosy and persons affected by leprosy [8,24,25], b) a 'communication needs assessment', and c) a workshop with input from persons affected by leprosy and other key stakeholders. A detailed description of the selection and development, including the pilot tests, of the interventions can be found as supporting information file (S1 Text).

Two interventions are assessed in this paper: (1) posters and (2) community meetings. Posters were available in three sizes (46x58 cm, 44x14 cm and 28x23 cm) and covered the following themes: symptoms, mode of transmission, cause, curability, (free) treatment, prevention of leprosy, and inclusion of persons affected by leprosy in the community. The posters and an English translation can be found as supporting information file (S2 Text). The villages in which the posters were put up were selected based on the number of leprosy patients registered at the health center since April 2014. Only villages in which at least two patients were registered were selected. The posters were placed at several locations in the villages (e.g., at the village leader's house, shops, the health facility, crossroads, ATMs, temples and the marketplace) and near sites of public transport (e.g., in buses and auto rickshaws, and at bus stops and railway stations).

The community meetings were held in villages selected from the list of 606 villages in which posters were put up. Villages in which the Pradhan (village leader) was available on scheduled meeting days and where the prior relationship with the Pradhan was good, were selected. Community members were invited to attend the meeting through the Pradhan and by door-to-door visits from community health workers (ASHA's). The meetings itself consisted of a short presentation about leprosy and the PEP++ project, followed by questions-and-answers and a discussion. In some meetings, two short videos about leprosy were also presented (due to technical issues this was not possible in all meetings). A health worker was present during the meetings. Meetings with key influential people were held at district or block level, while meetings with community members and persons affected by leprosy were held in the communities. Participants were also offered a leaflet with more information about leprosy (facts, myths and misconceptions) and the PEP++ project at the meetings. An overview of the reach of the two interventions can be found in Table 1. We report on perception and knowledge of leprosy before and after the interventions.

Table 1. An overview of the interventions, their target groups, periods of dissemination.

Intervention	Target group	Time period disseminated
Posters 16,070 large size posters (46x58 cm) and 8,384 smaller size posters (4,192 size 44x14 cm and 4,192 size 28x23 cm) were put up in 606 villages across the two districts. A total of six different formats (different images and key messages) were used.	• Persons affected by leprosy • Close contacts • Community members	October 2019 - April 2021 (ongoing for full project duration)
Community meetings 271 meetings were held across the two districts, reaching a total of 12,933 people. A total of 9,421 leaflets were disseminated at the meetings. Separate meetings were held per target group. Of the 271 meetings held: • 128 meetings were held for key influential people in the community, reaching 2,840 people • 98 meetings were held for community members, reaching 7,668 people • 12 "Shiv charcha" (religious) meetings were held in Chandauli, reaching 1,429 people • 33 meetings were held for persons affected by leprosy in Fatehpur, reaching 996 persons affected by leprosy.	• Key influential people in the community (teachers, informal practitioners, heads of the village, religious leaders and media personnel) • Community members • Persons affected by leprosy	December 2019 -February 2020

Outcomes

Four outcome measures were used to assess perception: (1) a knowledge, attitudes and practices (KAP) measure; (2) the Explanatory Model Interview Catalogue Community Stigma Scale (EMIC-CSS); (3) the Social Distance Scale (SDS); and (4) an intervention evaluation tool to assess exposure to the posters. In addition, in-depth interviews and focus group discussions were conducted.

The KAP measure was used to assess the knowledge, attitudes and practices of participants regarding leprosy. On some of the questions, multiple answers were possible. A maximum score of eight could be obtained on the KAP if all correct answers are provided, even if incorrect answers were present. We defined 'poor knowledge' as a score of two or less out of eight, 'moderate knowledge' as a score between two and six and 'adequate knowledge' as a score of six or more on the KAP. These cut-offs were chosen arbitrarily, as no external criterion was available. The KAP measure has been used in several leprosy studies in Nepal, India, Indonesia and Brazil between 2012 and 2018 [8,26–28].

The EMIC-CSS was used to measure perceived community attitudes and behavior towards persons affected by leprosy. A total maximum score of 30 can be obtained, ranging from zero (no negative attitudes) to 30 (most negative attitudes). The EMIC-CSS has been validated among community members of persons affected by leprosy in India [29]. We operationalized stigmatizing community attitudes towards leprosy as a sum score of 8 or higher on the EMIC-CSS, using the cut-off point of 8 that was proposed by Sermrittirong and colleagues [30].

The SDS was used to assess the social distance the participant wants to keep towards persons affected by leprosy. This measure was used as a proxy for personal attitudes and fears of the respondent. The SDS has 7 questions on which a maximum score of 21 can be obtained, ranging from zero (no negative attitudes) to 21 (most negative attitudes). The SDS has been translated to Hindi and was partially validated among community members of persons affected by leprosy in Uttar Pradesh, India [26]. We chose a cut-off for negative personal attitudes when participants answered at least 3 questions with 'probably not willing,' or at least one question with 'definitely not willing' and at least one question with 'probably not willing.'

The intervention evaluation tool consisted of questions about exposure to the posters. For example, participants were shown the posters and asked whether they had seen them recently, and participants were asked to identify correct messages about leprosy (read aloud while shown on the posters). The EMIC-CSS and SDS assess community stigma and were therefore not administered to persons affected by leprosy. The intervention evaluation tool was not administered to health workers, because they were not a target group for the posters and meetings.

Semi-structured in-depth interviews and focus group discussions were conducted for insight into specific local beliefs, myths and misconceptions of the participants towards leprosy and persons affected by leprosy. The interview guide was pilot tested before use [8].

Participant timeline

Survey 1 was conducted between March 2017 and December 2018. The outcomes represent the pre-intervention (baseline) information. After finalization of the interventions, dissemination of posters started from October 2019 and onwards, and the community meetings were held between September 2019 and February 2020. The post-intervention or evaluation study (Survey 2) was conducted between March and June 2020. An overview of the study design can be found in Figure 1.

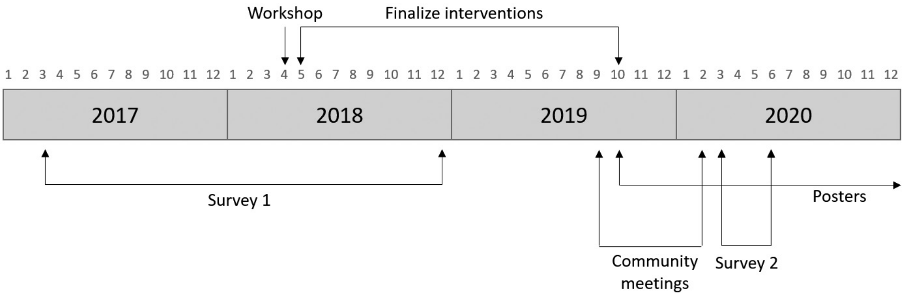

Figure 1. Study design and timeline. Survey 1 consisted of the KAP, EMIC-CSS, SDS, communication needs assessment, in-depth interviews, and focus group discussions. Survey 2 consisted of the KAP, EMIC-CSS, SDS, intervention evaluation tool, in-depth interviews, and focus group discussions.

Sample size

We aimed to include a random sample of at least 100 persons of each target group per district for the interview-administered questionnaires. This is based on an assumed prevalence of, for example, ‘negative attitudes’ of 50% at baseline and wanting to be able to detect a reduction of 20% or more (meaning that the prevalence of negative attitudes in the second survey is 30% or less). Using these parameters, a significance level of 0.05 and a power of 80%, 93 participants are needed before and 93 are needed after the intervention is implemented (calculated using Epi Info StatCalc for cross-sectional studies). To compensate for records that may not be usable or loss to follow up, we aimed to include at least 100 participants per target group. The data of community members for Survey 1 were collected in a separate study but using the same instruments, in the same area and timeframe. Thus, these data were included instead interviewing another sample of community members.

Recruitment and sampling procedure

For pre- and post-intervention assessments, the persons affected by leprosy were selected by stratified systematic sampling with a random start from a list of leprosy patients registered at the primary health care center. Close contacts of leprosy patients and community members were selected by convenience sampling from among those living in the same village or neighborhood as the person affected by leprosy. Health care workers were selected based on convenience sampling from among those present and available at the primary health care centers. Half of the health care workers had received training about leprosy and had specific responsibilities for leprosy treatment services. Details about the selection procedure have been published previously [8,24]. Different (randomly selected) participants were included in the first and second survey.

In addition, in each district we aimed to include six persons from each participant group in the in-depth interviews (IDI) and to conduct one focus group discussion (FGD) per participant group. These participants were a subset of those in the quantitative sample.

Data collection

Data for the perception studies (Survey 1 and Survey 2) were collected before and after the posters were distributed and community meetings were held. In Survey 2, additional demographic information was collected from the participants about income and caste. In addition, Survey 2 data were collected in the areas in which interventions were conducted. Participants were interviewed by a trained research assistant at or near their homes, at primary health centers or the district offices of NLR India. Details of Survey 1 have been published previously [8,24].

Data management

All participants provided informed consent prior to data collection. The hard copy informed consent forms are stored in a locked archive in the field offices of NLR India. Questionnaire data were collected on paper and a Data Entry Officer subsequently entered the responses into a database created in Epi Info. The interviews were recorded on a voice recorder and transcribed in Microsoft Word. The audio files of the interviews were deleted after transcription and data analysis.

Data analysis

Data analysis of the quantitative data were performed in SPSS. No records needed to be excluded from analysis. Simple descriptive methods were used to generate a demographic profile of the study sample. Differences between participants in the first and second survey were evaluated using an independent samples t-test for continuous variables (age) and Chi-square statistics for categorical variables. Corrected median differences and the statistical significance of changes ($p<0.05$) in KAP, EMIC-CSS and SDS scores between Survey 1 (before any intervention) and Survey 2 (after the posters and community meetings) were calculated using quantile regression in which we corrected for age, sex, district, education, religion, participant type and data collection period (Survey 1 or Survey 2). Correlations between exposure to the posters and KAP, EMIC-CSS and SDS scores were calculated using Spearman's rank correlation. Because of the differences in participants in Survey 1 and 2, we could not use a standardized method for effect size. We therefore used the corrected median difference in scale scores between Survey 1 and 2 as percentage of the maximum score that can be obtained on the KAP, EMIC-CSS and SDS to indicate the magnitude of the effect of the interventions.

In addition, we used stepwise multivariate regression with backward elimination to investigate the contribution of potential determinants (age, gender, participant type, marital status, education, occupation, knowing someone affected by leprosy, district and total number of posters seen) to the outcomes of interest (knowledge, stigma, social distance) for dependent variables that were normally distributed. We used bootstrapped stepwise multivariate regression with backward elimination for dependent variables that were not-normally distributed. Only variables that had a p-value of ≤ 0.2 in univariate analysis were considered for entry into the multivariable regression model. Variables were eliminated from the multivariate model one-by-one until only statistically significant variables ($p<0.05$) remained.

The in-depth interviews and focus group discussions were audio recorded, transcribed verbatim and translated from Hindi to English. The data were analyzed using open, inductive coding and content analysis. Qualitative data analyses were performed in the software program MAXQDA. All records were anonymized before analysis.

Results

Socio-demographic information

In total 1067 participants were included in the first survey and 843 participants in the second survey; see Table 2 for an overview of the demographic information of the participants. Statistically significant differences ($p<0.05$) between participants in the first and second survey were found for education level of persons affected by leprosy (participants in the second survey had in general had less education), for gender of health workers (more men were included in the second survey), and for all demographic variables of close contacts and community members.

In addition, after the intervention eight focus group discussions were conducted with 62 participants in total (one focus group with each target group in each district, n=47 male and n=17 female, average age 40 years, range 20-80 years) and 48 in-depth interviews (six per target group in each district, n=25 male and n=23 female, average age 37 years, range 19-58 years)

Table 2. Overview of the demographic characteristics of the participants included in Survey 1 (before any intervention, n=1067) and Survey 2 (after the posters and community meetings, n=842).

Variable	Persons affected by leprosy			Close contacts and community members			Health workers		
	Survey 1 (n=200)	**Survey 2 (n=201)**	***p*-value**[a]	**Survey 1 (n=767)**	**Survey 2 (n=541)**	***p*-value**[a]	**Survey 1 (n=100)**	**Survey 2 (n=101)**	***p*-value**[a]
Age, mean (SD)	39.1 (15.7)	41.9 (16.1)	0.087	40.5 (16.1)	36.7 (13.8)	<0.001	41.8 (11.1)	40.8 (10.7)	0.522
Sex, *n* (%)			0.385			0.021			0.001
Female	77 (38.5)	69 (34.3)		297 (38.7)	244 (45.1)		59 (59.0)	35 (35.0)	
Male	123 (61.5)	132 (63.7)		470 (61.3)	297 (54.9)		41 (41.0)	65 (65.0)	
District, *n* (%)			0.960			<0.001			1.000
From Fatehpur	100 (50.0)	101 (50.2)		296 (38.6)	271 (50.1)		50 (50.0)	50 (50.0)	
From Chandauli	100 (50.0)	100 (49.8)		471 (61.4)	270 (49.9)		50 (50.0)	50 (50.0)	
Education completed, *n* (%)			0.020			<0.001			0.317
No (formal)									
Primary	72 (36.0)	85 (42.3)		207 (27.0)	184 (34.0)		0 (0.0)	0 (0.0)	
Secondary or higher	23 (11.5)	40 (19.9)		104 (13.6)	115 (21.3)		0 (0.0)	1 (1.0)	
	105 (52.5)	76 (37.8)		456 (59.5)	242 (44.7)		100(100.0)	99 (99.0)	
Religion, *n* (%)			0.841			0.001			1.000
Hinduism	184 (92.0)	186 (92.5)		687 (89.6)	512 (94.6)		99 (99.0)	99 (99.0)	
Islam	16 (8.0)	15 (7.5)		72(9.4)	28 (5.2)		1 (1.0)	1 (1.0)	
Other	0 (0.0)	0 (0.0)		8 (1.0)	1 (0.2)		0 (0.0)	0 (0.0)	
Marital status, *n* (%)			0.159			<0.001			0.623
Currently married	150 (75.0)	162 (80.6)		313 (40.8)	425 (78.6)		91 (91.0)	89 (89.0)	
Never married	38 (19.0)	32 (15.9)		74 (9.6)	97 (17.9)		9 (9.0)	10 (10.0)	
Other[b]	12 (6.0)	7 (3.5)		9 (11.7)	19 (3.5)		0 (0.0)	1 (1.0)	
Missing	0 (0.0)	0 (0.0)		371 (48.4)	0 (0.0)		0 (0.0)	0 (0.0)	

[a] The p-value is based on independent samples t-test for continuous variables (age) and X2 statistics for categorical variables.

[b] Marital status ‘other’ refers to participants who are separated, divorced or widowed.

Overall impact of the interventions: difference between Survey 1 and 2

In Survey 1 the percentage of participants with adequate knowledge on the KAP measure was 13% (n=133; 13% of the persons affected by leprosy, 7% of the contacts and community members, 56% of the health workers). In Survey 2, 53% (n=448) of the participants had adequate knowledge of leprosy (78% of the persons affected by leprosy, 38% of the contacts and community members, 87% of the health workers). An overview of the distribution of the KAP measure scores in Survey 1 and 2 of the contacts and community group can be found in Figure 2. In addition, in Survey 1, 86% of participants had stigmatizing attitudes towards leprosy on the EMIC-CSS (n=747; 86% of the contacts and community members, 84% of the health workers). In Survey 2, this was 61% (n=393; 65% of the contacts and community members, 43% of the health workers). In addition, 37% of participants had negative personal attitudes on the SDS in Survey 1 (n=325; 41% of the contacts and community members, 14% of the health workers) and 19% in Survey 2 (n=121; 22% of the contacts and community members, 2% of the health workers).

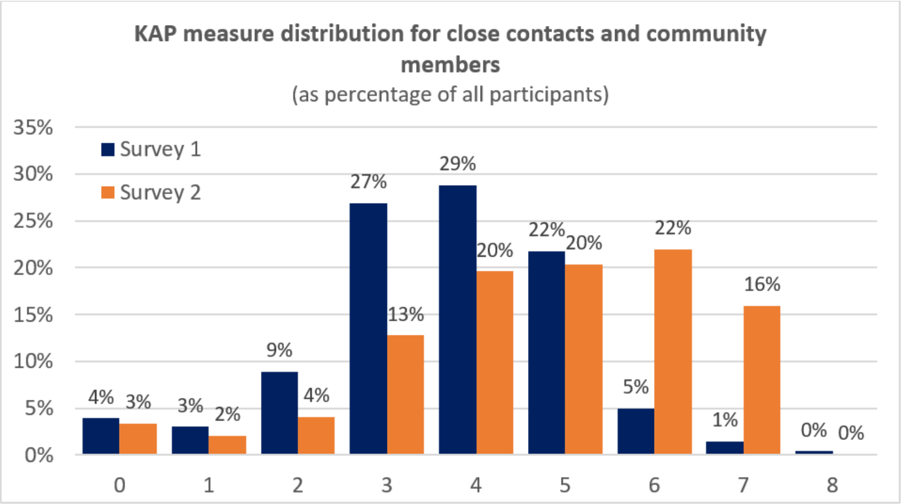

Figure 2. Distribution of the KAP measure scores for close contacts and community members in Survey 1 and Survey 2.

Tables 4 and 5 provide an overview of the differences in KAP, EMIC-CSS and SDS scores between Survey 1 and Survey 2 per district and per participant group. Compared to Survey 1, almost all KAP, EMIC-CSS and SDS scores improved. The scores that did not improve statistically significantly were the EMIC-CSS score for health workers in Fatehpur and the KAP and SDS scores for health workers in Chandauli (Table 4). When looking at the corrected median differences for all participants groups (the ‘whole dataset’ rows in Tables 4 and 5), the corrected median difference was 1.5 for the KAP (a change of 19% of the maximum score of 8 that can be obtained on the scale), -7.3 for the EMIC-CSS (a change of 24% of the total score of 30 that can be obtained) and -2.0 for the SDS (a change of 10% of the total score of 21 that can be obtained). The largest corrected median difference on the KAP was found for persons affected by leprosy (corrected median difference 3.0, 38% of total score, p<0.001). With corrected median differences of -10.0 and -10.3 (33-34% of the total score), the corrected median differences for the EMIC-CSS were larger in Chandauli district than in Fatehpur district. The largest corrected median differences (-3.0 and -2.9 or 14% of the total score of 21) for the SDS were found among contacts and community members in Chandauli and among health workers in Fatehpur (Table 3).

Table 3. Corrected median differences in KAP (range 0-8) scores between Survey 1 (n=1067) and Survey 2 (n=842).

Dataset	KAP measure				
	Survey 1, median (Q1-Q3)	Survey 2, median (Q1-Q3)	Corrected median difference[a]	Change in score[b]	p-value[c]
Whole dataset (n=1067~842)	**4.0 (3.0-5.0)**	**6.0 (4.0-7.0)**	**1.5**	**18.8%**	**<0.001**
Persons affected by leprosy (n=200~201)	4.0 (3.0-5.0)	7.0 (6.0-8.0)	3.0	37.5%	<0.001
Contacts and community (n=767~541)	4.0 (3.0-5.0)	5.0 (4.0-6.0)	1.0	12.5%	<0.001
Health workers (n=100~100)	6.0 (5.0-7.0)	7.0 (6.0-7.0)	1.0	12.5%	<0.001
Chandauli district (n=621~420)	**4.0 (3.0-5.0)**	**6.0 (5.0-7.0)**	**2.0**	**25.0%**	**<0.001**
Persons affected by leprosy (n=100~100)	3.0 (3.0-4.0)	6.0 (5.0-7.0)	3.0	37.5%	<0.001
Contacts and community (n=471~270)	4.0 (3.0-5.0)	5.0 (4.0-6.0)	2.0	25.0%	<0.001
Health workers (n=50~50)	6.0 (5.0-7.0)	7.0 (7.0-7.0)	0.0	0.0%	NS
Fatehpur district (n=446~422)	**4.0 (3.0-5.0)**	**5.0 (4.0-7.0)**	**1.3**	**16.3%**	**<0.001**
Persons affected by leprosy (n=100~101)	4.0 (3.0-5.0)	7.0 (6.0-8.0)	3.0	37.5%	<0.001
Contacts and community (n=296~271)	4.0 (3.0-5.0)	4.0 (3.0-6.0)	1.0	12.5%	<0.001
Health workers (n=50~50)	5.5 (4.0-6.3)	7.0 (6.0-7.0)	1.0	12.5%	0.014

[a] We corrected (adjusted) for age, sex, district, education, religion, participant type and data collection period (Survey 1 or Survey 2). Quantile regression models can be found in S3 Text.

[b] The corrected median difference as percentage of the maximum score that can be obtained on the scale.

[c] The p-value was calculated using quantile regression in which we corrected for differences in demographic information between the participants in Survey 1 and Survey 2. NS = not significant (p>0.05).

Table 4. Corrected median differences in EMIC-CSS (range 0-30) and SDS (range 0-21) scores between Survey 1 (n=867) and Survey 2 (n=641).

Dataset	EMIC-CSS					SDS				
	Survey 1, median (Q1-Q3)	Survey 2, median (Q1-Q3)	Corrected median difference[a]	Change in score[b]	p-value[c]	Survey 1, median (Q1-Q3)	Survey 2, median (Q1-Q3)	Corrected median difference[a]	Change in score[b]	p-value[c]
Whole dataset (n=1067~842)	**17.0 (11.0-21.0)**	**9.0 (5.0-14.0)**	**-7.3**	24.3%	**<0.001**	**5.0 (3.0-10.0)**	**3.0 (1.0-6.0)**	**-2.0**	9.5%	**<0.001**
Contacts and community (n=767~541)	17.0 (11.0-21.0)	9.0 (6.0-14.0)	-7.3	24.3%	<0.001	6.0 (3.0-10.0)	4.0 (2.0-7.0)	-2.7	12.9%	<0.001
Health workers (n=100~100)	15.0 (10.0-21.8)	6.0 (3.0-13.0)	-4.3	14.3%	<0.001	2.0 (0.0-6.0)	0.0 (0.0-2.0)	-1.0	4.8%	0.044
Chandauli district (n=621~420)	**18.0 (11.0-22.0)**	**7.0 (5.0-9.0)**	**-10.0**	33.3%	**<0.001**	**5.0 (2.0-9.0)**	**3.0 (1.0-4.0)**	**-2.0**	9.5%	**<0.001**
Contacts and community (n=471~270)	18.0 (12.0-22.0)	8.0 (5.8-10.0)	-10.0	33.3%	<0.001	5.0 (3.0-10.0)	3.0 (2.0-5.0)	-3.0	14.3%	<0.001
Health workers (n=50~50)	15.0 (8.8-23.3)	4.0 (2.0-6.0)	-10.3	34.3%	<0.001	1.0 (0.0-3.3)	0.0 (0.0-1.0)	0.0	0.0%	NS
Fatehpur district (n=446~422)	**15.0 (11.0-20.0)**	**14.0 (7.0-19.0)**	**-1.7**	5.7%	**0.029**	**6.0 (3.0-10.0)**	**4.0 (0.0-9.0)**	**-2.3**	11.0%	**<0.001**
Contacts and community (n=296~271)	15.0 (10.3-21.0)	14.0 (7.0-19.0)	-2.0	6.7%	0.018	6.0 (3.0-11.0)	5.0 (1.0-10.0)	-2.3	11.0%	<0.001
Health workers (n=50~50)	14.5 (11.0-20.0)	13.0 (6.0-18.5)	-1.6	5.3%	NS	3.5 (1.0-6.0)	0.0 (0.0-3.0)	-2.9	13.8%	<0.001

[a] We corrected (adjusted) for age, sex, district, education, religion, participant type and data collection period (Survey 1 or Survey 2). Quantile regression models can be found in S3 Text.

[b] The corrected median difference as percentage of the maximum score that can be obtained on the scale

[c] The p-value was calculated using quantile regression in which we corrected for differences in demographic information between the participants in Survey 1 and Survey 2. NS = not significant (p>0.05).

Multivariate analysis showed that the determinants of leprosy knowledge and community stigma were comparable for Survey 1 and Survey 2. In Survey 2, income, caste and 'having seen posters' were also included in the models. 'Having seen posters' alone had a larger effect on the KAP scores (R-squared=0.15, univariate analysis) than on the EMIC-CSS and SDS scores (R-squared=0.05 and 0.06 respectively, univariate analysis). The models for Survey 2 explained more of the variability of knowledge and stigma (Table 5). An overview of the full multivariate regression models can be found as supporting information file S4 Text.

Table 5. Correlations between level of knowledge (KAP score) about leprosy, community stigma (EMIC-CSS), social distance (SDS) and the other variables in the dataset including data of persons affected by leprosy, close contacts and community members. Participant type and district were included in all models to control for confounding.

Questionnaire	**Survey 1***		**Survey 2****	
	Variables included in the model	**R-squared**	**Variables included in the model**	**R-squared**
KAP measure (knowledge of leprosy)	*(Participant type, district)* No (formal) education Higher education	0.054	*(Participant type, district)* No (formal) education Higher education Income less than 5,000 Has seen PEP++ posters	0.355
EMIC-CSS (community stigma)	*(Participant type, district)*	0.105	*(Participant type, district)* Primary education Knowledge about leprosy (KAP) Income less than 1,000 Has seen PEP++ posters	0.292
SDS (social distance as a proxy for attitudes)	*(Participant type, district)* No (formal) education Gender Knowledge about leprosy (KAP)	0.050	*(Participant type, district)* No (formal) education Primary education Knowledge about leprosy (KAP) Occupation paid work Income 5,001 to 10,000 Has seen PEP++ posters	0.232

***** Variables included: participant type, district, age, gender, education, occupation, and for the EMIC-CSS and SDS also 'KAP score' and 'knowing someone affected by leprosy'.

** Variables included: participant type, district, age, gender, education, occupation, marital status, monthly household income, caste, having seen PEP++ posters, and for the EMIC-CSS and SDS also 'KAP score' and 'knowing someone affected by leprosy'.

In the in-depth interviews and focus group discussions, many participants indicated that there had been a change in perception in the community. Some participants indicated that people in the community used to believe something, but not anymore. Some participants related this change in perception of the community to (knowing about) preventive medication. One close contact explained:

> *"...Earlier people used to behave [negative] like this, now people have started understanding that untouchability does not happen [referring to transmission]..."*
> (30-year old close contact, male, FGD, Chandauli)

Furthermore, the transcripts of the interviews and focus group discussions revealed that over half of the participants knew that leprosy is caused by bacteria, and almost all participants mentioned loss of sensation and/or skin patches as early symptoms of leprosy and knew leprosy can be treated with medication. Knowledge about treatment, cause and symptoms was good. There were still some misconceptions regarding the cause of leprosy. For example, some participants thought leprosy is caused by a blood or vitamin deficiency, dirt or being unclean.

Approximately half of the participants indicated that (some) community members discriminate or keep a distance from persons affected by leprosy. Most participants said these community members behave this way because they have incorrect or insufficient knowledge about leprosy or because they are afraid of getting infected by the disease themselves. One participant explained:

> *"...Most people discriminate because they do not know about this disease, they feel that it is an untouchable disease, whereas this is not true...."*
> (58-year-old close contact, male, IDI, Chandauli)

Some participants stressed that community members only discriminate if leprosy is visible or if persons affected by leprosy are not treated. Approximately a quarter of all participants said that there is no discrimination, that community members behave well or normal towards persons affected by leprosy. Many of the participants who mentioned there is no discrimination, also mentioned that they advise persons affected by leprosy to get treatment. A few participants explicitly stated that they don't think persons affected should be discriminated.

> *"...They [community] behave differently, like talking with them [persons affected by leprosy] by keeping a distance etc. According to me, this is wrong, there should be no discrimination against them..."*
> (51-year-old community member, female, IDI, Fatehpur)

Another participant explained:

> *"...People of the community do not discriminate. Everyone sits together and tells [the person affected by leprosy] to get treatment for leprosy..."*
> (32-year-old close contact, male, IDI, Chandauli)

Finally, almost all participants had heard about post-exposure prophylaxis (PEP) and the PEP++ project. Everyone who knew about PEP had a positive attitude towards it. Many participants indicated that knowing about PEP and the possibility of PEP positively changed the perception of leprosy. A person affected by leprosy explained:

> *"…There is a change in thinking that now if you take [preventive] medicine before [you have symptoms] then there will be no disease. The medicine that prevents leprosy is a very good idea…"*
>
> (19-year-old person affected by leprosy, female, IDI, Chandauli)

Impact of the posters

Most participants (health care workers excluded) indicated they had seen a poster in the villages (34%, n=287) at the health facility (26%, n=220) or in public transport (10%, n=84). Almost two-third of the participants were able to identify at least one poster (61%, n=482). Participants correctly identified two posters on average. A total of 305 participants (36.2%) had not seen any poster and 196 participants (23.3%) had seen all five posters.

Persons affected by leprosy, close contacts and community members were also shown five posters and asked if they had seen them. Between 38% (n=281, fifth poster) and 48% (n=353, first poster) of the participants indicated that they had seen one of the five posters. Participants in Fatehpur (n=369) were also shown a poster that was never used and asked whether they had seen it. Two participants (0.5%) thought they had seen this poster and 367 participants (99.5%) said they had not seen the poster. Figure 3 gives an overview of the number of posters the participants had seen and their mean KAP, EMIC-CSS and SDS scores. There was an association between the number of posters seen and the KAP (n=738, rho=0.389, $p<0.001$), EMIC-CSS (n=539, rho=-0.208, $p<0.001$) and SDS (n=539, rho=-0.203, $p<0.001$) scores in Survey 2.

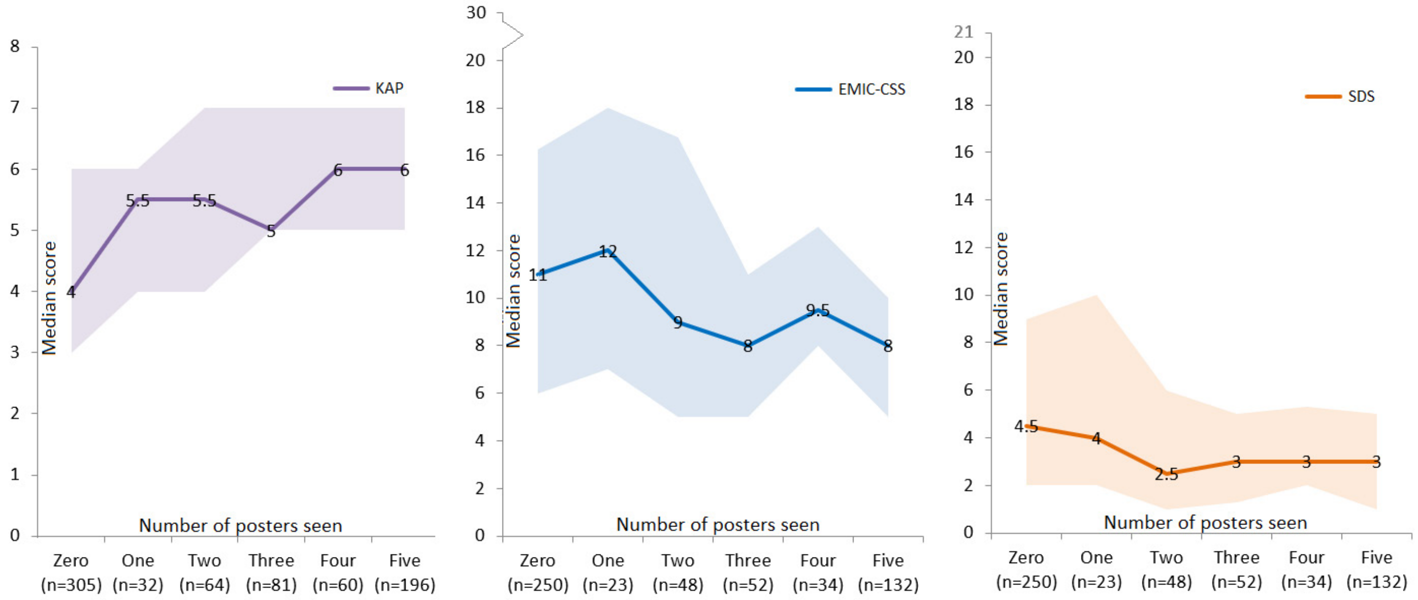

Figure 3. An overview of the number of posters seen and the corresponding median KAP (knowledge, ranging from 0-8 with higher scores denoting better knowledge), EMIC-CSS (community attitudes, ranging from 0-30 with higher scores denoting more negative attitudes) and SDS scores (personal attitudes, ranging from 0-21 with higher scores denoting more negative attitudes) and corresponding first and third quartiles. Please note that the graphs have different y-axes.

Discussion

Salient results

Findings from this study suggest that the contextualized posters and community meetings were effective in increasing knowledge of leprosy (a change of 19% of the maximum KAP score), in changing community attitudes (a change of 24% on the maximum EMIC-CSS score), and in changing personal attitudes (a change of 10% on the maximum SDS score) of all participant groups. In addition, when we used a cut-off point to determine adequate knowledge of leprosy and stigmatizing attitudes towards leprosy, the percentage of participants with adequate knowledge of leprosy was 13% in Survey 1 and 53% in Survey 2, and the percentage of participants with stigmatizing attitudes was 86% in Survey 1 and 61% in Survey 2, and the percentage of participants with negative personal attitudes was 37% in Survey 1 and 19% in Survey 2. We consider the effect high for knowledge of leprosy and community attitudes, and moderate for personal attitudes. It is likely that the change in community attitudes was greater than in personal attitudes, because the social distance score (SDS, personal attitudes) was already relatively low in Survey 1, so could decrease less.

The largest effect of the whole package of interventions was on knowledge of leprosy of persons affected by leprosy (a change of 38% of the maximum score) and on community attitudes of contacts and community members (a change of 24% of the maximum score). From other studies, we know that it is easier to improve knowledge than to change behavior [31,32], our findings are therefore very encouraging. It is possible that the effect of our interventions is an underestimation, because the post-intervention participants had lower education levels than the pre-intervention participants. This could have influenced our results, since less education was associated with less knowledge about leprosy and more negative attitudes.

The smallest effect of the whole package of interventions was seen among knowledge and personal attitudes of health workers. It is likely that the effect on health workers was smaller because they were not specifically targeted with the posters and community meetings. In addition, before the implementation of the interventions their knowledge of leprosy was already better and their attitudes more positive compared to the other participants in our study. Their scores could therefore increase (knowledge) or decrease (stigma) less. Health workers were asked to support and be involved in preparatory activities for the PEP++ project, such as finding houses of persons affected by leprosy and giving feedback on the posters, and they were informed about the project and preventive medication. In addition, they were exposed to the posters at the health centers daily. We assume that this involvement and exposure has positively influenced their perceptions.

Surprisingly, the effect of the total package of interventions on community stigma was larger in Chandauli district than in Fatehpur district. It is possible that the difference can partly be explained by the religious meetings featuring local artists (Shiv charcha) that were held only in Chandauli district in addition to the community meetings. In addition, according to the district teams, media exposure of the project and government interest and commitment were greater in Chandauli district. In Chandauli district, 52 news articles covered announcements of the community meetings and a brief explanation of the project, while this was covered in 26 news articles in Fatehpur district. The way persons affected are portrayed in the media reflects, defines, and perpetuates public perceptions of those who are portrayed [33]. In other fields, while media have been found to be a source of stigma through the negative portrayal of persons affected, they have also been found to reduce stigma by raising awareness (as was done in our study) [34–36]. However, we cannot offer a definitive explanation of the difference the interventions had on community stigma between the two districts.

From the exploratory and other studies, we know that knowledge about leprosy plays a crucial role in stigma [8,37–40]. While knowledge gaps can be addressed by information, to change attitudes and perceptions is more difficult and requires a combination of health education and behavioral change interventions [41,42]. Good knowledge of leprosy does not necessarily lead to more positive attitudes toward persons affected by leprosy [12]. Interestingly, the determinants of leprosy knowledge (education, income, exposure to posters) and community stigma (education, leprosy knowledge, income, occupation, exposure to posters) in the present study were similar before and after the interventions had been implemented. We collected additional information in the post-intervention measurement: income, caste and 'having seen posters' and included this in our analysis. The post-intervention models explained more of the variability of knowledge and stigma. This is in part explained by the effect of income and exposure to the posters.

In the present study there was a positive association between the number of posters seen and the level of knowledge and positive attitudes towards persons affected by leprosy. The more posters participants indicated to have seen, the better their knowledge and the more positive their attitudes. However, since we didn't assess the knowledge and attitudes of the participants in the second survey before exposure to the posters, it is uncertain that the impact can be attributed to the posters. Nevertheless, the correlation with the number of posters seen is strongly suggestive of such an effect. Beliefs about leprosy are often deeply rooted in people's culture [12]. To address this, we focused on local beliefs and misconceptions and have consulted the target populations in selecting the interventions and developing the posters. Printed media like posters, billboard and leaflets have been used to increase community awareness and reduce leprosy-related stigma in other studies also, but their impact has not been evaluated rigorously [16,43–45]. Although written materials, like posters and leaflets, are not the most suitable approach for populations with low educational levels [46], the contextualization and careful pretesting of the imagery used appears to have resulted in a positive impact on community knowledge and perception in the study.

Methodological considerations

A key feature of the interventions in the present study is that they are contextualized, relatively low-cost and easy to replicate. We ensured that the materials and messages were targeted and contextualized and that relevant topics were prioritized, e.g., cause, mode of transmission, symptoms and infectiousness of leprosy. Contextualized materials and messages are more effective than generic messages [47]. In addition, community consultation and involvement was used - this is more episodic community participation, in contrast to, for example, community engagement, which suggests an ongoing and active relationship [11]. The interventions were developed through collaborations and consultations between the target population, including persons affected by leprosy, researchers, health workers, leprosy experts, communication experts and policymakers. This maximizes the likelihood that the interventions fit with the target groups' needs and acceptability, and the uptake of the interventions by policymakers [23].

Several successful community-based stigma reduction interventions have been conducted in the field of leprosy, all including elements of community participation or engagement, such as informing, consulting, involving, collaborating and empowering communities [10,11]. These studies have shown encouraging results. Successful community-based stigma reduction interventions in the field of leprosy include education and counselling through (community) stigma reduction committees [9], stigma reduction interventions through groups of health workers, volunteers and persons affected in self-help groups [8], and rights-based counselling and contact events [4]. Indeed, community participation and engagement can ensure that research is relevant and impactful. Community engagement has been successful for control and elimination of other diseases also, such as malaria [12,13]. In the present study, in addition to the community's involvement in the development of the interventions, the community was also consulted and discussions were held in community meetings for the purpose of education and changing negative attitudes regarding leprosy. Efforts were made to ensure community and health worker engagement in the interventions, by consultation and by involving them in preparatory meeting.

We were not able to determine the actual change in behavior, given that there are no suitable measures to assess this. However, we measured knowledge, attitude and perceived practices and these measures in part reflect actual behavior (e.g., a person's perception of their behavior in a given situation), and our qualitative data show reflections of participants, indicating an actual change in perception and behavior. We therefore conclude that it is likely that in addition to a change in perceptions, there was likely also a change in behavior after the interventions. We would recommend that future studies explore meaningful ways to assess actual changes in behavior and indicators of behavior change, for example by asking persons affected by leprosy about their experiences at health facilities.

A novel feature of our study is that we determined and used a cut-off point for negative personal attitudes on the SDS, which was not yet available. A cut-off point for positive/negative attitudes is important, because it helps readers and practitioners to interpret the findings. It can also be used to estimate the magnitude of (meaningful) effect.

This study has several limitations. First, a randomized controlled design was not feasible given the nature of our intervention, namely, community-wide poster dissemination and meetings. Instead, we used a pre/post intervention design. It is possible that not all of the observed changes are due to the interventions – some changes may have been caused by other factors. We tried to minimize this by selecting a random sample. However, we cannot rule out other factors that may have contributed to the outcomes observed. While it is unclear how much of the effect found can be attributed to the interventions, given our findings it is very likely that the interventions have contributed to the outcomes. Second, there were differences between sociodemographic characteristics of the pre- and post-intervention participant groups. This was especially the case for close contacts and community members, with post-intervention participants having lower education levels. We have corrected for these differences in our analysis, but because of this, we were unable to use a standardized measure of effect size. This made it more difficult to determine the magnitude of the effect of the interventions. We recommend separately evaluating each element of an intervention in future studies (instead of the whole package of interventions), to gain a better understanding of the impact of each element. Finally, it would have been interesting if we had evaluated the impact of the community meetings on knowledge and perceptions of influential people specifically, they were not included as a separate target group in the surveys (evaluation) of the interventions.

Conclusions

The contextualized posters and community meetings in this study were effective in increasing leprosy-related knowledge and changing perceptions of leprosy in Fatehpur and Chandauli districts in Uttar Pradesh, India. The interventions in this study are relatively low-cost and are easy to replicate. Given that changing attitudes and perceptions is difficult and generally requires a combination of health education and behavioral change interventions, the results are very encouraging. Future studies should explore meaningful ways to assess actual changes in behavior and indicators of behavior change. In addition, the long-term effect of the interventions should be studied.

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References

1. Pickens J. Attitudes and perceptions. Organ Behav Heal Care 2005;4.
2. Hiebert PG. Transforming worldviews: An anthropological understanding of how people change. Baker Academic; 2008.
3. Broadbent E, Wilkes C, Koschwanez H, Weinman J, Norton S, Petrie KJ. A systematic review and meta-analysis of the Brief Illness Perception Questionnaire. Psychol Health 2015;30:1361–85.
4. Link BG, Struening EL, Rahav M, Phelan JC, Nuttbrock L. On stigma and its consequences: evidence from a longitudinal study of men with dual diagnoses of mental illness and substance abuse. J Health Soc Behav 1997:177–90.
5. Thornicroft G, Rose D, Kassam A, Sartorius N. Stigma: ignorance, prejudice or discrimination? Br J Psychiatry 2007;190:192–3.
6. Weiss MG, Ramakrishna J, Somma D. Health-related stigma: rethinking concepts and interventions. Psychol Health Med 2006;11:277–87.
7. Sermrittirong S, Van Brakel W. Stigma in leprosy: concepts, causes and determinants. Lepr Rev 2014;85:36–47.
8. van't Noordende AT, Korfage I, Lisam S, Arif MA, Kumar A, van Brakel WH. The role of perceptions and knowledge of leprosy in the elimination of leprosy: A baseline study in Fatehpur district, northern India. PLoS Negl Trop Dis 2019;13:e0007302.
9. Link BG, Phelan JC. Stigma and its public health implications. Lancet 2006;367:528–9.
10. Link BG, Phelan JC. Conceptualizing stigma. Annu Rev Sociol 2001;27:363–85.
11. Dadun D, Van Brakel WH, Peters RMH, Lusli M, Zweekhorst M, Bunders JGF, et al. Impact of socio-economic development, contact and peer counselling on stigma against persons affected by leprosy in Cirebon, Indonesia–a randomised controlled trial. Lepr Rev 2017;88:2–22.
12. Wong ML. Guest editorial: designing programmes to address stigma in leprosy: issues and challenges. Asia Pacific Disabil Rehabil J 2004;15:3–12.
13. Sermrittirong S, van Brakel WH, Bunders-Aelen JFG. How to reduce stigma in leprosy–a systematic literature review. Lepr Rev 2014;85:149–57.
14. Ebenso B, Fashona A, Ayuba M, Idah M, Adeyemi G, S-Fada S. Impact of socio-economic rehabilitation on leprosy stigma in Northern Nigeria: findings of a retrospective study. Asia Pacific Disabil Rehabil J 2007;18:98–119.
15. Sermrittirong S, Van Brakel WH, Bunders-Aelen JFG, Ariyothai N, Chaidee N. Addressing stigma related to leprosy: lessons from an intervention study in Thailand. Int J Trop Dis Heal 2014:48–63.
16. Raju MS, Rao PSS, Mutatkar RK. A study on community-based approaches to reduce leprosy stigma in India. Indian J Lepr 2008;80:267–73.
17. Parker R, Aggleton P. HIV and AIDS-related stigma and discrimination: a conceptual framework and implications for action. Soc Sci Med 2003;57:13–24.
18. Rao PS. Study on differences and similarities in the concept and origin of leprosy stigma in relation to other health-related stigma. Indian J Lepr 2010;82:117–21.
19. Birbeck G. Interventions to reduce epilepsy-associated stigma. Psychol Health Med 2006;11:364–6.

20. Opala J, Boillot F. Leprosy among the Limba: illness and healing in the context of world view. Soc Sci Med 1996;42:3–19.
21. Chen PC, Sim HC. The development of culture-specific health education packages to increase case-finding of leprosy in Sarawak. Southeast Asian J Trop Med Public Health 1986;17:427–32.
22. van't Noordende AT, Lisam S, Ruthindartri P, Sadiq A, Singh V, Arifin M, et al. Leprosy perceptions and knowledge in endemic districts in India and Indonesia: Differences and commonalities. PLoS Negl Trop Dis 2021;15:e0009031.
23. Wight D, Wimbush E, Jepson R, Doi L. Six steps in quality intervention development (6SQuID). J Epidemiol Community Heal 2016;70:520–5.
24. van't Noordende AT, Lisam S, Ruthindartri P, Sadiq A, Singh V, Arifin M, et al. Leprosy perceptions and knowledge in endemic districts in India and Indonesia: differences and commonalities. PLoS Negl Trop Dis 2021;15:e0009031.
25. van't Noordende AT, van Brakel WH. Towards a cross-NTD perception study toolkit: a prototype toolkit and implementation protocol developed in the field of leprosy. Submitt Publ Lepr Rev 2020.
26. Ballering A V, Peters RMH, Waltz MM, Arif MA, Mishra CP, van Brakel WH, et al. Community stigma and desired social distance towards people affected by leprosy in Chandauli District, India. Lepr Rev 2019;90:418–32.
27. Mieras L, Singh MK, Manglani PR, Arif MM, Banstola NL, Pandey B, et al. A single-dose of rifampicin to prevent leprosy; quantitative analysis of impact on perception, attitudes and behaviour of persons affected, contacts and community members towards leprosy in India, Nepal and Indonesia. Lepr Rev 2020;91:314–27.
28. Peters R, Mieras L, Subedi M, Apte H, Koesbardiati T, Banstola NL, et al. A single dose of rifampicin to prevent leprosy: qualitative analysis of perceptions of persons affected, contacts, community members and health professionals towards chemoprophylaxis and the impact on their attitudes in India, Nepal and Indonesia. Lepr Rev 2018;89:335–52.
29. Rensen C, Bandyopadhyay S, Gopal PK, Van Brakel WH. Measuring leprosy-related stigma–a pilot study to validate a toolkit of instruments. Disabil Rehabil 2011;33:711–9.
30. Sermrittirong S, Van Brakel WH, Kraipui N, Traithip S, Bunders-Aelen JF. Comparing the perception of community members towards leprosy and tuberculosis stigmatization. Lepr Rev 2015;86:54–61.
31. Bay JL, Mora HA, Sloboda DM, Morton SM, Vickers MH, Gluckman PD. Adolescent understanding of DOHaD concepts: a school-based intervention to support knowledge translation and behaviour change. J Dev Orig Health Dis 2012;3:469–82.
32. Barr RG, Barr M, Fujiwara T, Conway J, Catherine N, Brant R. Do educational materials change knowledge and behaviour about crying and shaken baby syndrome? A randomized controlled trial. Cmaj 2009;180:727–33.
33. Susman J. Disability, stigma and deviance. Soc Sci Med 1994;38:15–22.
34. Chapple A, Ziebland S, McPherson A. Stigma, shame, and blame experienced by patients with lung cancer: qualitative study. Bmj 2004;328:1470.
35. Bekalu MA, Eggermont S, Ramanadhan S, Viswanath K. Effect of media use on HIV-related stigma in sub-Saharan Africa: A Cross-Sectional Study. PLoS One 2014;9:e100467.
36. Niederkrotenthaler T, Reidenberg DJ, Till B, Gould MS. Increasing help-seeking and referrals for individuals at risk for suicide by decreasing stigma: the role of mass media. Am J Prev Med 2014;47:S235–43.

37. Adhikari B, Shrestha K, Kaehler N, Raut S, Chapman SR. Community attitudes towards leprosy affected persons in Pokhara municipality of western Nepal. J Nepal Health Res Counc 2014.
38. van den Broek J, O'Donoghue J, Ishengoma A, Masao H, Mbega M. Evaluation of a sustained 7-year health education campaign on leprosy in Rufiji District, Tanzania. Lepr Rev 1998;69: 57–74.
39. Singh R, Singh B, Mahato S. Community knowledge, attitude, and perceived stigma of leprosy amongst community members living in Dhanusha and Parsa districts of Southern Central Nepal. PLoS Negl Trop Dis 2019;13:e0007075.
40. Seshadri D, Khaitan BK, Khanna N, Sagar R. The tangled web: a study of knowledge and attitude towards leprosy from a tertiary care hospital in India. Indian J Lepr 2014:27–41.
41. Seligman HK, Wallace AS, DeWalt DA, Schillinger D, Arnold CL, Shilliday BB, et al. Facilitating behavior change with low-literacy patient education materials. Am J Health Behav 2007;31:S69–78.
42. Noar SM, Benac CN, Harris MS. Does tailoring matter? Meta-analytic review of tailored print health behavior change interventions. Psychol Bull 2007;133:673.
43. Organization WH. Leprosy elimination campaigns: Detecting and curing patients. Wkly Epidemiol Rec Relev Épidémiologique Hebd 1999;74:329–34.
44. Friedericks RC. A multi-media approach to health education. Lepr Rev 1991;62:329–32.
45. Ukpe IS. Educational posters and leaflets on leprosy: raising awareness of leprosy for health-care workers in rural South Africa. Public Health Rep 2008;123:217–21.
46. Gilder ME, Moo P, Hashmi A, Praisaengdet N, Wai K, Pimanpanarak M, et al. "I can't read and don't understand": Health literacy and health messaging about folic acid for neural tube defect prevention in a migrant population on the Myanmar-Thailand border. PLoS One 2019;14:e0218138.
47. Brown W. Can social marketing approaches change community attitudes towards leprosy? Lepr Rev 2006;77:89–98.

CHAPTER 8

A family-based intervention for prevention and self-management of disabilities due to leprosy, podoconiosis and lymphatic filariasis in Ethiopia: A proof of concept study

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Abstract

A key issue for persons with leprosy-, lymphatic filariasis- and podoconiosis-related disabilities is the life-long need to practice self-management routines. This is difficult to sustain without regular encouragement and support of others. Family-based support may be a sustainable and feasible strategy to practice self-management routines. This proof of concept study aimed to develop and pilot a family-based intervention to support prevention and self-management of leprosy, lymphatic filariasis and podoconiosis-related disabilities in Ethiopia.

We used a quasi-experimental pre/post intervention study design with a mixed methods approach. The study population included persons affected by leprosy, lymphatic filariasis and podoconiosis and their family members. All persons affected had visible impairments due to their condition. We collected physical impairment outcomes, data on activity limitations, stigma and family quality of life using the SALSA scale (range 0-80), the SARI stigma scale (range 0-63) and the Beach Centre Family Quality of Life scale (range 0-125) and conducted in-depth interviews and focus group discussions. Quantitative data were analysed using paired t-tests, unequal variances t-tests, linear regression and binary logistic regression. Qualitative data were coded using open, inductive coding and content analysis.

The family-based intervention consisted of self-management of disabilities, awareness raising and socio-economic empowerment. The intervention was delivered over several monthly group meetings over the course of several months. A total of 275 (100%) persons affected attended at least one session with a family member, and 215 (78%) attended at least three sessions. There was no significant improvement in eye and hand problems after the intervention. However, foot and leg impairments, number of acute attacks, lymphedema and shoe wearing all significantly improved at follow-up. In addition, family quality of life significantly improved from 67.4 at baseline to 89.9 at follow-up for family members and from 76.9 to 84.1 for persons affected ($p<0.001$). Stigma levels significantly decreased from 24.0 at baseline to 16.7 at follow-up ($p<0.001$). Activity levels improved, but not significantly.

This proof of concept study showed that the family-based intervention had a positive effect on impairments and self-management of disabilities, family quality of life and stigma. We recommend a large-scale efficacy trial, using a randomised controlled trial and validated measurement tools, to determine its effectiveness and long-term sustainability.

Introduction

Leprosy, lymphatic filariasis (LF) and podoconiosis are three skin-related Neglected Tropical Diseases (NTDs) [1]. All three conditions exhibit cutaneous manifestations such as patches and ulcers (leprosy), swollen limbs (LF and podoconiosis) and wounds, nodules or swelling (all three) [2–5]. If not diagnosed and treated early, all three conditions can lead to temporary and permanent impairments [1,2,6].

Leprosy, LF and podoconiosis-related impairments are major determinants of stigma and participation restrictions [7–9]. Stigma and physical impairments may also hamper people's daily functioning, for example their ability to work. This may deteriorate the economic situation of persons affected and may impose a social and economic burden on already marginalized families [5,10–13]. This while most impairments, particularly visible impairments such as wounds, swelling and contractures, are largely preventable. Relatively simple methods exist for self-management of impairments that can be practiced at home, without the need for a lot of medical supplies. Many of these methods for prevention and self-management of disabilities are suitable for use across different skin-related NTDs [14,15]. Too often, however, these methods are not taught to patients with neuropathic limbs or lymphedema, or if taught, they are not consistently practiced. Good self-care management practices are crucial to prevent further impairments, reduce symptoms, preserve quality of life and improve the ability to participate in work and social activities [16,17].

A key issue for persons with leprosy-, LF- and podoconiosis-related disabilities is the life-long need to practice such self-management routines. This is difficult to sustain without regular encouragement and support of others. A strategy shown to be successful is the formation of self-care groups in which persons affected by leprosy, LF and podoconiosis-related impairments support each other [18–20]. Self-care groups however, often have limited long-term sustainability and members may have problems in accessing the groups, for example because of financial or geographical barriers [19]. Family-based support may be a more sustainable and feasible strategy to practice self-management routines.

Several studies have indicated that family support is a highly significant factor in adherence to self-care [21,22]. When family functioning is not optimal, it is difficult to manage self-care and self-care is not as effective as it could be [21,23]. Strengthening social support and quality relationships, as is done through a family-based intervention, may also improve mental wellbeing of persons affected and their family members [24,25]. People with adequate social support seem to cope and adjust better with stressful events [26]. Since family-based support is practiced at home, no travel is required, and practicing self-care can be done at more flexible hours – a group facilitators is not required. Even though family-based support seems a sustainable and feasible strategy to practice self-management routines, especially in areas with limited health resources, it has received little attention to date. To our knowledge, no family-based intervention for leprosy, LF and podoconiosis-related disability management exists to date.

This study aimed to develop and pilot a family-based approach to support prevention and self-management of leprosy, lymphatic filariasis and podoconiosis-related disabilities in the Ethiopian context. We hypothesized that the family-based intervention would also impact psychosocial outcomes such as (family) quality of life, stigma and activity levels. The ultimate aim of this study is to improve the lives of the families involved in the study. This study builds on results of a recently published study that explored the quality of life of families with a family member affected by leprosy, LF and podoconiosis [13].

Methods

Ethics statement

Ethical approval was obtained from the Debre Markos University Health Science College Research Review Committee. In addition, the Awi zone (district) Health Desk office granted permission to conduct the study in the woredas. Since the literacy rate was low in our study area, all participants were verbally informed about the nature and objective of the study, of confidentiality of the data and the voluntary nature of the study prior to data collection. Verbal consent from each participant was obtained prior to data collection.

Intervention development

The family-based intervention was developed by the research team over the course of a year, based on an exploratory study conducted in 2017 [13]. The exploratory study consisted of in-depth interviews and focus group discussions (FGDs) and included a total of 86 participants, persons affected and their family members. Participants were asked about their self-care practices, family quality of life, and about ideas for family-based interventions to support prevention and self-management of disabilities. We found that many of the problems reported in the exploratory study were not only related to physical impairments, but also caused by stigma and poverty [13]. It was therefore decided to include the following two components in the family-based intervention, besides self-management of disabilities: (1) awareness raising, and (2) socio-economic empowerment. The family-based intervention was delivered over several monthly group meetings over the course of several months.

The main component of the family-based intervention, self-management of disabilities, used approaches that are appropriate for all three conditions as much as possible. These include inspection, foot hygiene using soap and water, skin care with removal of callous, application of ointment, elevation, exercises, bandaging and advice on appropriate footwear. The project made use of existing initiatives as much as possible, such as the WHO's Integrated morbidity management for LF and podoconiosis [27], the Ethiopian Ministry of Health's LF and podoconiosis morbidity management and disability prevention guidelines and the International Federation of Anti-Leprosy Association's guideline for prevention of disabilities in leprosy [28]. All participants received basic tools to practice self-care (Vaseline, a bucket, soap, and bandages if necessary). A detailed description of the family-based intervention can be found as supplementary information file (S1 Text).

Study design and study site

We used a quasi-experimental pre/post intervention study design with a mixed methods approach. The study was conducted in the Awi zone, located approximately 470 kilometres Northwest of the capital of Ethiopia, Addis Ababa. The Awi zone is one of the eleven zones in the Amhara region. The Awi zone was selected because the area is endemic for leprosy, LF and podoconiosis [29–31]. The study was conducted in Zigem, Guagusa Shikudad (Injibara town) and Fagita lekoma (Addis Kidam town) woreda (district).

Study population and sample

The study population included persons affected by leprosy, persons affected by LF and persons affected by podoconiosis ("persons affected") and their family members ("family members") living in the area where the intervention was offered. Since self-care practices for LF and podoconiosis are essentially the same, we did not make a distinction in our data between persons affected by LF and persons affected by podoconiosis.

This was a proof of concept study to see whether the family-based intervention had a positive impact on self-management and prevention of disabilities and to explore whether the intervention was feasible and acceptable. A sample size of 20-25 is adequate for studies that aim to demonstrate intervention efficacy and a sample size of 10-20 participants per group is adequate when trying to determine group differences [32]. To account for loss to follow up, we initially aimed to include at least 60 families in the intervention: 30 families of persons affected by leprosy and 30 families of persons affected by LF or podoconiosis. However, because we anticipated that participants would benefit from the intervention, we decided to include as many participants as possible, so more families would benefit.

We also administered questionnaires and conducted interviews and focus group discussions pre- and post-intervention. Results of the baseline interviews have been published previously and are therefore not reported in this paper [13]. We aimed to collect data until data saturation was reached. The participants in the qualitative sample will be a subset of those in the quantitative sample.

Eligibility criteria

Because our intervention aimed to improve prevention and self-management of disabilities, all persons affected had to have visible impairments due to their condition. In addition, all participants had to live in one of the three districts in which the intervention was offered. Family members had to live in the same household as their affected family member and had to know about the condition of their family member. Persons unable to give informed consent and persons younger than 15 years of age were excluded.

Sampling methods

Participants were selected using convenience sampling. For persons affected by podoconiosis and LF, local health posts were visited and a list with eligible persons in the study area was prepared. Persons on the list were visited in their home and asked to participate in the family-based intervention. Persons affected by leprosy were contacted through organisations of persons affected by leprosy in the study areas. This strategy was chosen because of the close connection of the Ethiopian National Association of Person Affected by Leprosy (ENAPAL), one of the organisations involved in the implementation of the intervention, with organisations of persons affected by leprosy in the study areas. Persons affected selected one family member from among those living in the same household, to participate in the intervention. This was done based on their availability.

Data collection

The baseline study was conducted in 2017 and 2018, the intervention ran from February to October 2019 and the follow-up study was conducted in October and November 2019. Mixed methods were used to get an understanding of the impact of the family-based intervention. Physical impairment outcomes were collected, three questionnaires were used, and in-depth interviews and focus group discussions were conducted. All data methods were administered pre- and post-intervention.

Physical impairment outcomes were assessed by noting down if impairments were present for eyes, hands and feet for leprosy (we scored impairments as either not present/0 or present/1), and frequency of acute attacks, shoe wearing behaviour and foot and leg circumference for LF and podoconiosis. Eye problems included difficulty seeing at six metres distance and lagophthalmos. Leg and foot problems included swelling, foot drop and shortening or loss of toes. Hand problems included claw hand and shortening or loss of fingers. These data were routinely collected from all persons affected during each family-based intervention group meeting.

The 25-item Beach Centre Family Quality of Life (FQoL) scale (range 0-125, with higher scores indicating higher family quality of life) was used to assess family quality of life of persons affected and their family members. The scale contains five subscales: family interaction, parenting, emotional well-being, physical/ material well-being, and disability-related support [33]. The FQoL scale has not been validated in Amharic. The FQoL scale was translated from English to Amharic, the translation was checked by translating the instrument back into English again using different interpreters. The FQoL scale was piloted tested among 20 participants before use.

The 20-item Screening of Activity Limitation and Safety Awareness (SALSA) scale was used to assess activity limitations of persons affected (range 0-80, with higher scores indicating more activity limitations). The questionnaire was developed in five countries in four continents among people affected by leprosy and diabetes [34]. The SALSA scale has been found to be a valid instrument to measure activity limitations in persons with a locomotor disability also [35]. In order to be able to compare results between conditions, we decided to use the scale for all three conditions. The SALSA scale has been validated in Amharic [36].

The 21-item SARI Stigma Scale (SSS) was used to assess stigma (range 0-63, with higher scores indicating more stigma experience). The SSS has been developed to assess leprosy-related stigma and assesses four aspects of stigma: personalised (experienced) stigma, disclosure concerns, internalised stigma and anticipated or perceived stigma. We believe the SSS can be used to assess stigma experience in other NTDs also, given that the areas of life affected by stigma are similar for people with (stigmatized) health conditions [37]. The SSS has not been validated in Amharic. The SSS was translated from English to Amharic, translated back into English again using different interpreters, and pilot tested before use. The SSS was pilot tested among 15 participants (who did not participate in the pilot test of the FQoL) before use.

Qualitative interviews, both in-depth interviews and focus group discussions, were conducted pre-intervention to develop the family-based intervention and post-intervention to evaluate the impact of the intervention.

Four facilitators and seven local area health workers received a four-day training on how to implement the intervention. Pre- and post-intervention date were collected by the four facilitators, all facilitators were local health extension workers who spoke both Amharic and Agew (local) language. The interviewers were trained in the three conditions (both clinical and psychosocial aspects) and interviewing techniques prior to data collection. The questionnaires interview guides were pilot tested before data collection commenced, these participants were not included in the final sample. Minor revisions were made based on the pilot interviews. The interviews were conducted either in participants' homes, in a private space near the patient organisation or at the location of the family-based intervention. The in-depth interviews and focus group discussions were audio recorded. A coordinator monitored the entire process.

Data analysis

The quantitative data were analysed using SPSS version 24. To correct for the large differences in sample size between baseline and the final sessions, we calculated the differences in physical impairment outcomes by comparing the baseline scores of participants with their last measurement. For some participants, their last measurement was the second or third session. For eye, hand, foot and leg impairments, acute attacks and shoe wearing, paired t-tests were performed to check whether the scores were significantly different between the baseline and the (participants') last measurement. Binary logistic regression was performed to see if there was a relationship between gender and age and physical impairment outcomes at final measurement (number of acute attacks, shoe wearing behaviour and hand, eye and foot impairments). Simple descriptive methods were used to generate a demographic profile of the study sample. Welch's unequal variances t-tests were performed to check whether the scores on the three questionnaires were significantly different pre- and post-intervention ($p\text{-value} < 0.05$). Mean overall scores, scores per participant group and where relevant scores per domain were calculated. Qualitative data were analysed using Open Code 4.03 software. The qualitative data were transcribed and coded using open, inductive coding and content analysis. All data were anonymised before data analysis.

Results

Demographic information

A total of 312 persons affected were identified to be included in the family-based intervention. Of the 312 persons listed, 275 (88%) persons affected (115 affected by leprosy and 160 affected by LF or podoconiosis) could be located and were invited to participate. All 275 persons affected who were approached were enrolled and attended the first group meeting with a family member.

A little over half (n=151, 55%) of the persons affected who participated in the intervention were female. The mean age of the participants was 51 (± 15 SD). An overview of all demographic information can be found in Table 1. No record was kept of the relationship of family members with the persons affected, however, all family members were household members (parent, child, sibling, or partner).

Table 1. Demographic information of the persons affected (n=275) who attended the first family-based intervention session.

	Persons affected by leprosy (n=115)	Persons affected by lymphatic filariasis or podoconiosis (n=160)	Total (n=275)
Average age, mean ± SD	59 ± 12.7	45 ± 14.1	51 ± 15.1
Gender, *n* (%)			
Female	35 (30.4)	116 (72.5)	151 (54.9)
Male	80 (69.6)	44 (27.5)	124 (45.1)
Living area, *n* (%)			
Zigem	5 (4.3)	160 (100.0)	165 (60.0)
Addis Kidam	86 (74.8)	0 (0.0)	86 (31.3)
Injibara	24 (20.9)	0 (0.0)	24 (8.7)
Impairments at enrolment*, *n* (%)			
Eyes	41 (35.7)	n/a	41 (14.9)
Hands	42 (36.5)	n/a	42 (15.3)
Feet	46 (40.0)	19 (11.9)	65 (23.6)
Legs	n/a	67 (41.9)	67 (24.4)
Acute attacks	n/a	137 (85.6)	137 (49.8)
Number of sessions attended, *n* (%)			
At least one	115 (100.0)	160 (100.0)	275 (100.0)
At least two	91 (79.1)	157 (98.1)	248 (90.2)
At least three	73 (63.5)	142 (88.8)	215 (78.2)
At least four	63 (54.8)	102 (63.8)	165 (60.0)
At least five	51 (44.3)	23 (14.4)	74 (26.9)
At least six	37 (32.3)	-	37 (13.5)
At least seven	18 (15.7)	-	18 (6.5)
At least eight	8 (7.0)	-	8 (2.9)

* Impairments either on the left side, right side or both sides (for example impairments in the left eye, right eye or both eyes). Participants who were lost to follow-up are also included.

Table 2 provides an overview of the number of participants out of the total of 275 participants who were invited to participate in the study, who were administered the questionnaires and who were interviewed pre- and post-intervention. Out of the 275 participants who were invited to participate in the intervention, a total of 212 participants (95 persons affected and 117 family members) were administered the FQoL, 94 persons affected the SSS and 71 persons affected the SALSA scale at baseline. Follow-up data was collected of 219 participants for the FQoL (141 persons affected and 78 family members), 149 persons affected the SSS and 126 persons affected the SALSA. We did not keep a record of if participants were interviewed both at baseline and follow-up. In addition, in-depth follow-up interviews were conducted with 25 participants (18 persons affected and 7 family members) and 58 participants were included in a total of nine focus group discussions (40 persons affected and 18 family members).

Table 2. The total number of participants included per participant group, pre- and post-intervention.

	Persons affected by leprosy		Persons affected by lymphatic filariasis or podoconiosis		Family members	
	Baseline	**Follow-up**	**Baseline**	**Follow-up**	**Baseline**	**Follow-up**
FQoL scale	48	73	47	68	117	78
SALSA scale	43	75	28	51	-	-
SARI scale (SSS)	62	78	32	71	-	-
Interviews	-	11	-	7	-	7
Focus group discussions	-	26	-	14	-	18

Group meeting attendance

There were monthly follow-up visits (group meetings) of the family-based intervention. A total of 74 different group meetings were organised, spread over eight 'sessions' (each session had on average 9 different group meetings). These groups met on different days. On average 12 persons affected with one family member each participated in each meeting.

Sometimes participants were for example present at the first, third and fourth session, but not at the second. A total of 275 (100%) persons affected attended at least one session, 248 (90%) attended at least two sessions and 215 (78%) attended at least three sessions (Table 1). In most cases, different sessions were held for persons affected by leprosy and their family members, and for persons affected by podoconiosis or LF and their family members, because of the distribution of diseases per district (Table 1).

Physical outcomes

Table 3 shows the number of persons affected by leprosy who had eye, hand or foot impairments at baseline and at their last follow-up. For a quarter of the participants, their last follow-up was the second or third session (n=25, 27.5%). There was no change in the number of participants with eye impairments. In addition, logistic regression showed that as age increased, the odds of having eye impairments also increased ($p<0.05$, odds ratio 1.047, with 95%CI 1.006-1.091). The number of participants with hand impairments decreased at follow-up, but this was not significant. The number of persons affected by leprosy with foot impairments significantly decreased (Table 3). There was no relationship between age and gender and number of acute attacks, shoe wearing behaviour, and hand and foot impairments and between gender and eye impairments at final assessment ($p>0.05$, logistic regression).

Table 3. The number of persons affected by leprosy with eye, hand and/or foot impairments and the number of participants affected by lymphatic filariasis or podoconiosis with leg impairments, at least one acute attack per month and who wears shoes and at baseline and at participants' last family-based intervention session.

	Total number of participants baseline and follow-up	**Baseline, *n* (%)**	**Final/last assessment[a], *n* (%)**	**Difference (%)**	**p-value[b]**
Has eye impairments	91	31 (34.1)	31 (34.1)	0 (0)	NS
Has hand impairments	91	36 (39.6)	33 (36.3)	3 (8.3)	NS
Has foot impairments	91	44 (48.4)	36 (39.6)	8 (18.2)	0.011
Has leg impairments	145	62 (42.8)	30 (20.7)	32 (51.6)	0.000
Has at least one acute attack per month	146	126 (85.7)	22 (15.2)	104 (82.5)	0.000
Wears shoes	146	136 (93.2)	142 (97.3)	6 (4.4)	0.014

[a] The final assessment is the last session the participant attended. For persons affected by leprosy this was the second (13% of the participants), third (14%), fourth (9%), fifth (8%), sixth (18%), seventh (13%) or eighth (25%) session, not including the 24 participants that only attended the baseline session. For persons affected by lymphatic filariasis or podoconiosis this was at the second (9%), third (35%), fourth (41%) or fifth (15%) session, the 13 participants that only attended the baseline session not included.

[b] Test used is paired t-test with a significance level of 0.05. NS = not significant ($p>0.05$).

The number of persons affected by LF and podoconiosis who had leg impairments and who had at least one acute attack per month significantly decreased and who wore shoes significantly increased between baseline and participants' last follow-up (Table 3). In addition, the mean values of the measurements of the right and left leg (leg lymphedema) and right and left foot all significantly decreased between baseline and the final assessment (Table 4).

Table 4. Leg circumference for persons affected by lymphatic filariasis and podoconiosis at each family-based intervention session.

	Baseline		Final/last assessment[a]		Difference (%)	95% CI[b]	p-value
	n	Mean (SE)	n	Mean (SE)			
Circumference right leg	139	26.5 (0.35)	139	24.7 (0.32)	1.8 (6.8)	1.4-2.2	0.000
Circumference left leg	132	26.8 (0.32)	132	25.1 (0.32)	1.7 (6.3)	1.4-2.0	0.000
Circumference right foot	143	25.2 (0.22)	143	23.4 (0.21)	1.8 (7.1)	1.6-2.2	0.000
Circumference left foot	136	25.6 (0.26)	136	23.7 (0.21)	1.9 (7.4)	1.6-2.3	0.000

[a] The final assessment is the last session the participant attended. This was either at the second (for 9-10% of participants), third (36-37% of participants), fourth (40-41% of participants) or fifth (14% of participants) session that was held. The 13 participants that only attended the baseline session are not included.

[b] 95% Confidence interval for mean reduction. Test used is a paired t-test with a significance level of 0.05.

Awareness raising

Participants were asked about the awareness raising component of the intervention during the in-depth interviews and focus group discussions. All participants said they were positive about (participating in) the family-based intervention and explained that the intervention had improved their knowledge about the three conditions and of self-management. One participant explained:

> *"…Previously I did not think this system [self-care] gives me relief from my illness but after starting implementation of self-care practices, I saw change within two weeks. During the monthly follow-up time research assistants measured my leg circumference and they told me my swelling shows decrement, during this time I felt empowered…"*
>
> (65-year old man affected by podoconiosis, focus group discussion).

One family member said:

> *"…I understood that these diseases are non-communicable after treatment, this gave me high motivation to support my [affected] father…"*
>
> (20-year old female family member of person affected by leprosy, in-depth interview).

Flyers with disease specific information were also prepared, but only disseminated to the participants after the intervention was completed.

Socio-economic empowerment

Socio-economic empowerment consisted the formation of establishing Disabled People's Organisations (DPOs). Two DPOs existed already (initiated by the Ethiopian National Association of Persons Affected by Leprosy/ENAPAL, one of the partners in the project): the leprosy specific group in Addis Kidam and in Injibara. A DPO for all three conditions was established in Zigem. Each DPO collected a small contribution fee from its participants, 5 to 20 birr each month (less than one dollar or euro). These fees were used to provide loans for the participants (micro-finance). DPOs also lobbied for 'benefits', e.g. the use of land, from the government. These groups met monthly. While the facilitators of the project helped to establish these groups and were present during the meeting, they did not give any guidance on the management of the groups. This was done by persons affected themselves.

Both persons affected and their family members said the establishment of the associations was important to them. Participants explained that the association addressed their economic difficulties by providing (self-saved) money:

> *"...[The association] encourages saving. We save and can take a loan and use it when there is a challenge..."*
>
> (65-year old man affected by leprosy, FGD).

Some participants were critical towards the leadership of the (already existing) leprosy association, who they felt not always provided loans to everyone in the organisation.

Family quality of life, stigma and activity limitations

Family quality of life

Table 5 shows the differences in mean family quality of life scores of persons affected and their family members at baseline and follow-up. Higher values indicate better family quality of life. The increase in family quality of life scores for persons affected and for family members are significant (Table 5), indicating that family quality of life has significantly improved after the intervention. Mean family quality of life scores of subgroups improved also, but this difference was not significant for the subgroup of persons affected by leprosy.

Table 5. Mean differences between baseline and follow-up in family quality of life (Beach Centre FQoL), stigma (SARI stigma scale) and activity limitations (SALSA scale) per subgroup.

		Baseline total score, mean (95%CI)	Follow up total score, mean (95%CI)	Difference (%)	p-value[a]
Family quality of life	All persons affected (n=95 ~ n=141)	76.9 (74.2-79.6)	84.1 (81.2-87.0)	7.2 (9.4)	0.000
	Persons affected leprosy[b] (n=48 ~ n=73)	79.1 (75.2-83.0)	79.9 (74.4-85.4)	0.8 (1.0)	NS
	People affected podoconiosis or lymphatic filariasis[b] (n=47 ~ n=68)	74.6 (70.9-78.4)	88.6 (87.8-89.5)	14 (18.8)	0.000
	All family members (n=117 ~ 78)	67.4 (65.1-69.7)	89.9 (87.2-92.6)	22.5 (33.4)	0.000
Stigma	People affected (total) (n=94 ~ n=149)	24.0 (21.6-26.5)	16.7 (14.6-18.9)	7.3 (30.4)	0.000
	People affected leprosy[b] (n=62 ~ n=78)	22.7 (19.5-26.0)	16.5 (13.0-20.0)	6.2 (27.3)	0.011
	People affected podoconiosis or lymphatic filariasis[b] (n=32 ~ n=71)	26.6 (23.2-30.0)	17.0 (14.3-19.6)	9.6 (36.1)	0.000
Activity limitations	People affected (total) (n=71 ~ n=126)	38.5 (35.1-41.9)	36.0 (33.9-38.2)	2.5 (6.5)	NS
	People affected leprosy[b] (n=43 ~ n=75)	44.2 (39.8-48.6)	39.9 (36.7-43.0)	4.3 (9.7)	NS
	People affected podoconiosis or lymphatic filariasis[b] (n=28 ~ n=51)	29.8 (26.2-33.4)	30.3 (28.6-32.0)	0.5 (1.7)	NS

[a] Difference between baseline and follow-up scores, calculated using Welch's unequal variances t-test. NS = not significant (p>0.05).

[b] These are subgroups of the ‘persons affected (total)’ group.

When looking at the five domains of the Beach Centre FQoL scale (family interaction, parenting, emotional wellbeing, physical wellbeing and disability-related support), there was a significant improvement in the domains emotional, physical and disability-related support for persons affected at baseline compared to follow-up (p<0.001, unequal variances t-test). The mean scores on these domains have improved over 17%. All five domains have significantly improved for family members at follow-up (p<0.001, unequal variances t-test). The domains with the biggest mean improvement (>40%) for family members are emotional, physical and disability-related support. An overview can be found in supporting information file S2 Text.

Stigma

Mean (SARI) stigma scores for persons affected significantly decreased from 24.0 at baseline to 16.7 at follow-up. This indicates that stigma for persons affected significantly decreased after the intervention (lower scores on the SARI stigma scale indicate less stigma). A significant decrease was also found for both subgroups (Table 4). Mean scores on three out of the four domains of the SARI stigma scale significantly decreased after the intervention (please see S2 Text), these include experienced, internalised and anticipated stigma ($p<0.05$, unequal variances t-test). The mean difference on the domain disclosure decreased, but this difference was not significant ($p<0.05$, unequal variances t-test).

Some participants in the in-depth interviews emphasized that they felt more confident after the intervention. In addition, some participants said their family dynamics and social participation had improved. One participant explained:

> *"…I saw many changes, my wound healed, I had a bad smell before but now after you [project staff] came here my wound is healed. Now I can go to church, I bake injera [Ethiopian flatbread] and prepare a soup like I used to. There is a big change [after the intervention]…"*
>
> (66-year old woman affected by leprosy, FGD).

Another participant said:

> *"…Previously people discriminated us [the family], we were not allowed to drink coffee with them, but now there is no stigma and discrimination…"*
>
> (29-year old woman affected by podoconiosis, in-depth interviews).

Activity limitations

With a mean age of 53 (SD 14.4), participants who were administered the SALSA at follow-up were on average a few years older than participants included at baseline (mean 48, SD 16.5). Table 5 shows differences in mean activity limitation scores of persons affected at baseline and follow-up. Lower activity limitations (SALSA) scores indicate less activity limitations. The mean activity limitations decreased at follow-up for persons affected in general and for the persons affected by leprosy subgroup, but increased for the persons affected by podoconiosis and LF subgroup. These differences were not significant ($p>0.05$, Table 5).

An overview of the different categories of the SALSA (no, mild, moderate, severe or extreme activity limitations) and the number of participants in the in each category at baseline and follow-up can be found as supporting information file (S2 Text).

The change in the severe limitations group was the only significant difference between baseline and follow-up. The percentage of persons affected with severe limitations significantly decreased at follow-up ($p<0.05$, unequal variances t-test, pooled data of persons affected by leprosy, podoconiosis and LF).

Discussion

Findings from the present study show that the family-based intervention had a positive impact on impairments and self-management of disabilities. Persons affected and family members were enthusiastic and had a positive attitude towards participating in the intervention. In addition, family quality of life improved and stigma decreased at follow-up. Several studies have found teaching persons affected basic self-care techniques to be a successful approach for morbidity management [15,16,18–20,22,38–41,42], this finding is supported by the present study.

Family-based format

A new aspects of our intervention is its family-based format. Group-based disability care, often in the form of self-care groups, are common, especially for persons affected by leprosy and LF [15,16,18–20,22,38–41]. To our knowledge, the current intervention is the first family-based intervention for leprosy and podoconiosis-related disability management and the first family-based self-care intervention for leprosy, LF and podoconiosis. Only a few home-based self-care interventions have been conducted for LF [43–45]. In line with our findings, these studies reported good physical impairment outcomes [43–45]. This suggests that family-based self-care is a feasible alternative for self-care groups. Challenges often reported for self-care groups include long distances that need to be travelled to attend group meetings [19], lack of time to attend meetings [46] and sustainability of the groups [18,47]. These challenges do not apply to family-based interventions, where participants do not need to travel to attend and no facilitator is needed. In addition, a family-based intervention is relatively inexpensive since this can be practiced at home and no travel is required. Furthermore, encouragement by significant others can increase motivation, which is essential for self-care behaviour [23,47]. Family-based support therefore seems like a sustainable option, especially in contexts where resources are scarce, which is often the case in areas where leprosy, LF and podoconiosis are endemic [17,19,48–50].

Components: self-management of disabilities, awareness, socio-economic empowerment

The intervention in the present study consisted of three components: self-management of disabilities awareness raising, and socio-economic empowerment, with self-management as main component. These three components were developed based on findings from an exploratory study, where we found that many problems faced by participants were not only related to physical impairments, but also caused by stigma and poverty [13]. These findings are supported by a study among over 1,000 persons affected by leprosy in Indonesia, in which the authors stress that “stigma reduction activities and socio-economic rehabilitation are urgently needed in addition to strategies to reduce the development of further physical impairment after release from treatment” [7].

While we have identified ‘awareness raising’ as a separate component, health education and self-care training are often integrated in self-care and morbidity management training in other studies [16,40,43,44,51]. In addition to providing education about self-care and clinical manifestations during the group sessions, in the present study it was initially also planned to distribute printed material in the communities, to raise awareness and reduce stigma. Due to time constraints, the printed materials were only distributed after the follow-up assessments had been conducted. In leprosy, stigma and a lack of knowledge have been identified as obstacles to case finding and adherence to treatment [52,53]. In addition, social stigma has been associated with poor psychosocial health outcomes [54,55]. Stigma reduction is therefore a crucial component of morbidity management interventions [7]. We recommend including pre- and post-intervention assessment of knowledge and community stigma in future studies, to get a better understanding of the changes in knowledge and community stigma.

Socio-economic empowerment was one of the three components of the intervention in the present study and consisted of the exploration and formation of (disease-specific) Disabled People’s Organisations. These organisations/groups collected monthly fees from participants, that were used to provide loans to group members. Unfortunately, we did not collect any information on how much money was collected by the DPOs as monthly fees, and if any loans were provided. Several studies have emphasized the marginalized position of persons affected in their communities, poverty and a lack of resources for income generation are challenges often reported for persons affected by leprosy, LF and podoconiosis [5,17,19,48–50]. Sometimes, income is too low to acquire basic materials for self-care [46,48]. Costs for treatment, associated non-medical costs and reduced ability to work may cause a financial burden on the entire household of persons affected [5,17,49,50]. We therefore consider socio-economic empowerment an essential component of interventions for self-management and prevention of disabilities. Without income, self-care items such as Vaseline and shoes cannot be bought. Previous studies have also reported positive results in terms of reducing community stigma among persons affected by leprosy, using micro-credit loans and vocational training [56,57]. In addition, if material wellbeing (family income) is positively influenced, family quality of life is higher and all family members, including the affected person, benefit [58].

Primary outcomes: physical impairments

In the present study, foot and leg impairments, number of acute attacks, lymphedema and shoe wearing all significantly improved at follow-up. Eye and hand impairments did not improve after the intervention. We believe the lack of improvement in eye impairments relates to their more chronic, permanent nature. Vision loss is not (naturally) reversible. Unfortunately, we did not record severity of impairments. We believe some of the hand impairments reported were either permanent (e.g. loss of digits), may only have been reversed after a longer period of time, or with reconstructive surgery. This is supported by findings from a study in China, that found that regularity in self-care of persons affected by leprosy was only established after three monthly reinforcements [59]. Findings from the present study also indicate that more attention should be paid to self-care practices of hand impairments, something that should be taken into account in future studies.

Secondary outcomes: family quality of life, stigma, activities

The intervention in the present study improved family quality of life for persons affected and their family members and decreased stigma. This is an important finding, since leprosy, LF and podoconiosis can negatively impact individual [60–66] and family quality of life [13]. Stigma and visible impairments can also deteriorate quality of life [64]. Quality of life is crucial in the evaluation of health care interventions [67]. Leprosy, LF and podoconiosis-related disabilities require life-long care. Effective morbidity management can lessen impairments and disabilities and is therefore imperative to improve individual quality of life of persons affected [17,67]. It is therefore promising that the intervention in the present study improved family quality of life. We believe the improvement in physical impairments of persons affected in the present study contributed to the improvement in quality of life and reduction in stigma we found, a finding that is supported by a study in Bangladesh [64]. Visible impairments are major determinants of stigma and participation restrictions [7–9].

Even though most physical impairment outcomes improved at the final assessment in the present study, activity levels (assessed by the SALSA scale) did not significantly improve. This is likely related to the lack of improvement found in hand impairments and in part related to the slightly older age (51 years) of the participants in the present study. In addition, during the study we discovered that the SALSA scale is not the most suitable tool for persons affected by LF and podoconiosis. Fifteen out of the total of 20 questions on the SALSA scale relate to hand movements and strength. It is therefore not surprising that persons affected by podoconiosis and LF had less activity limitations on the SALSA scale, given that these conditions mostly cause lymphoedema of the leg(s) or swelling of other organs like the scrotum [6]. Since the SALSA scale is more sensitive to limitations caused by leprosy, future studies that aim to assess activity limitations among persons affected by LF and podoconiosis would benefit from using a different tool than the SALSA scale.

Strengths, limitations and recommendations

The present study assessed the short-term outputs and effect of a family-based self-care intervention among a relatively small sample of persons affected by leprosy, LF and podoconiosis. Because of the non-random sample and relatively short follow-up time, effectiveness of the intervention could not be assessed. In addition, the present study did not use a control group of persons not participating in the family-based intervention. A comparison with a control group would have provided additional evidence to the study. However, since this was a proof of concept study, we believe we have demonstrated the feasibility and potential of the family-based intervention. A strength of this study is the mixed-method approach that allowed for triangulation of the data.

To date most prevention of disability effectiveness studies have been flawed by failing to use a randomised controlled design. This has resulted in a lack of evidence about the effectiveness of these interventions. Further research using a randomised controlled design with a larger sample is needed. As a result of this proof of concept study, a randomised controlled trial study using validated measurement tools and more physical impairment outcomes is in development.

Because family structure, roles and functioning are influenced by culture, which in turn influences self-care behaviours, it would be interesting for future studies to explore the self-care behaviours of families in different cultures.

Conclusions

This proof of concept study showed that the family-based intervention had a positive effect on impairments and self-management of disabilities, family quality of life and stigma. We recommend a large-scale efficacy trial, using a randomised controlled trial and validated measurement tools, to determine its effectiveness and long-term sustainability. Future studies who aim to assess activity limitations among persons affected by lymphatic filariasis and podoconiosis are recommended not to use the SALSA scale, as this scale is more sensitive to limitations caused by leprosy.

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References

1. Mitjà O, Marks M, Bertran L, Kollie K, Argaw D, Fahal AH. Integrated Control and Management of Neglected Tropical Skin Diseases. PLoS Negl Trop Dis 2017;11:e0005136.
2. Suzuki K, Akama T, Kawashima A, Yoshihara A, Yotsu RR, Ishii N. Current status of leprosy: epidemiology, basic science and clinical perspectives. J Dermatol 2012;39:121–9.
3. Davey G. Podoconiosis, non-filarial elephantiasis, and lymphology. Lymphology 2010;43:168.
4. Babu S, Nutman TB. Immunology of lymphatic filariasis. Parasite Immunol 2014;36:338–46.
5. Chandler DJ, Grijsen ML, Fuller LC. With Bare Feet in the Soil: Podoconiosis, a Neglected Cause of Tropical Lymphoedema. Dermatology 2020:1–12.
6. Lyons OTA, Modarai B. Lymphoedema. Surg 2013;31:218–23.
7. Van Brakel WH, Sihombing B, Djarir H, Beise K, Kusumawardhani L, Yulihane R, et al. Disability in people affected by leprosy: the role of impairment, activity, social participation, stigma and discrimination. Glob Health Action 2012;5:18394.
8. Tora A, Franklin H, Deribe K, Reda AA, Davey G. Extent of podoconiosis-related stigma in Wolaita Zone, Southern Ethiopia: a cross-sectional study. Springerplus 2014;3:647.
9. Zeldenryk LM, Gray M, Speare R, Gordon S, Melrose W. The Emerging Story of Disability Associated with Lymphatic Filariasis: A Critical Review 2011.
10. Awofeso N. Stigma and socio-economic reintegration of leprosy sufferers in Nigeria. Acta Leprol 1996;10:89–91.
11. Abdulmalik J, Nwefoh E, Obindo J, Dakwak S, Ayobola M, Umaru J, et al. Emotional difficulties and experiences of stigma among persons with lymphatic filariasis in Plateau State, Nigeria. Health Hum Rights 2018;20:27.
12. Person B, Bartholomew LK, Gyapong M, Addiss DG, van den Borne B. Health-related stigma among women with lymphatic filariasis from the Dominican Republic and Ghana. Soc Sci Med 2009;68:30–8.
13. van't Noordende AT, Aycheh MW, Schippers A. The impact of leprosy, podoconiosis and lymphatic filariasis on family quality of life: A qualitative study in Northwest Ethiopia. PLoS Negl Trop Dis 2020;14:e0008173.
14. Jansen B. Systematic review: skin and wound management interventions related to skin-neglected tropical diseases. VU University Amsterdam, 2017.
15. Pryce J, Mableson HE, Choudhary R, Pandey BD, Aley D, Betts H, et al. Assessing the feasibility of integration of self-care for filarial lymphoedema into existing community leprosy self-help groups in Nepal. BMC Public Health 2018;18:201.
16. Narahari SR, Bose KS, Aggithaya MG, Swamy GK, Ryan TJ, Unnikrishnan B, et al. Community level morbidity control of lymphoedema using self care and integrative treatment in two lymphatic filariasis endemic districts of South India: A non randomized interventional study. Trans R Soc Trop Med Hyg 2013;107:566–77.
17. Douglass J, Graves P, Gordon S. Self-care for management of secondary lymphedema: a systematic review. PLoS Negl Trop Dis 2016;10.
18. Benbow C, Tamiru T. The experience of self-care groups with people affected by leprosy: ALERT, Ethiopia. Lepr Rev 2001;72:311–21.

19. Deepak S, Hansine PE, Braccini C. Self-care groups of leprosy-affected people in Mozambique. Lepr Rev 2013;84:283.
20. Ebenso J, Muyiwa LT, Ebenso BE. Self care groups and Ulcer prevention in Okegbala Nigeria. Lepr Rev 2009;80:187.
21. Grey M, Knafl K, McCorkle R. A framework for the study of self-and family management of chronic conditions. Nurs Outlook 2006;54:278–86.
22. Li J, Mu H, Ke W, Bao X, Wang Y, Wang Z, et al. The sustainability of self-care in two counties of Guizhou Province, Peoples' Republic of China. Lepr Rev 2008;79:110–7.
23. Dunbar SB, Clark PC, Quinn C, Gary RA, Kaslow NJ. Family influences on heart failure self-care and outcomes. J Cardiovasc Nurs 2008;23:258.
24. Connell J, Brazier J, O'Cathain A, Lloyd-Jones M, Paisley S. Quality of life of people with mental health problems: a synthesis of qualitative research. Health Qual Life Outcomes 2012;10:138.
25. Baumeister RF, Leary MR. The need to belong: desire for interpersonal attachments as a fundamental human motivation. Psychol Bull 1995;117:497.
26. Gurung R, Sarason B, Sarason I. Close personal relationships and health outcomes: A key to the role of social support. Handb Pers Relationships Theory, Res Interv (2nd Ed) Chichester, UK Wiley 1997:547–73.
27. Deribe K, Kebede B, Tamiru M, Mengistu B, Kebede F, Martindale S, et al. Integrated morbidity management for lymphatic filariasis and podoconiosis, Ethiopia. Bull World Health Organ 2017;95:652.
28. ILEP. ILEP Learning Guide four: How to prevent disability in leprosy. 2006.
29. World Health Organization. Weekly Epidemiological Record, 31 August 2018. Global leprosy update, 2017: reducing the disease burden due to leprosy. vol. 93. 2018.
30. World Health Organization. Weekly Epidemiological Record, 2 November 2018. Global programme to eliminate lymphatic filariasis: progress report, 2017. vol. 93. 2018.
31. Deribe K, Cano J, Trueba ML, Newport MJ, Davey G. Global epidemiology of podoconiosis: A systematic review. PLoS Negl Trop Dis 2018;12:e0006324.
32. Hertzog MA. Considerations in determining sample size for pilot studies. Res Nurs Health 2008;31:180–91.
33. Hoffman L, Marquis J, Poston D, Summers JA, Turnbull A. Assessing family outcomes: Psychometric evaluation of the beach center family quality of life scale. J Marriage Fam 2006;68:1069–83.
34. Group SCS, Group SCS. The development of a short questionnaire for screening of activity limitation and safety awareness (SALSA) in clients affected by leprosy or diabetes. Disabil Rehabil 2007;29:689–700.
35. Mol S. Examining the validity of the SALSA scale as a generic measure of activity. VU University, 2010.
36. Wijk U, Brandsma JW, Dahlström O, Björk M. The concurrent validity of the Amharic version of Screening of Activity Limitation and Safety Awareness (SALSA) in persons affected by leprosy. Lepr Rev 2013;84:13–22.
37. Van Brakel WH. Measuring health-related stigma—a literature review. Psychol Health Med 2006;11:307–34.

38. Aggithaya MG, Narahari SR, Vayalil S, Shefuvan M, Jacob NK, Sushma K V. Self care integrative treatment demonstrated in rural community setting improves health related quality of life of lymphatic filariasis patients in endemic villages. Acta Trop 2013;3:198–204.
39. Madhavan K, Vijayakumaran P, Ramachandran L, Manickam C, Rajmohan R, Mathew J, et al. Sustainable leprosy related disability care within integrated general health services: findings from Salem District, India. Lepr Rev 2007;78:353–62.
40. Chen S, Zhang L, Wang Z, Zhou J, Liu Y, Mao C. Experiences from a collaborative project on the prevention of disability in leprosy patients in Shandong Province, the People's Republic of China. Lepr Rev 2001;72:330–6.
41. Ziperstein J, Dorkenoo M, Datagni M, Drexler N, Murphy M, Sodahlon Y, et al. Final program evaluation methods and results of a National Lymphedema Management Program in Togo, West Africa. J Epidemiol Glob Health 2014;4:125–33.
42. Addiss DG, Louis-Charles J, Roberts J, LeConte F, Wendt JM, Milord MD, et al. Feasibility and effectiveness of basic lymphedema management in Leogane, Haiti, an area endemic for bancroftian filariasis. PLoS Negl Trop Dis 2010;4.
43. Yahathugoda TC, Weerasooriya M V, Samarawickrema WA, Kimura E, Itoh M. Impact of two follow-up schemes on morbidity management and disability prevention (MMDP) programme for filarial lymphedema in Matara, Sri Lanka. Parasitol Int 2018;67:176–83.
44. Jullien P, Somé J d'Arc, Brantus P, Bougma RW, Bamba I, Kyelem D. Efficacy of home-based lymphoedema management in reducing acute attacks in subjects with lymphatic filariasis in Burkina Faso. Acta Trop 2011;120:S55–61.
45. Organization WH. Lymphatic filariasis: progress of disability prevention activities. Wkly Epidemiol Rec Relev Épidémiologique Hebd 2004;79:417–24.
46. Lima MC V, Barbosa FR, Santos DCM, Nascimento RD, D'Azevedo SSP. Practices for self-care in Hansen's disease: face, hands and feet. Rev Gaúcha Enferm 2018;39:e20180045.
47. Cross H. The prevention of disability for people affected by leprosy: whose attitude needs to change? Lepr Rev 2007;78:321–9.
48. Shahvisi A, Meskele E, Davey G. A human right to shoes? Establishing rights and duties in the prevention and treatment of podoconiosis. Health Hum Rights 2018;20:53.
49. Tembei AM, Kengne-Ouaffo JA, Ngoh EA, John B, Nji TM, Deribe K, et al. A comparative analysis of economic cost of podoconiosis and leprosy on affected households in the northwest region of Cameroon. Am J Trop Med Hyg 2018;98:1075–81.
50. Caprioli T, Martindale S, Mengiste A, Assefa D, Tamiru M, Negussu N, et al. Quantifying the socio-economic impact of leg lymphoedema on patient caregivers in a lymphatic filariasis and podoconiosis co-endemic district of Ethiopia. PLoS Negl Trop Dis 2020;14:e0008058.
51. Suma TK, Shenoy RK, Kumaraswami V. Efficacy and sustainability of a footcare programme in preventing acute attacks of adenolymphangitis in Brugian filariasis. Trop Med Int Heal 2002;7:763–6.
52. Heijnders ML. Experiencing leprosy: perceiving and coping with leprosy and its treatment. A qualitative study conducted in Nepal. Lepr Rev 2004;75:327–37.
53. Rafferty J. Curing the stigma of leprosy. Lepr Rev 2005;76:119–26.
54. Litt E, Baker MC, Molyneux D. Neglected tropical diseases and mental health: a perspective on comorbidity. Trends Parasitol 2012;28:195–201.

55. Staring ABP, Van der Gaag M, Van den Berge M, Duivenvoorden HJ, Mulder CL. Stigma moderates the associations of insight with depressed mood, low self-esteem, and low quality of life in patients with schizophrenia spectrum disorders. Schizophr Res 2009;115:363–9.
56. Ebenso B, Ayuba M. “Money is the vehicle of interaction”: insight into social integration of people affected by leprosy in northern Nigeria. Lepr Rev 2010;81:99.
57. Dadun D, Van Brakel WH, Peters RMH, Lusli M, Zweekhorst M, Bunders JGF, et al. Impact of socio-economic development, contact and peer counselling on stigma against persons affected by leprosy in Cirebon, Indonesia–a randomised controlled trial. Lepr Rev 2017;88:2–22.
58. Park J, Turnbull AP, Turnbull III HR. Impacts of poverty on quality of life in families of children with disabilities. Except Child 2002;68:151–70.
59. Smith W, Cairns S, Zhang G, Zheng T, Watson JM, Lehman LF, et al. Prevention of impairment in leprosy; results from a collaborative project in China. Int J Lepr Other Mycobact Dis 1995;63:507–17.
60. Borges-de-Oliveira R, Rocha-Leite CI, Araujo-de-Freitas L, Queiroz DA, Machado PRL, Quarantini LC. Perception of social exclusion, neuropathy, and quality of life among Hansen’s disease patients. Int J Psychiatry Med 2015;49:176–86.
61. Lustosa AA, Nogueira LT, Pedrosa JI dos S, Teles JBM, Campelo V. The impact of leprosy on health-related quality of life. Rev Soc Bras Med Trop 2011;44:621–6.
62. Morgan PA, Franks PJ, Moffatt CJ. Health-related quality of life with lymphoedema: a review of the literature. Int Wound J 2005;2:47–62.
63. Mousley E, Deribe K, Tamiru A, Davey G. The impact of podoconiosis on quality of life in Northern Ethiopia. Health Qual Life Outcomes 2013;11:122.
64. Tsutsumi A, Izutsu T, Islam AM, Maksuda AN, Kato H, Wakai S. The quality of life, mental health, and perceived stigma of leprosy patients in Bangladesh. Soc Sci Med 2007;64:2443–53.
65. Santos VS, Oliveira LS, Castro FDN, Gois-Santos VT, Lemos LMD, do CO Ribeiro M, et al. Functional activity limitation and quality of life of leprosy cases in an endemic area in Northeastern Brazil. PLoS Negl Trop Dis 2015;9:e0003900.
66. Thomas C, Narahari SR, Bose KS, Vivekananda K, Nwe S, West DP, et al. Comparison of three quality of life instruments in lymphatic filariasis: DLQI, WHODAS 2.0, and LFSQQ. PLoS Negl Trop Dis 2014;8:e2716.
67. Kumari AK, Krishnamoorthy K, Harichandrakumar KT, Das LK. Health Related Quality of Life, an appropriate indicator to assess the impact of morbidity management and disability prevention activities towards elimination of lymphatic filariasis. Filaria J 2007;6:8.

CHAPTER 9

Key sources of strength and resilience for persons receiving services for Hansen's disease (leprosy) in Porto Velho, Brazil: What can we learn for service development?

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Abstract

Background

Individuals affected by Hansen's disease (leprosy) often experience stigma and discrimination. Greater psychosocial resilience may enable people to deal with such discrimination. This study aimed to explore sources of strength and resilience for individuals affected by Hansen's disease in Brazil.

Methods

We used a cross-sectional study design with a qualitative approach. Semi-structured focus groups were conducted. Analysis comprised thematic categorization of transcripts.

Results

Thirty-one participants were included: 23 individuals affected by Hansen's disease and 8 healthcare providers. We found that while a few individuals affected were provided with formal psychological support in the early phases of their treatment, many noted the importance of providing such support at this time. Most participants described relationships with and social support from family members, friends and with others affected by Hansen's disease as their primary source of resilience. A key context for building resilience was through the peer-level sharing and engagement experienced in self-care and support groups. Participants also emphasised the importance of providing appropriate information about Hansen's disease and the importance of beliefs and spirituality.

Conclusions

Hansen's disease services should seek to build resilience in early treatment through counselling, and during treatment and beyond by having people affected getting together. Across both settings supporting family and social relationships, providing accurate information, and acknowledging spiritual beliefs are important.

Introduction

Since the introduction of multidrug therapy (MDT) in 1982, the management of Hansen's disease (leprosy), a neglected tropical disease (NTD), has advanced considerably, and millions have been cured from the disease. However, the transmission of its pathogen *Mycobacterium leprae* is ongoing, so the primary focus of Hansen's disease services in many countries is on disease surveillance and medical treatment. However, many people affected emphasize that the adverse psychosocial consequences of the disease outweigh the physical and functional dimensions [1–3]. In response, therefore, meaningful Hansen's disease services should also attend to psychosocial corollaries of the disease [4].

A commonly reported psychosocial concern for many individuals affected by the disease is Hansen's disease-related stigma and discrimination. This adversely affects psychological wellbeing and is linked with depression and even suicide [3]. It also impacts social participation [5–8], with negative consequences for interpersonal relationships, social inclusion, social status, education and even employment [7–9]. It is therefore not surprising that a diagnosis of Hansen's disease can profoundly and negatively affect a person's general quality of life [5,10–12] and specifically, their psychosocial wellbeing [4,10,12,13].

Within psychologically-oriented research, efforts to address Hansen's disease-related stigma and discrimination have largely focused on the stigmatised person, the stigmatising context and broader systems [14,15]. Corresponding interventions have sought to treat the individual affected, or influence community attitudes or change existing laws and policies. Few approaches have sought to address discrimination from a resilience perspective. That is, few have sought to enhance wellbeing by building the psychosocial capacity of the stigmatised person to withstand social challenges and overcome discrimination. Resilience, the capacity to overcome and thrive in the face of adversity [16], aligns closely with some of the skills which may benefit people to overcome discrimination at an individual level.

Resilience-related skills and thinking may enable people to thrive when faced with adversity [17], and may have a positive effect on general quality of life, happiness, psychosocial wellbeing and even longevity [17,18]. Factors associated with resilience include optimism, positive thinking, self-efficacy, problem-solving skills, attachment to others and faith. Resilience also appears to be associated with enhanced social support [16,18–21], which may in itself be a protective factor in stressful situations [22]. Research suggests that while all people are resilient to some degree, there are also a number of resilience-related behaviours or qualities that can be learned [16,21,23].

This would indicate that in the Hansen's disease context, if more opportunities and resources were available to build and enhance the psychosocial resilience of people affected, then they may be better resourced to recognise, counteract and deal with discriminatory behaviour. To date, the concept of resilience has not been the focus of much research in the Hansen's disease and NTD area. Greater understanding of what factors contribute to the resilience of individuals affected and their family members could meaningfully inform Hansen's disease service development. Given the similarities in stigma types, manifestations and impact among NTDs [24], lessons learned from this research can also inform service development for other stigmatized conditions, such as other NTDs.

The current study is part of a larger project that aims to develop and pilot an intervention to build individual and family resilience against Hansen's disease-related discrimination. The project has three phases: a scoping review [15] about evidence-based resilience promoting interventions in the context of stigma and discrimination; the current study explores the sources of strength and resilience of an initial sample of individuals affected by Hansen's disease and experienced health service providers in the context of Hansen's disease treatment; and a third phase underway in India, where the principles of resilience promotion found in the scoping review plus the lessons learned from the current study inform the design of a pilot intervention to build resilience among families that are experiencing Hansen's disease.

Materials and methods

Study design

This study used a cross-sectional design with a qualitative approach. Semi-structured focus group discussions were conducted to gain insight in the sources of strength and resilience of an initial sample of individuals affected by Hansen's disease and experienced service providers.

Study site

Focus groups were conducted at a secondary level public health facility, the Osvaldo Cruz Policlinic, a State of Rondônia Reference Center for Hansen's disease located in Porto Velho, Brazil, in December 2018. Approximately 300 people affected by Hansen's disease are seen at the facility each month. The location caters to a large number of patients coming from various regions of the State of Rondônia, northern Brazil, in the Amazon region, who, despite having completed the drug regimen for active Hansen's disease, still require care. Reaction management, physical rehabilitation and prevention of disabilities self-care groups are some of the services provided in addition to regular MDT treatment. It is located in an urban center of about 539.354 inhabitants according to the Population Estimate for 2020 by the Brazilian Institute of Geography and Statistics. In 2019 the number of new cases registered in the health surveillance system was 122 [25].

Study population and sampling methods

People with understanding of Hansen's disease and Hansen's disease services, namely (a) people directly affected by Hansen's disease, and (b) clinic staff working closely with persons affected, were included in the study. Data was collected until data saturation was reached, we estimated that this would be reached when at least 10 individuals per participant group were included. People aged <18y and those unable to speak Portuguese were excluded. Healthcare providers were only included if they had specific responsibilities for Hansen's disease services.

Participants were selected through convenience sampling from among those present at the clinic at the time of the group discussions. Potential participants were contacted via the Osvaldo Cruz Policlinic, with general and personal invitations offered to people with understanding of Hansen's disease and Hansen's disease services.

Data collection

Data were collected over a 2-wk period in December 2018. We used semi-structured small focus groups. In these groups, an interview guide with points and questions to trigger and sustain the discussion and engagement of all participants was used. Participants were asked to speak about what in their experience gave them (or people affected by Hansen's disease) strength, courage and the means to face discrimination. In addition to audio recording, the facilitator (second author, individual affected by Hansen's disease, highly experienced in qualitative research), took written notes and sought clarification where required. All focus groups were conducted in a private space in the health facility. Separate focus groups were held for health workers and for people affected. Except for one focus group, participants who were in self-care groups and who were not, were not mixed. All focus group discussions were conducted in Portuguese and audio recorded, the discussions lasted from 19 min to 1 h 33 min.

Data analysis

The audio recordings of the focus group discussions were transcribed verbatim, also noting periods of silence and emotional expressions. Where there were local language expressions, they were edited to common Portuguese grammar. These transcripts were translated to English by the second author (ZBSP) and a translator who are fluent in English and Portuguese, with care to ensure the meaning of terms and expressions was communicated. A total of 234 pages of transcript was reviewed by the three investigators (ZBSP, PK, AvtN). Based on agreed summaries, investigators identified main categories of relevance to the notion of building resilience in the context of Hansen's disease services. Whenever necessary, the original transcription in Portuguese was referred to for clarity.

Ethical considerations

Ethical clearance for this project was provided by the Federal University of Santa Maria, Santa Maria, Brazil as a post-doctoral project within an umbrella project entitled "Precarious lives in the Cyber World" (Approval number 3.750.927). All participants were informed about the objective of the study, the voluntary nature of participation, and of confidentiality of the data. Written informed consent was obtained from all participants before data collection commenced.

Results

Demographics

Thirty-one participants were included in 10 small focus group discussions (2-5 participants per group). Three groups comprised only female participants, one group consisted only males and the other six groups were mixed gender. A total of 23 individuals affected by Hansen's disease and 8 healthcare providers were included. Of the total of 23 people affected by Hansen's disease, 12 (n=12/23) were in self-care groups. These self-care groups consist if persons undergoing MDT, rehabilitation, or both. Their meetings focus on physical self-care and social activities to promote self-confidence and self-esteem.

Most participants were female (n=18/31). The average age of all participants was 54 (range 26-75) y. Most participants did not have a paid job, they received social welfare benefits due to advanced age or due to physical disability (n=17/31). The other participants were unskilled urban workers, a rural worker, a homemaker or were not in an occupation but without receiving any welfare benefits (n=6/31). As a qualitative pilot study, focused on understanding the nature and variety of participants' experiences, rather than comparing groups or variables, we did not collect specific clinical data; however, an overview of the demographic information per participant group can be found in Table 1.

Table 1. Demographic information of participants.

	No. of individuals affected (n=23)	Health providers (n=8)
Average age, median (range)	53 (26-70)	52 (31-67)
Gender, *n* (%)		
Female	12 (52%)	6 (75%)
Male	11 (48%)	2 (25%)
Education, *n* (%)		
No education	4 (17%)	0 (0%)
Primary level incomplete	13 (56%)	0 (0%)
Primary level complete (8 yrs)	3 (13%)	0 (0%)
Secondary level complete (12 yrs)	3 (13%)	1 (12%)
University level complete (16+ yrs)	0 (0%)	7 (87%)
Occupation, *n* (%)		
Health worker (higher education)	0 (0%)	6 (75%)
Health worker (secondary education)	0 (0%)	2 (25%)
Rural worker	1 (4%)	0 (0%)
Home maker	1 (4%)	0 (0%)
Urban unskilled worker	3 (13%)	0 (0%)
Not occupied	1 (4%)	0 (0%)
Social security beneficiary	17 (74%)	0 (0%)
Group support		
Not in self-care groups	11 (48%)	n/a
In self-care groups	12 (52%)	n/a

Based on our focus group feedback, all participants with Hansen's disease described a degree of discrimination in at least some, if not most, aspects of their lives. The group interviews explored 'sources of strength and resilience' for individuals affected by Hansen's disease through open-ended questions. The following themes were identified.

Themes

There is a need for psychosocial support in early treatment to facilitate resilience

Despite the existence of psychological services in the Hansen's disease program in Brazil, several participants noted that they went through the process of diagnosis and early treatment without any formal psychosocial support.

> *"...No doctor helped me! They just gave the medicine, I took the medicine, I did not even tell my story because I was ashamed there..."*
>
> *(FG6, women affected by Hansen's disease).*

Focus group discussions indicated that participants were expecting that psychosocial and adjustment issues would be addressed alongside initial medical care. However, in most cases, they only met medical personnel, who often did not have the time or capacity to focus on these issues.

> *"...When I came in I already spilled everything out right away, I was crying, I was telling everything [to the doctor]..."*
>
> *(FG6, women affected by Hansen's disease).*

Some participants explained that their experience of early treatment and diagnosis undermined their strength and resilience.

The overall picture of early treatment as described in these focus groups was that there were few opportunities for focusing on psychosocial resilience. Likewise, staff explained that such services were not a high priority, and difficult to provide with scheduling challenges and staff turnover. It seemed that the provision of psychological support was not a high priority in this service.

> *"...She [psychologist] does not have a proper corner to talk. [She sees people] there, with everyone else walking in and out, it does not work..."*
>
> *(FG9, men and women affected by Hansen's disease).*

Indeed, this was also true of other services:

> *"...Often, patients from other health facilities, already under treatment, arrive without any [psychological] guidance..."*
>
> *(FG5, health workers).*

However, it was also clear that some early treatment staff took the time to listen and support patients. People affected drew a sense of resilience from the positivity provided by staff.

> *"…The treatment we do here [at the facility], they take good care of us. We receive that strength and encouragement not to give up…"*
>
> *(FG9, men and women affected by Hansen's disease).*

In one focus group, the example was given of a medical practitioner who was very helpful in providing people with psychological and emotional support. This was also noted by service providers. It was clear from this focus group that such support by medical staff was deeply appreciated.

Counselling in early treatment is helpful in supporting resilience

A few participants reported receiving formal psychological support as part of their early treatment. They noted that they had drawn strength from this one-to-one counselling, and described it as crucial, enabling them to understand the effects of the disease, to cope with their situation and start to build their strength.

> *"…It was very difficult! It was the stage of treatment that was the most difficult … [The counsellor] helped me a lot. Because until then … I did not talk to anyone. That thing was trapped inside me. That was very bad for me. I cried a lot. I got depressed once. After I started having psychological counselling, I started to improve…"*
>
> *(FG8, women affected by Hansen's disease).*

> *"…That day, when the doctor told me [I had Hansen's disease], I was finished! Then he saw that I was very worried, so he sent me to talk to her [psychologist]. From that time on I talked to her, it was … It was like growing up, everything changed in me…"*
>
> *(FG9, men and women affected by Hansen's disease).*

Family and social relationships contribute to resilience

In most focus groups, participants talked about relationships and the social support they received as their primary source of strength. Focus group participants described drawing strength and resilience from their relationships with family members, from relationships with close friends, and importantly, from relationships that they had subsequently developed with others affected by Hansen's disease.

In many cases, participants described family as their key source of strength. This was true of both family of origin and for their spouse and children

> *"…For me, what … gave me more strength was my children…"*
> *(FG6, women affected by Hansen's disease.)*

> *"…In my family I said: 'I have this disease here!' But everyone supported me, no one turned their back on me…"*
> *(FG4, men and women affected by Hansen's disease).*

While the importance of family as a source of strength was evident for the majority of participants, we noted that for some, family could be a source of rejection and discrimination.

In some cases, friendships were seen a source of considerable practical support, which enabled participants to be more resilient.

> *"…She said: 'Come here, to my house, with your children.' I with my children! She had five and I three [children]. [She said] 'So let's raise these children…'"*
> *(FG6, women affected by Hansen's disease).*

Some participants described their existing and new friendships with individuals not affected by the disease as a key source of strength.

> *"…My friends always supported me, thank God I never suffered any prejudice!…"*
> *(FG4, men and women affected by Hansen's disease).*

Responses from individuals affected and service providers showed that many people looked to their friendships for emotional strength. In many cases, friends supported their resilience. However, as with families, in a few notable examples, the opposite was true.

Peer relationships contribute to resilience

Beyond the theme of family relationships and friendships as sources of resilience, the majority of our focus group participants emphasized the importance of relationships with peers affected by Hansen's disease, as a source of strength. Relationships with peers were instrumental for psychosocial support and sharing of experience.

> *"…My strength was…my friends from the [self-care group] meeting here…"*
> *(FG10, men and women affected by Hansen's disease).*

Participants saw their relationships with those also affected by Hansen's disease as core to effectively coping and being resilient.

> *"...Because we talk to friends and we get stronger. One is giving strength to the other..."*
>
> *(*FG7, men affected by Hansen's disease).

Indeed, many emphasized that their communication with and relationships with peers comprised of sharing of information, venting of frustrations and sharing of experiences. This reciprocity and ability to identify which each other's experience and situation, resulted in mutual benefit and contributed to their resilience.

> *"...To give strength to others, start helping others who have the disease. You feel stronger!..*."
>
> *(FG9, men and women affected by Hansen's disease).*

Self-care groups and support groups contribute to resilience

A core theme of our focus group discussions related to participants' experience of self-care and support groups as a key source of strength. Interestingly the healthcare providers interviewed also emphasised the importance of peer support, self-care and self-help groups. They emphasised that it is important to actively encourage these groups and to include family members to further reduce stigma:

> *"... She managed to recover, and improved her physiognomy, her self-esteem was up there ... In self-care, she found support..."*
>
> *(FG1, health workers).*

Participants gave strong indications of the psychosocial strength they had derived from self-care and support groups.

> *"... [There are times] that you do not want to do anything... Do not want to see anybody... Do not want to talk to anyone... You feel down ... And do not want to do anything, but the group gives us self-esteem to talk, to be entertained, it is where we tell our problems, where we share what we are feeling... I take it as a life lesson. I listen to them talk and take some lesson for me. I'm sure that what I say, they also take a little bit for them. Because what I've been through... Today I have overcome ... I am already a victor for having gone through what I went through: the difficulties, the sadness, the depression that I went through, and I am here today, and I say: 'I'm very strong, despite all that...'*
>
> *(FG8, women affected by Hansen's disease).*

As with the examples of strength derived from friendships with others affected by Hansen’s disease, many focus group discussions emphasised the sharing of life experience with others. It appears the mutual nature of the discussions helped them build resilience. Despite not being intended for addressing psychosocial concerns, self-care and self-help groups provide an opportunity for learning from those who have been in similar situations, thereby increasing learning and resilience.

Resilience-related benefits described from engaging in such groups include deriving support from and providing support to those who are new or at their most vulnerable.

> *“...We are very strong for each other. When we meet, people talk a lot, when we meet, people give strength to one another. Sometimes we come and see the other, like her [another participant] there who cannot talk, and we talk a lot to her, give her a lot of affection. ... Today she cannot speak, but in a month or two months, she can do it...”*
>
> *(FG8, women affected by Hansen’s disease).*

Self-care and support groups are also a source of fun, which was an important source of strength.

> *“...We get involved, which is very cool. The games are very good, we danced, we played, we jumped, we have fun here, you know, for me the self-care group is very good, especially for my mind...”*
>
> *(FG6, women affected by Hansen’s disease).*

Providing information about Hansen’s disease is beneficial in enhancing resilience

Our focus groups with affected individuals, as well as those with health staff, emphasised the importance of providing accurate and accessible information about Hansen’s disease in building resilience in the face of Hansen’s disease and discrimination. Healthcare providers recognised that correct knowledge can combat stigma and contribute to resilience.

> *“...This stigma is still very much related to ... The ancient leprosy ... There's still this prejudice, this stigma ... The lack of information. I think we have to inform better...”*
>
> *(FG2, health workers).*

People affected also noted the link between information and stigma:

> *"...This lack of knowledge causes stigma..."*
> *(FG7, men affected by Hansen's disease).*

However, they emphasised that such information had benefit for their own adjustment and understanding, and that it was beneficial to discuss the information with peers. In the context of a focus group discussion on the topic of accurate information on how contagious Hansen's disease is, one participant stated:

> *"...It is very good to talk to the doctor, to get rid of the doubts, and to always have a group, so as to be able to ask questions, to talk about the experiences..."*
> *(FG4, men and women affected by Hansen's disease).*

Including beliefs and spirituality is beneficial for greater resilience

Finally, another source of strength and resilience reported by some of our participants was their beliefs, their spiritual life and their relationship with God.

> *"...It is God! God gives us much strength! I seek God! I seek and thank God. He has helped me very, very, very much..."*
> *(FG4, men and women affected by Hansen's disease*).

Many participants referred to praying to God. Participants stressed the importance of religion and the strength they gained from praying.

> *"...I prayed to the Lord, to my Father in Heaven, I prayed a lot to the Lord... My strength, I asked God for it. And I thought a lot about my kids, because I, my problem, was very difficult..."*
> *(FG6, men and women affected by Hansen's disease).*

Discussion

In keeping with the widely recognised psychosocial needs of persons affected by Hansen's disease [26], and the potential importance of psychosocial resilience in the face of discrimination [15], the current qualitative study provides key perspectives on what services might do to help persons affected and families to build strength and resilience. Based on the perspectives of people affected and service providers in Brazil, services should: provide psychosocial support (particularly one-to-one counselling) in early treatment; recognise that family and social relationships are very important for building resilience; foster peer relationships (ideally incorporated into self-care groups); ensure that accurate information about Hansen's disease is provided, and acknowledge the place of beliefs and spirituality in building psychosocial resilience.

The theme of direct psychological support and counselling was key in our findings. This study found that people affected drew strength from psychosocial support and specifically the opportunity for counselling in early treatment. The importance of such support is also stressed in the WHO guidelines for strengthening participation of individuals affected by Hansen's disease in Hansen's disease services, i.e 'psychological support and counselling are crucial to the successful treatment of people diagnosed with leprosy' [27]. It is clear that counselling can help individuals cope with hardship and challenges and, indeed, may help people to become more resilient [28]. In light of indications from our recent literature review [15] and focus group data, it would appear that counselling might also be a vital tool for services to enhance psychosocial resilience in the context of Hansen's disease.

In the current study, those participants who had only been seen by medical personnel (who may not have had the time or capacity to focus on their psychosocial and adjustment concerns), also described feeling that their resilience was undermined. Our focus group participants were clear that formal psychosocial support provided in early treatment aided their resilience. It is noteworthy that this study was conducted in Brazil, where there is a degree of psychological support available in the general Hansen's disease program. While it may be that the presence of some services may have increased awareness of the need for such support, it also provides an indication for those countries where no such services exist, i.e. that people affected may gain great benefit from such support.

Another major finding in our study was the importance of family and other relationships and social support in building resilience. Participants described gaining strength from their relationships and the support they received. While it is generally understood that social relationships and support play a key role in a person's health and psychosocial wellbeing [29], these findings align with some more specific studies which link social support with resilience [18–22,30]. Social support appears to be beneficial for people experiencing stress [31,32], may protect against depression and anxiety [33] and may assist people to cope with stressful events [33].

Substantial literature associates social support with psychosocial wellbeing [31] and quality of life [34,35]. The current findings suggest that such social support is also vitally linked with resilience [20]. Social support can help build resilience because it can be a means of gaining functional, emotional, material and informational assistance [20]. Relationships can increase resilience by helping people to regulate emotions and solve problems by talking through issues [21,36,37].

The importance of the role of family and other relationships for building strength and resilience was reported by all focus groups. This aligns well with the resilience literature, which clearly links such connections with a person's ability to cope with adversity [30,38]. Family relationships provide many of the advantages of social relationships noted above (reducing stress and anxiety, improving quality of life, and providing practical supports) but at a more significant level. While it is also true that some of our participants described family and other relationships as sources of discrimination and stress, it was clear from the responses that they saw the need for services to optimize such relationships, towards building resilience and strength.

Another key finding in our study was the importance of self-care and peer support groups in fostering resilience. Such groups are very common in Hansen's disease services, albeit with a primary focus on ulcer treatment and disability prevention [39,40]. Self-care groups have been seen to have substantial benefits in managing impairment [39,41–43], and group members often report benefits beyond improved wound care, such as in social participation [39,41–43]. There were strong indications from our study that such groups also provide substantial psychosocial benefit and contribute to resilience. Again, this aligns well with previous research identifying such groups as effective for promoting resilience. The benefits of connection to a group of peers is known to have a variety of physical and psychological benefits [33,44–46] and promote resilience [15,20,22]. People affected by Hansen's disease in self-care groups in Ethiopia also reported increased confidence, dignity, self-respect and a sense of belonging [41]. Peer support has been found to build autonomy and community [46], and may enable stigmatised people to develop new ways of thinking about themselves and develop a more positive self-image [46], all vital to a greater sense of resilience. These findings suggest that Hansen's disease services should seek to build resilience in early treatment, possibly by utilising (peer) group counselling.

Our focus group participants also emphasized the importance of accurate information, provided to them, as well as to their families and others around them. For the layperson in Brazil, Hansen's disease is largely unknown, unlike more common tropical diseases that affect large numbers of people. As such many are unaware of it until the point diagnosis [47]. Providing people affected and families with accurate and meaningful information about the disease would appear to challenge myths and stigma [15,48], which also contributes to resilience. Indeed the provision of information is a vital dimension of most interventions for resilience in the face of discrimination [15].

Finally, another common source of strength and resilience reported in the present study was participant's beliefs and their spiritual life. Studies in other fields have also found that spirituality and faith can increase resilience to stigma [49,50]. Among our participants, in response to questions about 'sources of strength' there were clear indications that beliefs were important. Earnshaw and colleagues [50] argued that spirituality can restore self-worth and perception of sense of control. These would appear to be particularly valuable assets in the face of stigma and discrimination. It has similarly been reported in the literature that spirituality can buffer people from the negative impact of stigma, that is, the meaning-making and framing that comes through faith, can enhance resilient behaviours [49,50]. Our results align closely with the observation that relying on faith can provide individuals with a sense of control, positively contributing to resilience and the capacity to overcome adversity [51].

While the above themes were apparent in the feedback provided by focus group members and align with indications in literature from a variety of sources, it should be acknowledged that this study also had a number of limitations. First, although we did not seek a large number of participants, the sample for this study was small, highly localised and narrowly selected. Typically qualitative studies such as this seek to explore in-depth with a few participants rather than strive for a representative sample, so while the study is appropriate for its intended purpose, this information may not be representative of all individuals affected.

Second, including service providers in our sample was both a strength and a limitation. The strength resided in being able to include service provider perspectives (given that our interest is relevance to current service provision), however recognising their small number led us to conclude that a separate study focus on a diversity of service providers across a variety of locations may have been preferable.

Third, some factors that are unique to the Brazilian context should also be acknowledged. As noted above, Brazilian public health services which include psychological services may not be representative of Hansen's disease services in other endemic countries. Likewise, the strongly Roman Catholic and Christian profile of the Brazilian population may have influenced our findings, but may not necessarily be representative of other Hansen's disease-endemic countries.

Finally, it should be acknowledged that while the above themes are informative from the perspective of key stakeholders, they are not necessarily going to be effective in increasing resilience. The issue of effectiveness is a topic for future research. In reality, some aspects which deplete or prevent resilience may not be amenable to change within the resources of Hansen's disease services. However, despite these qualifications, the current study provides some useful indicators of what actions services might support or initiate to help individuals affected and their families to build psychosocial strength and resilience.

Conclusions

Based on the current findings, interventions for promoting resilience for people affected by Hansen's disease who face stigma and discrimination could be targeted at two main points. First, during the initial part of treatment, after diagnosis, there would appear to be substantial benefit in providing resilience-focused counselling. Providing such a safe environment for people to express emotions and concerns early on was strongly recommended.

Second, during treatment and beyond, having people affected getting together (in self-care or self-help groups) seems to be beneficial. This is likely to help people experience a sense of partnership and a shared understanding of resilience. Across both settings, the importance of supporting family and social relationships, providing accurate information, and acknowledging spiritual beliefs are important.

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References

1. Rafferty J. Curing the stigma of leprosy. Lepr Rev 2005;76:119–26.
2. Cross H. Interventions to address the stigma associated with leprosy: a perspective on the issues. Psychol Health Med 2006;11:367–73.
3. Litt E, Baker MC, Molyneux D. Neglected tropical diseases and mental health: a perspective on comorbidity. Trends Parasitol 2012;28:195–201.
4. Tsutsumi A, Izutsu T, Islam MDA, Amed JU, Nakahara S, Takagi F, et al. Depressive status of leprosy patients in Bangladesh: association with self-perception of stigma. Lepr Rev 2004;75:57–66.
5. Brouwers C, van Brakel W, Cornielje H. Quality of life, perceived stigma, activity and participation of people with leprosy-related disabilities in south-east Nepal. Disabil CBR Incl Dev 2011;22:16–34.
6. Stevelink SAM, Van Brakel WH, Augustine V. Stigma and social participation in Southern India: Differences and commonalities among persons affected by leprosy and persons living with HIV/AIDS. Psychol Health Med 2011;16:695–707.
7. Weiss MG. Stigma and the social burden of neglected tropical diseases. PLoS Negl Trop Dis 2008;2:e237.
8. Van Brakel WH, Sihombing B, Djarir H, Beise K, Kusumawardhani L, Yulihane R, et al. Disability in people affected by leprosy: the role of impairment, activity, social participation, stigma and discrimination. Glob Health Action 2012;5:18394.
9. Singh GP. Psychosocial aspects of Hansen's disease (leprosy). Indian Dermatol Online J 2012;3:166.
10. Tsutsumi A, Izutsu T, Islam AM, Maksuda AN, Kato H, Wakai S. The quality of life, mental health, and perceived stigma of leprosy patients in Bangladesh. Soc Sci Med 2007;64:2443–53.
11. Lustosa AA, Nogueira LT, Pedrosa JI dos S, Teles JBM, Campelo V. The impact of leprosy on health-related quality of life. Rev Soc Bras Med Trop 2011;44:621–6.
12. Yamaguchi N, Poudel KC, Jimba M. Health-related quality of life, depression, and self-esteem in adolescents with leprosy-affected parents: results of a cross-sectional study in Nepal. BMC Public Health 2013;13:22.
13. Rocha-Leite CI, Borges-Oliveira R, Araújo-de-Freitas L, Machado PRL, Quarantini LC. Mental disorders in leprosy: An underdiagnosed and untreated population. J Psychosom Res 2014;76:422–5.
14. Cook JE, Purdie-Vaughns V, Meyer IH, Busch JTA. Intervening within and across levels: A multilevel approach to stigma and public health. Soc Sci Med 2014;103:101–9.
15. Van't Noordende AT, Kuipers P, Pereira ZBD. Strengthening personal and family resilience: a literature review for the leprosy context. Lepr Rev 2019;90:88–104.
16. Walsh F. Family resilience: A framework for clinical practice. Fam Process 2003;42:1–18.
17. Xu J, Ou L. Resilience and quality of life among Wenchuan earthquake survivors: the mediating role of social support. Public Health 2014;128:430–7.
18. MacLeod S, Musich S, Hawkins K, Alsgaard K, Wicker ER. The impact of resilience among older adults. Geriatr Nurs (Minneap) 2016;37:266–72.

19. Connor KM, Davidson JRT. Development of a new resilience scale: The Connor-Davidson resilience scale (CD-RISC). Depress Anxiety 2003;18:76–82.
20. Southwick SM, Sippel L, Krystal J, Charney D, Mayes L, Pietrzak R. Why are some individuals more resilient than others: the role of social support. World Psychiatry 2016;15:77.
21. Earvolino-Ramirez M. Resilience: A Concept Analysis. Nurs. Forum, vol. 42, 2007, p. 73.
22. Pietrzak RH, Johnson DC, Goldstein MB, Malley JC, Rivers AJ, Morgan CA, et al. Psychosocial buffers of traumatic stress, depressive symptoms, and psychosocial difficulties in veterans of Operations Enduring Freedom and Iraqi Freedom: the role of resilience, unit support, and postdeployment social support. J Affect Disord 2010;120:188–92.
23. Edward K, Warelow P. Resilience: When coping is emotionally intelligent. J Am Psychiatr Nurses Assoc 2005;11:101–2.
24. Hofstraat K, van Brakel WH. Social stigma towards neglected tropical diseases: a systematic review. Int Health 2016;8:i53–70.
25. Brazil G of. Informações de Saúde (TABNET) - DATASUS 2020. http://tabnet.datasus.gov.br/cgi/tabcgi.exe?sinannet/cnv/hanswro.def (accessed November 18, 2020).
26. Scott J. The psychosocial needs of leprosy patients. Lepr Rev 2000;71:486–91.
27. Organization WH. Guidelines for strengthening participation of persons affected by leprosy in leprosy services. WHO Regional Office for South-East Asia; 2011.
28. Enache R. Psychological counseling and developing resilience for young people with psychological trauma in childhood or adolescence. J Exp Psychother 2016;19:73.
29. Tough H, Siegrist J, Fekete C. Social relationships, mental health and wellbeing in physical disability: a systematic review. BMC Public Health 2017;17:414.
30. Walsh F. Family resilience: a developmental systems framework. Eur J Dev Psychol 2016;13:313–24.
31. Thoits PA. Mechanisms linking social ties and support to physical and mental health. J Health Soc Behav 2011;52:145–61.
32. Thoits PA. Stress, coping, and social support processes: Where are we? What next? J Health Soc Behav 1995:53–79.
33. Gurung R, Sarason B, Sarason I. Close personal relationships and health outcomes: A key to the role of social support. Handb Pers Relationships Theory, Res Interv (2nd Ed) Chichester, UK Wiley 1997:547–73.
34. Baumeister RF, Leary MR. The need to belong: desire for interpersonal attachments as a fundamental human motivation. Psychol Bull 1995;117:497.
35. Connell J, Brazier J, O’Cathain A, Lloyd-Jones M, Paisley S. Quality of life of people with mental health problems: a synthesis of qualitative research. Health Qual Life Outcomes 2012;10:138.
36. Cohen S. Psychosocial models of the role of social support in the etiology of physical disease. Heal Psychol 1988;7:269.
37. Hatzenbuehler ML, Nolen-Hoeksema S, Dovidio J. How does stigma “get under the skin”? The mediating role of emotion regulation. Psychol Sci 2009;20:1282–9.
38. Walsh F. Strengthening family resilience. Third. Guilford Publications; 2015.
39. Ebenso J, Muyiwa LT, Ebenso BE. Self care groups and Ulcer prevention in Okegbala Nigeria. Lepr Rev 2009;80:187.

40. Rahmawati I. The experiences of people affected by leprosy who participated in self-care groups in the community: A qualitative study in Indonesia. Lepr Rev 2017;88:543–53.
41. Benbow C, Tamiru T. The experience of self-care groups with people affected by leprosy: ALERT, Ethiopia. Lepr Rev 2001;72:311–21.
42. Cross H, Choudhary R. STEP: an intervention to address the issue of stigma related to leprosy in Southern Nepal. Lepr Rev 2005;76:316–24.
43. Deepak S, Hansine PE, Braccini C. Self-care groups of leprosy-affected people in Mozambique. Lepr Rev 2013;84:283.
44. Hale CJ, Hannum JW, Espelage DL. Social support and physical health: The importance of belonging. J Am Coll Heal 2005;53:276–84.
45. Solomon P. Peer support/peer provided services underlying processes, benefits, and critical ingredients. Psychiatr Rehabil J 2004;27:392.
46. Mead S, Hilton D, Curtis L. Peer support: A theoretical perspective. Psychiatr Rehabil J 2001;25:134.
47. Lira KB, Leite JJG, Maia DCB de S, Freitas R de MF, Feijão AR. Knowledge of the patients regarding leprosy and adherence to treatment. Brazilian J Infect Dis 2012;16:472–5.
48. van't Noordende AT, Korfage I, Lisam S, Arif MA, Kumar A, van Brakel WH. The role of perceptions and knowledge of leprosy in the elimination of leprosy: A baseline study in Fatehpur district, northern India. PLoS Negl Trop Dis 2019;13:e0007302.
49. Ögtem-Young Ö. Faith resilience: Everyday experiences. Societies 2018;8:10.
50. Earnshaw VA, Bogart LM, Dovidio JF, Williams DR. Stigma and racial/ethnic HIV disparities: moving toward resilience. 2015.
51. Yasien S, Nasir JA, Shaheen T. Relationship between psychological distress and resilience in rescue workers. Saudi Med J 2016;37:778.

CHAPTER 10

Strengthening individual and family resilience against leprosy-related discrimination: a pilot intervention study

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Abstract

Background

Leprosy and leprosy-related stigma can have a major impact on psychosocial wellbeing of persons affected and their family members. Resilience is a process that incorporates many of the core skills and abilities which may enable people to address stigma and discrimination. The current study aimed to develop and pilot an intervention to strengthen individual and family resilience against leprosy-related discrimination.

Methodology

We used a quasi-experimental, before-after study design with a mixed methods approach. The 10-week family-based intervention was designed to strengthen the resilience of individuals and families by enhancing their protective abilities and capacity to overcome adversity. The study was conducted in two sites, urban areas in Telangana state, and in rural areas in Odisha state, India. Persons affected and their family members were included using purposive sampling. Two questionnaires were used pre-and post-intervention: the Connor-Davidson Resilience Scale (CD-RISC, maximum score 100, with high scores reflecting greater resilience) and the WHOQOL-BREF (maximum score of 130, with higher scores reflecting higher quality of life). In addition, semi-structured interviews were conducted post-intervention. Data were collected at baseline, a few weeks after completion of the intervention, and in the Odisha cohort again at six months after completion. Paired t-tests measured differences pre- and post- intervention. Qualitative data were thematically analysed.

Findings

Eighty participants across 20 families were included in the study (23 persons affected and 57 family members). We found a significant increase in CD-RISC scores for persons affected and family members from Odisha state (baseline 46.5, first follow-up 77.0, second follow-up 70.0), this improvement was maintained at six-month follow-up. There was no increase in CD-RISC scores post-intervention among participants from Telangana state. WHOQOL-BREF scores were significantly higher at follow-up for persons affected in both states, and for family members in Odisha state. No families dropped out of the study. In the qualitative feedback, all participants described drawing benefit from the programme. Participants especially appreciated the social dimensions of the intervention.

Conclusion

This pilot study showed that the 10-week family-based intervention to strengthen resilience among persons affected by leprosy and their family members was feasible, and has the potential to improve resilience and quality of life. A large-scale efficacy trial is necessary to determine the effectiveness and long-term sustainability of the intervention.

Introduction

Leprosy is a chronic infectious disease caused by *Mycobacterium leprae* [1]. Although curable by a combination of drugs, the disease may damage the peripheral nerves and skin, which, especially if diagnosed late, can result in impairment [1]. Such impairments and other factors (including social myths, attitudes cultural and religious beliefs and fear), may lead to persons affected by leprosy being stigmatised [2,3]. Social stigma may impact the emotions, thoughts, behaviour and relationships of persons who are stigmatised [4]. Not surprisingly, leprosy has been associated with anxiety disorders, depression, suicide (attempts), mental distress and emotions such as fear and shame, low self-esteem and reduced quality of life [5]. Close contacts of persons affected, such as family members and friends, may also be negatively impacted by such social and psychological consequences of the disease [5].

Though leprosy can have a major impact on psychosocial wellbeing [5], there is also evidence that persons affected by leprosy can overcome experiences of discrimination and exclusion [6]. Studies indicate that people who have faced severe stigmatisation and have gone on to overcome this adversity demonstrate numerous dimensions of resilience [7,8]. They also tend to show normal or high levels of mental wellbeing despite exposure to psychological or physical adversity [9]. It would appear that resilience is a process that incorporates many of the core skills and abilities which may be required to address stigma and discrimination.

Resilience can be understood as a dynamic and complex process (as well as outcome) of successfully adapting to difficult or challenging life experiences, especially through mental, emotional and behavioral flexibility and adjustment to external and internal demands [10,11]. Many factors can contribute to how well or poorly people adapt to adversity, including how they perceive and engage with the environment, their social resources and their use of coping strategies [12–15]. Many of these “protective” or adaptive factors can be learned and strengthened, such as emotional regulation, self-efficacy, support seeking behaviour, communication skills, problem solving abilities and engaging in a supportive environment [12–15]. In the face of stressors, these protective factors are said to modify the individual’s or groups’ response to adversity, thereby reducing the likelihood of negative outcomes [15].

Evidence-based approaches to building resilience within families have been described [12], including where a family member has health and disability problems [16], or is stigmatised and in resource-poor settings [17]. Versions of these approaches are now also conducted as brief interventions [18], addressing family stress, conflict, cohesion, adaptation, working through adversity, beliefs and spirituality, and broader communication beyond the family. Unfortunately, most of them are highly resource intensive, with even the brief interventions requiring multiple counselling sessions with each family over many months. In addition, most are not designed for neglected tropical disease (NTD) services such as leprosy programmes. The key challenge is how to make these highly strategic interventions more suited to the realities of treatment, rehabilitation and other services, and thereby more accessible to people in leprosy-endemic countries.

The current study aimed to develop and pilot an intervention to strengthen individual and family resilience against leprosy-related discrimination. It is based on a scoping review [19] to determine principles of evidence-based interventions to strengthen personal and family resilience, as well as a qualitative exploration on sources of strength and resilience [20].

Methods

Ethics statement

Ethical approval was obtained from the Technical Advisory Group and the Institutional Ethics Committee of the Lepra-Blue Peter Public Health and Research Centre (BPHRC). Written informed consent was obtained from all participants prior to data collection.

Study design and study area

We used a quasi-experimental, before-after study design using mixed methods data collection. The study was conducted in urban areas in Hyderabad district in Telangana state and in rural areas in Subarnnapur, Nabarangpur and Koraput districts in Odisha state, India. In 2019, the prevalence of leprosy was 1.45 per 10,000 population in Odisha state and 0.62 per 10,000 population in Telangana state (State NLEP report, 2019-2020).

Description of the intervention

The intervention was designed to strengthen the resilience of families struggling with leprosy-related discrimination, by strengthening their protective abilities and capacity to overcome adversity. The contents of the intervention were based on a recent scoping review [19], a qualitative exploration on sources of strength and resilience [20], determinants of individual, family and community resilience among families in low- and middle-income contexts [21] and the family resilience framework [12]. An overview of the content of each session and corresponding resilience factors can be found as supporting information file (S1 Text).

Our scoping review identified a number of key process-related factors: the intervention should have more than one session, it should be spread over several weeks, it should ideally involve persons affected themselves in planning or executing the intervention, it should be targeted to fit the audience and it should include multiple intervention components [19]. The intervention to strengthen resilience was therefore designed to have ten weekly sessions in a family-based format. Each session adopted an action learning and problem-solving approach to the activities design. In addition, a separate session about rights was added, since our scoping review identified a human rights foundation as an important component [19]. In addition, our qualitative exploration on sources of strength and resilience identified the importance of supporting family and social relationships, providing accurate information about leprosy, and acknowledging spiritual beliefs [20]. We therefore added a separate session about knowledge about leprosy, and also integrated these other concerns. The scoping review endorsed the importance of providing correct knowledge about the stigmatized condition to empower the individual or group to challenge misconceptions about their condition.

While it was not logistically possible to include persons affected in executing the intervention for this pilot, the study team included a person affected (ZBSP), and the intervention was developed with reference to the ILEP Advisory Panel, comprising women and men affected by leprosy. In addition, a three-day interactive training workshop was conducted in Hyderabad by three authors with the staff members who had been selected to deliver the intervention. In this workshop the assessments were presented and discussed, and the intervention content and resource materials were discussed and refined.

In order to make the concept of resilience more tangible to the participants (largely poor Indian families), bamboo was chosen as emblematic of the intervention pictorially and as a catch-phrase: ‘being strong and bouncing back like the bamboo in a storm’. We identified four main themes: strong roots of the bamboo plant (knowledge, sessions 1-2), strong trunk (thoughts and behaviour, sessions 3-5), strong branches and leaves (rights and spirituality, sessions 6-7), and strong soil (relationships and social support, sessions 8-10).

Participants and sampling procedure

Persons who had been treated for leprosy (‘persons affected’) and their family members were included in the study. Participants lived in urban slum areas of Hyderabad, and in rural tribal areas of Subarnapur, Nabarangpur and Koraput districts of Odisha. Family members included in the study were all people in the same household as the identified person affected by leprosy, they were related by blood or by marriage. Persons below the age of 18 and those unwilling or unable to give informed consent were excluded. Those living in single person households were also excluded.

Participants were purposively sampled; contacted through the networks of Lepra Society (Odisha cohort) and Hyderabad Leprosy Control & Health Society (HLCHS) (Telangana cohort). Identified families were visited in their home to invite them to participate and to check whether they met the inclusion criteria. Sampling also sought a mix of women and men affected by leprosy, of different ages.

According to Hertzog [22], if the aim of the pilot study is to demonstrate intervention efficacy in a single group, a sample size of 20-25 is adequate. A sample size of 10-20 participants per group is adequate when trying to determine group differences [22]. Since this was a proof of concept (pilot) study in which we wanted to explore outcomes of a trial intervention, but also wanted to determine if there are any differences between rural and urban areas, we aimed to include families from rural as well as urban areas.

Data collection

Data collection tools

Demographic information gathered included sex, age, education, occupation, religion, role in the family, and for persons affected, also included disability grade and year of diagnosis. We used two validated questionnaires: the Connor-Davidson Resilience Scale (CD-RISC) and WHOQOL-BREF. In addition, semi-structured interviews to assess participant satisfaction were conducted with each family post-intervention, with facilitators taking notes of the responses of participants to each question.

The CD-RISC was used to assess (protective factors related to) resilience. The scale comprises of 25 items, each item can be rated from 0 ('not true at all') to 4 ('true nearly all the time'). A total score of 100 can be obtained, with high scores reflecting greater resilience [23]. The CD-RISC has been validated in Urdu [24] and Hindi [25]. The CD-RISC has not been validated in Odia language (spoken in Odisha), so we used an external language expert to translate it (using both English and Hindi versions as reference). Local bilingual supervisors checked and corrected this version, which was subsequently cross checked by bilingual implementing staff. Supervisors and implementing staff then developed local protocols for administering the scale in the local context.

The WHOQOL-BREF was used to assess quality of life, a concept that is closely linked to well-being and broadly encompasses someone's perception of how 'good' several aspects of their life are [26]. The WHOQOL-BREF comprises 26 items, each rated from 1 (negative response) to 5 (positive response). A total score can be obtained for each of the four domains: physical health (raw total score 35), psychological (30), social relationships (15) and environment (40). Question 1 (overall perception of quality of life) and question 2 (overall perception of health) are not included in the domain scores [27]. Even though there is no official total score for the WHOQOL-BREF [27], we calculated a sum score of 130 by adding the scores obtained on each of the 26 questions to be able to compare the total sum scores pre- and post-intervention. This was done after transforming the scores on negatively framed questions (question 3, 4 and 26). Higher scores on the WHOQOL-BREF denote higher quality of life. The WHOQOL-BREF has been validated in Urdu [28], Hindi [29] and Odia [30].

In addition, staff records were maintained documenting session dates, number of participants and staff comments. Both the intervention and the interviews were conducted by trained facilitators working in leprosy for several years, in participants' homes.

Phases of follow-up

Three rounds of data collection were completed in Odisha state: the baseline assessment was conducted in October 2019, the first follow-up in February 2020 (one week after the intervention had been completed) and the second follow-up in August 2020 (six months after the intervention had been completed). Two rounds of questionnaires were conducted in Telangana state: the baseline assessment was conducted in November 2019 and the follow-up in April 2020 (three and a half weeks after the intervention had been completed). A subsequent follow-up interview was conducted by telephone in Telangana state in October 2020 (six months after the intervention had been completed). Telephone administration was used due to the high number of people affected by COVID-19 in the area.

Data analysis

Quantitative data were collected on paper forms and entered in a database created using Epi Info. The data were analysed using IBM SPSS Statistics 24. Differences between participants from Odisha and Telangana state were evaluated using the Mann-Whitney U test for continuous variables, X2 statistics for categorical variables and Fisher's Exact test for categorical variables for which the expected values in one of the cells of the contingency table was less than five. Frequencies and percentages were calculated to depict demographic information of the participants. Median total scores and the interquartile range (IQR) of the CD-RISC and WHOQOL-BREF were calculated for the different participant groups to summarize resilience and quality of life scores pre- and post-intervention. The Wilcoxon signed-rank test was performed to check whether the scores were significantly different pre- and post- intervention. In addition, we created a new variable that contained the absolute difference between the baseline and follow-up scores of the CD-RISC and WHOQOL-BREF and used bootstrapped stepwise multivariate linear regression with backward elimination to see if there were associations between these variables and the other variables in our dataset (sex, age, participant type, occupation, education, role in the family, and in Telangana state religion also). Bootstrapping was performed to correct for non-normality of the data. Variables were included in the model if they had a p-value of <0.2 in univariate analysis. We made separate models for each state. Statistical significance level was set a priori at $p<0.05$. All data were anonymised before data analysis.

Qualitative data (detailed notes of family responses to interview questions, as well as staff records of weekly meetings) were thematically analysed, identifying key elements of the intervention described as beneficial by participants.

Results

Demographic information

A total of 80 participants were included in the study. A little over half of the participants (n=41, 51%) were from urban areas in Telangana state. The median age of the participants was 35 years. Half of the participants had not had any (formal) education (n=40, 50%). About one third had paid work (n=24, 30%), and a third were unemployed (n=23, 29%). While all participants in Odisha state were Hindu (n=39, 100%), the participants in Telangana state were either Muslim (n=21, 51%) or Hindu (n=20, 49%). An overview of all participant characteristics can be found in Table 1.

Table 1. Demographic information of the study population.

	Odisha state (n=39)	Telangana state (n=41)	Total (n=80)	p-value[b]
Age, median (interquartile range)	35.0 (24.0-50.0)	36.0 (27.0-59.0)	35.0 (25.3-55)	0.187
Sex, *n* (%)				0.642
Female	22 (56.4)	21 (51.2)	43 (53.8)	
Male	17 (43.6)	20 (48.8)	37 (46.2)	
Living area, *n* (%)				0.000
Rural	39 (100.0)	0 (0.0)	39 (48.8)	
Urban	0 (0.0)	41 (100.0)	41 (51.3)	
Language, *n* (%)				0.000
Hindi	1 (2.6)	19 (46.3)	20 (25.0)	
Urdu	1 (2.6)	20 (48.8)	21 (26.3)	
Odia	37 (94.9)	0 (0.0)	37 (46.3)	
Telegu	0 (0.0)	18 (43.9)	18 (22.5)	
Religion, *n* (%)				0.000
Hindu	39 (100.0)	20 (48.8)	59 (73.8)	
Muslim	0 (0.0)	21 (51.2)	21 (26.3)	
Occupation at baseline, *n* (%)				0.001
Paid work	9 (23.1)	15 (36.6)	24 (30.0)	
Unemployed	6 (15.4)	17 (41.5)	23 (28.8)	
Other[a]	24 (61.5)	9 (22.0)	33 (41.3)	
Education, *n* (%)				0.733
No or no formal education	21 (53.8)	19 (46.3)	40 (50.0)	
Primary	12 (30.8)	16 (39.0)	28 (35.0)	
Secondary or higher	6 (15.4)	6 (14.6)	12 (15.0)	
Participant type, *n* (%)				0.697
Persons affected	12 (30.8)	11 (26.8)	23 (28.8)	
Family member	27 (69.2)	30 (73.2)	57 (71.3)	

[a] Occupation 'other' included non-paid work, self-employed and retired.

[b] The tests used are the Mann-Whitney U test for continuous variables (age), X2 statistics for categorical variables (sex, occupation, education and participant type) and Fisher's Exact test for categorical variables for which the expected values in one of the cells of the contingency table was less than five (living area, language and religion).

Persons affected (n=23, 29%) and family members (n=57, 71%) were included. Most persons affected (n=16, 70%) had grade 2 disabilities, followed by grade 0 (n=5, 22%) and grade 1 (n=4, 17%). Most persons affected were diagnosed between five and ten years ago (n=14, 61%). Only one participant (n=1, 4%) was diagnosed over ten years ago. Family members included caregivers (n=21, 37%), breadwinners (n=14, 25%), or heads of the household/decision maker (n=23, 35%).

Family-based sessions

In most cases, about four people participated in the intervention (range 3-6). The intervention consisted of ten weekly sessions that were held in participants' homes. On average, all family members were present for eight of the ten sessions. Each session lasted on average 63 minutes (74 minutes in Odisha state and 51 minutes in Telangana state). In general, the duration of each session increased with family size.

Short-term impact on resilience and quality of life

Table 2 shows the median difference in CD-RISC (resilience) and WHOQOL-BREF (quality of life) scores between the baseline and first follow-up assessment. Higher scores reflect greater resilience and higher quality of life. The increase in resilience scores is significant for persons affected and family members from Odisha. There is no significant improvement in CD-RISC scores among persons from Telangana state pre- and post-intervention. However, there is a significant improvement post-intervention when only looking at the Hindu participants from Telangana state (Wilcoxon signed-rank test, p=0.048) In addition, there is a significant increase in quality of life scores for all participant groups pre- and post-intervention, except for family members from Telangana state (Table 2).

Table 2. Difference in baseline and first follow-up in resilience scores (CD-RISC) and quality of life scores (WHOLQOL-BREF).

		Baseline Median (IQR)	Follow-up Median (IQR)	Difference (%)	p-value[a]
CD-RISC	Participants from Odisha (n=38)	46.5 (39.8-56.0)	77.0 (68.0-86.0)	30.5 (65.6)	0.000
	Persons affected from Odisha (n=12)	40.5 (32.3-57.0)	75.0 (65.0-84.8)	34.5 (85.2)	0.002
	Family members from Odisha (n=26)	47.5 (42.0-56.0)	77.0 (68.0-87.0)	29.5 (62.1)	0.000
	Participants from Telangana (n=41)	49.0 (46.5-51.5)	50.0 (47.0-52.0)	1.0 (2.0)	0.471
	Persons affected from Telangana (n=11)	49.0 (44.0-50.0)	47.0 (43.0-50.0)	2.0 (4.1)	1.000
	Family members from Telangana (n=30)	49.0 (47.0-53.3)	50.0 (47.0-52.3)	1.0 (2.0)	0.362

		Baseline Median (IQR)	Follow-up Median (IQR)	Difference (%)	p-value[a]
WHOQOL-BREF	Participants from Odisha (n=39)	75.0 (67.0-81.0)	100.0 (94.0-105.0)	25.0 (33.3)	0.000
	Persons affected from Odisha (n=12)	66.5 (62.5-77.3)	97.0 (92.5-101.8)	30.5 (45.9)	0.002
	Family members from Odisha (n=27)	77.0 (73.0-83.0)	101.0 (94.0-107.0)	24.0 (31.2)	0.000
	Participants from Telangana (n=41)	69.0 (62.0-73.5)	76.0 (67.5-79.5)	7.0 (10.1)	0.004
	Persons affected from Telangana (n=11)	65.0 (62.0-69.0)	78.0 (71.0-81.0)	13.0 (20.0)	0.010
	Family members from Telangana (n=30)	75.5 (65.0-79.0)	71.5 (62.0-74.3)	4.0 (5.3)	0.108

[a] P-value of the baseline versus first follow-up scores, calculated using the Wilcoxon signed-rank test.

An overview of the median difference pre- and post-intervention per question of the CD-RISC and WHOQOL-BREF and of the difference in domain scores on the WHOQOL-BREF can be found as supporting information file (S2 Text).
In Odisha state, all domains of the WHOQOL-BREF (physical health, psychological, social relationships and environment) significantly improved post-intervention. In Telangana state, only the domains social relationships and environment significantly improved post-intervention (S2 Text).

Factors associated with short-term increase in scores

Some factors were associated with short-term increase on the CD-RISC and WHOQOL-BREF scales (please see S3 Text). We developed two models for each state, given the large differences in median increase on the two scales per state.

Multivariate analysis showed that participants from Odisha state, with occupation ‘other’ (e.g. day labourer) had significantly less improvement on the CD-RISC between baseline and the first follow-up assessment (Table 1). This model explained 16% of the variability of increase in resilience score in Odisha state. In addition, multivariate analysis showed that men had significantly more improvement on the WHOQOL-BREF between baseline and the first follow-up assessments (Table 2). This model explained 17% of the variability of increase in quality of life score in Odisha state.

Multivariate analysis of the data from Telangana state showed that participants who had another occupation than paid work and participants who were Hindu had significantly more improvement on the CD-RISC between baseline and the first follow-up assessments (Table 3). This model explained 24% of the variability of increase in resilience score in Telangana state. In addition, analysis showed that persons affected by leprosy and participants who were Hindu had significantly more improvement on the WHOQOL-BREF between baseline and the first follow-up, this model explained 50% of the variability of increase in quality of life score in Telangana state – religion alone explained 40% of the variability (r-squared 0.401).

Impact on resilience and quality of life after six months

Participants from Odisha state underwent an additional follow-up assessment at six months post-intervention, which coincided with the COVID-19 lockdowns in India. Table 3 shows the median difference in CD-RISC and WHOQOL-BREF scores between baseline, first follow-up and second follow-up assessments in Odisha state. All median scores decreased between first and second follow-up. This decrease was significant for all subgroups on both scales, except for the resilience score of persons affected by leprosy in Odisha state. Even though the resilience and quality of life scores decreased between first and second follow-up, the scores for all subgroups on both scales remained significantly higher than baseline (Table 3).

Table 3. Difference in baseline and first and second follow-up in resilience scores (CD-RISC) and quality of life scores (WHOQOL-BREF) in Odisha state.

		Baseline Median (IQR)	First follow-up Median (IQR)	Second follow-up Median (IQR)	p-value baseline versus first follow-up	p-value baseline versus second follow-up
CD-RISC	All participants from Odisha (n=38)	46.5 (39.8-56.0)	77.0 (68.0-86.0)	70.0 (64.0-79.0)	0.000	0.000
	Persons affected from Odisha (n=12)	40.5 (32.3-57.0)	75.0 (65.0-84.8)	73.5 (61.5-86.5)	0.002	0.002
	Family members from Odisha (n=26)	47.5 (42.0-56.0)	77.0 (68.0-87.0)	69.0 (65.0-76.0)	0.000	0.000
WHOQOL-BREF	Participants from Odisha (n=39)	75.0 (67.0-81.0)	100.0 (94.0-105.0)	92.0 (87.0-98.0)	0.000	0.000
	Persons affected from Odisha (n=12)	66.5 (62.5-77.3)	97.0 (92.5-101.8)	91.5 (87.5-95.0)	0.002	0.002
	Family members from Odisha (n=27)	77.0 (73.0-83.0)	101.0 (94.0-107.0)	95.0 (86.0-100.0)	0.000	0.000

[a] We used the Wilcoxon signed-rank test..

Qualitative data

It was evident from the interview notes that participants greatly valued the 10-week program and enjoyed the practical, vignette- and story-based approach. Qualitative feedback indicated that participants enjoyed the social dimensions of the program. In feedback some noted that the project gave them more confidence to “develop relationship(s) with others” (AD:BS).

Across interview notes there were clear indications that the family-based approach enabled greater connection and social strengthening, promoting understanding and acceptance “We could discuss about health issue together with family members by this programme” (AJ:PN), and that it “brought the change among us” (BS:NN).

Nearly all participants mentioned that they appreciated and drew benefit from the visual image and repeated metaphor of ‘being strong and bouncing back like the bamboo in a storm’. Interviews also reflected that participants understood that resilience was multifactorial, including health related, psychological, behavioural, family, social, rights and other dimensions. All interviewees expressed some degree of improvement in at least some of these aspects.

Across all participants, week 10 (social activity with peers), week 2 (knowledge about leprosy), week 6 (understanding your rights), week 8 (family relationships) and week 3 (positive thinking, understanding thoughts and emotions) were identified as the most beneficial. A number of participants indicated that they thought that week 7 (spirituality) could be improved.

Analysis of staff weekly notes about the 10-week program indicated that staff seemed to appreciate the detailed program which highlighted a number of issues for discussion and action. In many settings, even community agencies, leprosy services are quite narrow, focusing on treatment and follow-up. Notes indicated that for some staff, the project was an entry point to addressing a broader range of important issues for families. The manual appeared to give staff both the content and frameworks to promote discussion on a number of topics, and offer more psycho-socially oriented support. Staff seemed to like the program approach of working with families (as opposed to individuals) and used the program to incorporate a number of psycho-emotional, social, rights-based, and practical issues (pertaining to benefits and treatments) into their service provision.

Discussion

The high prevalence and negative impact (including psychosocial impact) of leprosy-related stigma is well described in the literature [31–36]. However, there are few psychosocial and resilience interventions designed to assist people in NTD and leprosy programs. Further, many of the existing resilience interventions are highly resource intensive and suited to Western contexts. While it is clear that resilience can be strengthened [12–15,19,37,38], the present pilot study indicates the potential of such actions in the context of leprosy-related discrimination. Findings from this study suggest that a 10-week family-based intervention to strengthen resilience among persons affected by leprosy and their family members is feasible, and has the potential to improve resilience and quality of life.

Content of intervention

The intervention in the present study consisted of ten sessions that focused on four main themes: knowledge, thoughts and behaviour, rights and spirituality, and relationships and social support. These broad themes incorporated elements similar to those found in a recent review by Chmitorz and colleagues [39], who reviewed and evaluated 43 randomized controlled training programs to foster psychological resilience. They found that these programmes most often included cognitive restructuring, stress management, problem-solving and coping strategies.

While most components of the current intervention were based on resilience factors relevant to families in low- and middle-income contexts [12,21], the sessions about ‘knowledge’ and ‘rights’ were added based on our recent scoping review [19] and qualitative exploration of sources of strength and resilience [20]. Several studies have shown that accurate knowledge about leprosy is associated with reduced stigma [40–42]. Misconceptions about leprosy, often linked to fear of the disease and fear of transmission, can increase stigma [2,43–45]. We therefore considered knowledge a tool to help family members address and challenge misconceptions and reduce (community) stigma. Knowledge can also reduce internalised stigma, by helping increase someone’s self-image. This was illustrated by Lusli and colleagues [46], who found that increased knowledge helped persons affected by leprosy to see themselves as cured and no longer infectious, rather than infectious and uncured. This enhanced their self-perception [46]. Human rights was identified as an important component to include in the intervention [19]. Persons affected by leprosy who are aware of their rights have been found to be more confident and less afraid to take initiatives [46].

Qualitative responses indicated that the social dimensions of the intervention were especially appreciated by the participants. Accordingly, there was a significant increase of scores in the domains “social relationships” and “environment” of the WHOQOL-BREF scale in both states post-intervention. Many studies have emphasized the importance of social support in fostering resilience [12,47–49]. Support for resilience is provided by family, friends, neighbours and mentors [47]. In children, peer acceptance and friendships have been found to act as moderators between family adversity and child adjustment to adversity [48]. Werner [47] found that self-esteem and self-efficacy are promoted through supportive relationships. Family relationships provide practical and emotional support in the context of discrimination [10,12]. In addition, people feel less stressed when they have good family and social support [10]. Good social relationships are vital for resilience [12] and we consider it a key component of the current intervention.

Family-based format

The present study used a family-based format. This was done for two main reasons. First, for many people, families are the bedrock of identity and wellbeing [50]. Family members and partners play a crucial role in resilience by supporting, believing in, and encouraging family members [12,49]. Family systems are said to mediate and regulate individual vulnerability and the impact of adverse events. Relationships with kin, intimate partners and mentors play a crucial role in resilience [12,48,49]. And second, stigma and discrimination have an impact on the whole family. In addition, a family-based intervention can be practiced at home and no travel is required, and is therefore relatively inexpensive (compared to e.g. peer support groups or counselling).

Impact of intervention on resilience

The intervention in the present study significantly improved resilience scores in participants from Odisha state post-intervention, but not in participants from Telangana state. In Odisha, this improvement was maintained at six-month follow-up. The baseline score of the participants in Odisha and Telangana state in the present study, were similar to that of elderly patients with depression in a study in Maharashtra state in India [51]. Follow-up scores in Odisha state were similar to that of healthy, elderly controls in the same study [51], but somewhat higher than the scores found in two cross-sectional studies in Karnataka state among parents of children with intellectual disability and adult offspring of parents with schizophrenia [52,53].

Several interventions aiming to improve resilience and wellbeing have found increases in CD-RISC scores, with most reporting an increase of around 15% [54]. The present study found no increase in resilience in Telangana state and 66% increase in Odisha state. This is a much higher increase than similar studies have reported [54], Indeed there have also been several studies who found no increase [55,56] or a decrease on the CD-RISC after intervention [57,58].

We surmised that this substantial discrepancy may in part be attributable to differences in context and community demographics [59,60]. Community ties, opportunities for participation in community life, opportunities to connect with others, as well as religious or other groups, can affect resilience and family functioning [61]. It is likely that community bonds were stronger in the rural areas of Odisha state as opposed to the Telangana participants, who were mostly extremely poor urban slum residents (almost half of the participants from Telangana state were unemployed). In addition, Hinduism in Telangana state was associated with more improvements in resilience and quality of life post-intervention. The controversial 'Citizenship Amendment Act' that was passed by the Parliament of India in December 2019, that offers citizenship to non-Muslims fleeing religious persecution from nearby countries, and related protests and demonstrations likely also had a negative impact on the Muslim participants of Telangana state. This is an area for future exploration. Importantly, the intervention also coincided with the COVID-19 pandemic and restrictions in Telangana (which were not as severe in Odisha). It is possible that the psychosocial nature of the intervention was not substantial enough to address their more fundamental and overwhelming needs. On the other hand, the Odisha participants were mostly tribal people, for whom such interventions were entirely novel. Further, the effects of the pandemic were less substantial and had not yet penetrated their region at the time of follow-up. It is possible that this was the first psycho-social intervention they had ever experienced and therefor drew considerable benefit. Finally, given the crucial role of social support in fostering resilience [12,47–49], it is possible that the participants from Odisha state had stronger social relationships and support prior to the intervention.

Impact of intervention on quality of life

The intervention in the present study significantly improved the quality of life of persons affected from both states, and of family members from Odisha state. In Odisha, where we conducted a second follow-up assessment, the improvement in quality of life remained six months after the intervention. This is an important finding, given that leprosy and leprosy-related stigma can have a negative impact on quality of life [36,62,63]. Studies have shown that resilience and quality of life are positively correlated [64,65]. Our results confirm the mediating role of resilience and social support on quality of life found in other studies [16,66,67].

The improvement in quality of life noted for people affected, but not for their family members, in Telangana suggests that the attention provided to them through the project may have had substantial benefit for them, but as noted above, was insufficient to improve the quality of life of their family members. It is self-evident that improving the resilience of families in extreme poverty requires more of a long-term, multifaceted and systemic intervention. This is an important lesson for future research.

Study limitations

As noted in the discussion, our study was substantially affected by the COVID-19 pandemic, which took effect in India while the intervention was underway. COVID-19 affected our data collection (limited follow-up in Telangana), and also appears to have adversely affected our outcome measure scores, but the exact nature and extent of that influence is a matter of conjecture. Suffice to say that we took confidence in the appeal of our intervention in that no families dropped out of the pilot study, and in the qualitative feedback, all described drawing benefit from the programme.

Conducting the project across multiple languages and cultures was also a substantial limitation that may have constrained comprehension at some points. In both cohorts there were a number of local dialects/languages, however all participants understood the language of intervention and outcome measurement (Urdu in Telangana, and Odia in Odisha). This limitation was to some extent mitigated by using local staff who were familiar with the families and the local dialects/languages. Further, while our own internal translation of the CD-RISC into Odia was certainly beneficial, it was not validated in that language, so results should be considered cautiously. Indeed the discrepancies of scores across cohorts may in part be attributable to language and translation concerns.

We recommend including pre- and post-intervention assessment of knowledge and community stigma in future studies, in order to ensure a better understanding of the changes in knowledge and community stigma. For future studies it would also be worthwhile to assess internalised stigma, mental wellbeing, physical health and stressor exposure also, to explore what role these factors play in strengthening resilience and interventions to strengthen resilience. The relationship between resilience and religion is another area for future exploration. As noted above, since this pilot study included relatively small numbers of participants across various religious groups, and coincided with religious persecution due to the ‘Citizenship Amendment Act’, interpretation of these data must be cautious. However, in response to these indications and current literature on this topic [11], there is great need for more detailed exploration of the interplay between resilience and religion, beliefs, spirituality and faith practices.

Conclusions

This pilot study showed that the 10-week family-based intervention to strengthen resilience among persons affected by leprosy and their family members is feasible, and has the potential to improve resilience and quality of life. This is one of the first interventions designed to strengthen psychosocial resilience of persons affected by NTDs such as leprosy in a developing country context. A large-scale efficacy trial is necessary to determine the effectiveness and long-term sustainability of the intervention.

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References

1. Lastória JC, Abreu MAMM de. Leprosy: review of the epidemiological, clinical, and etiopathogenic aspects-part 1. An Bras Dermatol 2014;89:205–18.
2. Sermrittirong S, Van Brakel W. Stigma in leprosy: concepts, causes and determinants. Lepr Rev 2014;85:36–47.
3. Bainson KA, Van den Borne B. Dimensions and process of stigmatization in leprosy. Lepr Rev 1998;69:341–50.
4. Rafferty J. Curing the stigma of leprosy. Lepr Rev 2005;76:119–26.
5. Somar PMW, Waltz MM, van Brakel WH. The impact of leprosy on the mental wellbeing of leprosy-affected persons and their family members–a systematic review. Glob Ment Heal 2020;7.
6. Cross H, Choudhary R. STEP: an intervention to address the issue of stigma related to leprosy in Southern Nepal. Lepr Rev 2005;76:316–24.
7. Shih M. Positive stigma: Examining resilience and empowerment in overcoming stigma. Ann Am Acad Pol Soc Sci 2004;591:175–85.
8. Earnshaw VA, Lang SM, Lippitt M, Jin H, Chaudoir SR. HIV Stigma and Physical Health Symptoms: Do Social Support, Adaptive Coping, and/or Identity Centrality Act as Resilience Resources? AIDS Behav 2015;19:41.
9. Kalisch R, Baker DG, Basten U, Boks MP, Bonanno GA, Brummelman E, et al. The resilience framework as a strategy to combat stress-related disorders. Nat Hum Behav 2017;1:784–90.
10. Windle G. What is resilience? A review and concept analysis. Rev Clin Gerontol 2011;21:152–69.
11. Sivilli TI, Pace TWW. The Human Dimensions of Resilience: A Theory of Contemplative Practices. Garrison Inst 2014.
12. Walsh F. Family resilience: A framework for clinical practice. Fam Process 2003;42:1–18.
13. Edward K, Warelow P. Resilience: When coping is emotionally intelligent. J Am Psychiatr Nurses Assoc 2005;11:101–2.
14. Earvolino-Ramirez M. Resilience: A Concept Analysis. Nurs. Forum, vol. 42, 2007, p. 73.
15. Fletcher D, Sarkar M. Psychological resilience: A review and critique of definitions, concepts, and theory. Eur Psychol 2013;18:12.
16. Rolland JS, Walsh F. Facilitating family resilience with childhood illness and disability. Curr Opin Pediatr 2006;18:527–38.
17. Wouters E. Life with HIV as a chronic illness: A theoretical and methodological framework for antiretroviral treatment studies in resource-limited settings. Soc Theory Heal 2012;10:368–91.
18. Saltzman WR. The FOCUS family resilience program: An innovative family intervention for trauma and loss. Fam Process 2016;55:647–59.
19. Van't Noordende AT, Kuipers P, Pereira ZBD. Strengthening personal and family resilience: a literature review for the leprosy context. Lepr Rev 2019;90:88–104.
20. Van't Noordende AT, Pereira Z, Kuipers P. Sources of strength and resilience for persons affected by leprosy in Brazil: What can we learn for service development? Accept Publ Int Heal 2020.
21. Bhana A, Bachoo S. The determinants of family resilience among families in low-and middle-income contexts: a systematic literature review. South African J Psychol 2011;41:131–9.

22. Hertzog MA. Considerations in determining sample size for pilot studies. Res Nurs Health 2008;31:180–91.
23. Connor KM, Davidson JRT. Development of a new resilience scale: The Connor-Davidson resilience scale (CD-RISC). Depress Anxiety 2003;18:76–82.
24. Murtaza G, Sultan S, Ahmed F, Mustafa G. Exploring construct validity of Resilience Scale in Pakistani youth. J Appl Env Biol Sci 2016;6:79–83.
25. Junnarkar M. Validation of Hindi Translated Scales on Grit, Resilience and Well-being 2018.
26. Camfield L, Skevington SM. On subjective well-being and quality of life. J Health Psychol 2008;13:764–75.
27. Organization WH. WHOQOL-BREF: introduction, administration, scoring and generic version of the assessment: field trial version, December 1996. World Health Organization; 1996.
28. Lodhi FS, Raza O, Montazeri A, Nedjat S, Yaseri M, Holakouie-Naieni K. Psychometric properties of the Urdu version of the World Health Organization’s quality of life questionnaire (WHOQOL-BREF). Med J Islam Repub Iran 2017;31:129.
29. Saxena S, Chandiramani K, Bhargava R. WHOQOL-Hindi: A questionnaire for assessing quality of life in health care settings in India. Natl Med J India 1998;11:160–5.
30. Kar N, Swain SP, Patra S, Kar B. The WHOQOL-BREF: Translation and validation of the odia version in a sample of patients with mental illness. Indian J Soc Psychiatry 2017;33:269.
31. Tsutsumi A, Izutsu T, Islam AM, Maksuda AN, Kato H, Wakai S. The quality of life, mental health, and perceived stigma of leprosy patients in Bangladesh. Soc Sci Med 2007;64:2443–53.
32. Rensen C, Bandyopadhyay S, Gopal PK, Van Brakel WH. Measuring leprosy-related stigma–a pilot study to validate a toolkit of instruments. Disabil Rehabil 2011;33:711–9.
33. Ibikunle PO, Nwokeji SC. Assessment of stigma among people living with Hansen’s disease in south-east Nigeria. Lepr Rev 2017;88:43–57.
34. Tsutsumi A, Izutsu T, Islam MDA, Amed JU, Nakahara S, Takagi F, et al. Depressive status of leprosy patients in Bangladesh: association with self-perception of stigma. Lepr Rev 2004;75:57–66.
35. Van Brakel WH, Sihombing B, Djarir H, Beise K, Kusumawardhani L, Yulihane R, et al. Disability in people affected by leprosy: the role of impairment, activity, social participation, stigma and discrimination. Glob Health Action 2012;5:18394.
36. van’t Noordende AT, Aycheh MW, Schippers A. The impact of leprosy, podoconiosis and lymphatic filariasis on family quality of life: A qualitative study in Northwest Ethiopia. PLoS Negl Trop Dis 2020;14:e0008173.
37. Earnshaw VA, Bogart LM, Dovidio JF, Williams DR. Stigma and racial/ethnic HIV disparities: moving toward resilience. 2015.
38. Zautra AJ, Hall JS, Murray KE, 1 the RSG. Resilience: a new integrative approach to health and mental health research. Health Psychol Rev 2008;2:41–64.
39. Chmitorz A, Kunzler A, Helmreich I, Tüscher O, Kalisch R, Kubiak T, et al. Intervention studies to foster resilience–A systematic review and proposal for a resilience framework in future intervention studies. Clin Psychol Rev 2018;59:78–100.
40. van’t Noordende AT, Korfage I, Lisam S, Arif MA, Kumar A, van Brakel WH. The role of perceptions and knowledge of leprosy in the elimination of leprosy: A baseline study in Fatehpur district, northern India. PLoS Negl Trop Dis 2019;13:e0007302.

41. Seshadri D, Khaitan BK, Khanna N, Sagar R. The tangled web: a study of knowledge and attitude towards leprosy from a tertiary care hospital in India. Indian J Lepr 2014:27–41.
42. Singh R, Singh B, Mahato S. Community knowledge, attitude, and perceived stigma of leprosy amongst community members living in Dhanusha and Parsa districts of Southern Central Nepal. PLoS Negl Trop Dis 2019;13:e0007075.
43. Sottie CA, Darkey J. Living with stigma: Voices from the Cured Lepers' village in Ghana. Soc Work Health Care 2019;58:151–65.
44. Hofstraat K, van Brakel WH. Social stigma towards neglected tropical diseases: a systematic review. Int Health 2016;8:i53–70.
45. de Stigter DH, Geus LD, Heynders ML. Leprosy: between acceptance and segregation. Community behaviour towards persons affected by leprosy in eastern Nepal. Lepr Rev 2000;71:492–8.
46. Lusli M, Peters R, van Brakel W, Zweekhorst M, Iancu S, Bunders J, et al. The impact of a rights-based counselling intervention to reduce stigma in people affected by leprosy in Indonesia. PLoS Negl Trop Dis 2016;10:e0005088.
47. Werner EE. Risk, resilience, and recovery: Perspectives from the Kauai Longitudinal Study. Dev Psychopathol 1993;5:503–15.
48. Criss MM, Pettit GS, Bates JE, Dodge KA, Lapp AL. Family adversity, positive peer relationships, and children's externalizing behavior: A longitudinal perspective on risk and resilience. Child Dev 2002;73:1220–37.
49. Walsh F. The concept of family resilience: Crisis and challenge. Fam Process 1996;35:261–81.
50. Zuna NI, Brown I, Brown RI. Family quality of life in intellectual and developmental disabilities: A support-based framework. Int Public Heal J 2014;6:161.
51. Singh S, Deshmukh P, Ungratwar A, Subramanyam AA, Kamath R. Does resilience affect illness perception and well-being in the elderly? J Geriatr Ment Heal 2014;1:94.
52. Herbert HS, Manjula M, Philip M. Resilience and factors contributing to resilience among the offsprings of parents with schizophrenia. Psychol Stud (Mysore) 2013;58:80–8.
53. Rajan AM, John R. Resilience and impact of children's intellectual disability on Indian parents. J Intellect Disabil 2017;21:315–24.
54. The Connor-Davidson Resilience Scale. Home. The Scale. User Guide Page 5. Table 11a. Changes in CD-RISC 25 Associated with Intervention or Treatment. 2020. http://www.connordavidson-resiliencescale.com/user-guide-5.php.
55. White B, Driver S, Warren AM. Resilience and indicators of adjustment during rehabilitation from a spinal cord injury. Rehabil Psychol 2010;55:23.
56. Naylor JC, Dolber TR, Strauss JL, Kilts JD, Strauman TJ, Bradford DW, et al. A pilot randomized controlled trial with paroxetine for subthreshold PTSD in Operation Enduring Freedom/ Operation Iraqi Freedom era veterans. Psychiatry Res 2013;206:318–20.
57. Guest R, Craig A, Nicholson Perry K, Tran Y, Ephraums C, Hales A, et al. Resilience following spinal cord injury: A prospective controlled study investigating the influence of the provision of group cognitive behavior therapy during inpatient rehabilitation. Rehabil Psychol 2015;60:311.
58. Rogers D. Does a brief workshop change clinical associate students' resilience? African J Prim Heal Care Fam Med 2016;8:1–4.

59. Bhattarai M, Maneewat K, Sae-Sia W. Psychosocial factors affecting resilience in Nepalese individuals with earthquake-related spinal cord injury: a cross-sectional study. BMC Psychiatry 2018;18:60.
60. Campbell-Sills L, Forde DR, Stein MB. Demographic and childhood environmental predictors of resilience in a community sample. J Psychiatr Res 2009;43:1007–12.
61. Blyth DA, Roehlkepartain EC. Healthy Communities, Healthy Youth: How Communities Contribute to Positive Youth Development. 1993.
62. Brouwers C, van Brakel W, Cornielje H. Quality of life, perceived stigma, activity and participation of people with leprosy-related disabilities in south-east Nepal. Disabil CBR Incl Dev 2011;22:16–34.
63. Lustosa AA, Nogueira LT, Pedrosa JI dos S, Teles JBM, Campelo V. The impact of leprosy on health-related quality of life. Rev Soc Bras Med Trop 2011;44:621–6.
64. Bodde MI, Schrier E, Krans HK, Geertzen JH, Dijkstra PU. Resilience in patients with amputation because of Complex Regional Pain Syndrome type I. Disabil Rehabil 2014;36:838–43.
65. Velickovic K, Rahm Hallberg I, Axelsson U, Borrebaeck CAK, Rydén L, Johnsson P, et al. Psychometric properties of the Connor-Davidson Resilience Scale (CD-RISC) in a non-clinical population in Sweden. Health Qual Life Outcomes 2020;18:1–10.
66. Migerode F, Maes B, Buysse A, Brondeel R. Quality of life in adolescents with a disability and their parents: The mediating role of social support and resilience. J Dev Phys Disabil 2012;24:487–503.
67. Alriksson-Schmidt AI, Wallander J, Biasini F. Quality of life and resilience in adolescents with a mobility disability. J Pediatr Psychol 2007;32:370–9.

CHAPTER 11

General discussion

In this thesis the perception of leprosy, interventions to change the perception of leprosy at community level, and interventions to reduce the impact of leprosy at individual and family level were explored. Four research questions were addressed in the articles presented in chapters 2 to 10. In this general discussion, the main findings of this thesis are described and interpreted, methodological considerations are discussed, and recommendations for policy, practice and future research are given.

Main findings

Research question 1: What are determinants of perception and knowledge of leprosy in endemic communities?

Answer:

We found that determinants of knowledge of leprosy in endemic districts in India and Indonesia include literacy, education, (not) knowing anyone affected by leprosy and living area. Living area was also a determinant of community attitudes towards persons affected by leprosy. Determinants of personal attitudes, represented by social distance towards persons affected by leprosy, include sex, education, knowledge about leprosy and living area.

There were differences in levels of knowledge and stigma between participant groups, for example, knowledge was better among health workers and stigma was high among community members (chapters 2 and 3). We found living area to be a determinant of perception, living area was a determinant of knowledge of leprosy, community attitudes, and personal attitudes. This likely reflects the differences in local beliefs and misconceptions about the cause and mode of transmission of leprosy between participants from India and Indonesia. For example, in Indonesia a common perception was that leprosy runs in the family for seven generations, in India a common perception was that leprosy is a result of karma. Indeed, while the origin of stigma varies between conditions [1] and between countries (chapter 3), the unfair treatment and social exclusion is the same for all health-related stigma [1].

There are understandable reasons for leprosy to be perceived negatively, given the examples of discrimination, the risk of leprosy reactions and disability, and the need for treatment with side effects. However, fortunately, leprosy is not highly contagious and can be treated and cured. After patients take their first dose of MDT, the disease is no longer contagious. Most impairments and disabilities can be prevented if treatment is taken in time, and chemoprophylaxis such as PEP++ can prevent the development of the disease in people who may carry the bacteria but are not aware of it yet. Early detection and prompt treatment are essential to interrupt the transmission of *M. leprae* and to reduce the number of new leprosy patients globally [2–4]. Early diagnosis and treatment are crucial to prevent impairments and disabilities [2] and play a role in stigma, because visible impairments are often linked to negative perceptions and stigma [5–9]. Early reporting of leprosy requires knowledge and awareness of leprosy in the community [10]. In addition, voluntary and early reporting of leprosy is hampered by stigma [4]. The disease is often concealed due to fear of potential discrimination [11]. Concealment of one's leprosy status is also a potential barrier to contact-based interventions such as post-exposure prophylaxis [12]. Improving knowledge of leprosy and reducing stigma is therefore essential to improve (strategies for) early case detection.

We identified three main drivers of negative attitudes towards (persons affected by) leprosy: (1) poor knowledge and misconceptions about leprosy, (2) local beliefs, and (3) fear of contagion. Our findings confirm results from other studies that also found that lacking knowledge about leprosy [13–17], (socio)cultural beliefs about leprosy [13,17–23] and fear of infection [19,20,24,25] are associated with negative attitudes towards persons affected by leprosy. These drivers should receive specific attention when designing interventions to change the perception of leprosy.

Although we identified three main drivers of negative attitudes, the determinants we included in our study explained only little of the variability of perception (knowledge of leprosy and personal and community attitudes). We included determinants that are generally included in knowledge, attitudes, and practices (KAP) studies, such as sex, age, education, occupation, marital status, religion, knowing a person affected by leprosy, living area and knowledge of leprosy. We did not include marital status or religion in our analysis due to missing data and contextual differences, respectively. We found a number of determinants of knowledge and attitudes, but these explained very little of the variability of knowledge (10-29%) and attitudes (3-10%; chapter 3). Studying the variability by district did not provide more insight into this variability. Our findings show that only including the 'obvious determinants' is not sufficient. Some studies have found additional determinants of attitudes towards persons affected by leprosy, such as regulations regarding leprosy [19], exposure to leprosy health promotion messages [18,20], marital status [15,22], economic status [26], ethnicity [13,15,27], distance of residence from the hospital [13] and migrant status [27]. We also found that exposure to leprosy health promotion messages and income were determinants of perception and knowledge of leprosy when we conducted a second perception study in India (chapter 7). In addition, some studies have suggested that perceptions are influenced by religious beliefs and religious teachings about leprosy [18–20,28]. In other studies, we also identified religion and spirituality as a source of strength to handle leprosy-related discrimination (chapter 9) and we identified the relationship between religion and resilience as an area for future exploration (chapter 10). We consider exploring the interplay between religion/spirituality, religiosity, resilience, and perception an important area for future research.

We conclude that it is necessary to include more personal and environmental factors of the participants in studies that explore determinants of perception and knowledge, to get a better understanding of the underlying mechanisms behind perception and knowledge, and ultimately to change these perceptions. In addition, it would be helpful to explore the influence of religion further, not just taking religious beliefs, but also level of religious faith and dedication into account.

Research question 2: How does leprosy impact marital and family life?

Answer:

We found that leprosy can influence marital and family life in several ways. Women in Nepal experienced problems in marital life during leprosy treatment, such as more distance from close others, couples sleeping in separate beds and having no sexual intercourse because the husband was afraid of contagion. In Ethiopia, we found that leprosy-related stigma and physical impairments can make it difficult for persons affected to find a spouse, can be a reason for divorce, and may cause reduced productivity and discrimination. Sometimes family members also experienced discrimination and difficulties in finding a spouse. In addition, reduced productivity of persons affected can impact the whole family, through loss of income, which may cause financial problems. Finally, having to provide for an affected family member can cause stress, school dropouts and an additional workload.

Difficulties in marital and family life are not unique to persons affected by leprosy. In our studies in Nepal and Ethiopia, we explored experiences of persons affected by leprosy, but also experiences of other marginalised groups, such as persons affected by podoconiosis and lymphatic filariasis, and women with disabilities. We found that these participants also experienced problems in their marital and family life. It is likely that the visibility of their symptoms and their marginalised position in the society and family (e.g., a lack of acceptance of their disease and a perceived lower social standing because of their disease) contributed to the problems they experienced. Appearance was one of the most frequently reported reasons for stigmatisation found in a review of stigma towards neglected tropical diseases (NTDs) [24] and is considered an important contributor to perceptions and stigma of leprosy (chapters 3 and 7 and [5–9]). Leprosy, podoconiosis, lymphatic filariasis and physical disabilities are often visible, which identifies these persons as different from others in the community. In addition, people with an “atypical” appearance are sometimes considered inferior [29]. This may have contributed to the experiences of our participants. It is known that some diseases are less stigmatised than others, for instance, persons with diabetes have been found to experience less stigma than persons with HIV, leprosy or schizophrenia and some NTDs like human African trypanosomiasis are less stigmatised than other NTDs [24,30]. People affected by some diseases may therefore experience less severe stigma. However, studies have also reported that if a condition is stigmatised, the impact of such stigma on people’s lives is similar across such stigmatised conditions [1,31,32].

In addition, we found that it was sometimes difficult to distinguish the problems due to the impact of leprosy on marital and family life from those that might have occurred otherwise (without leprosy). There were also factors that contributed to problems in marriage and family that were found in all participant groups (including a control group of ‘able bodied’ women in Nepal). This points to more structural problems in society. For example, the high frequency of alcohol abuse among the husbands of our participants in Nepal, the lower status of women compared to men in Nepal, and the lack of financial means of our participants in Ethiopia. However, it appeared that persons affected by leprosy faced problems in their marital relationship and family life that specifically related to leprosy. The women in Nepal faced additional problems during treatment, such as no intercourse due to fear of contagion, and social distance, while in Ethiopia, divorce seemed to occur more often among persons affected by leprosy than among persons affected by podoconiosis or lymphatic filariasis. However, it should be noted that our studies included only small samples of non-randomly selected participants. Therefore, a larger survey using random sampling will have to confirm whether the greater frequency of problems in marital and family life in these studies is real, or a result of selection bias.

We found that many of the problems persons affected by leprosy experienced in their marital and family life likely result from a lack of knowledge about leprosy, insufficient access to (sexual) health education and the negative perception of leprosy in the family and community. In our studies in Nepal (chapter 5) and Ethiopia (chapter 6), many participants had poor knowledge about the cause of leprosy. In our studies in India and Indonesia (chapters 2 and 3) we found that lower knowledge of leprosy was associated with more negative attitudes towards (persons affected by) leprosy. Indeed, the perception (misconception) of leprosy as a hereditary disease, or fear of costs of treatment and of disability can negatively influence marriage prospects and marital stability, not only for persons affected but also for their family members. Similarly, the belief (misconception) that leprosy is sexually transmitted or very infectious may cause the spouse or other family members to keep their distance. Such perceptions may also result in family members experiencing courtesy stigma (stigma by association), which can negatively impact employment and romantic relationships of family members [33–35]. Indeed, families are interconnected, problems experienced by individuals within the family can affect all family members [36,37]. Family members can also be a source of stigma [38,39]. Although this is not something we found in our study in Ethiopia, we did find that husbands were a source of discrimination in Nepal.

In conclusion, we found that (perception of) leprosy can negatively impact different aspects of marital and family life of persons affected and of their family members. It was sometimes difficult to distinguish the problems arising due to the impact of leprosy, from those that might have occurred regardless of people's leprosy status. Further research is recommended to investigate the extent of the problems in marital and family life and ways to ameliorate the situation of persons affected and their family members. Since some problems seemed to have been caused by misconceptions about leprosy and a lack of knowledge, education of leprosy and counselling of persons affected and their family members at diagnosis may help prevent some of the problems reported.

Research question 3: How effective are posters and community meetings in changing perception and knowledge of leprosy at community level?

Answer:

Contextualised posters and community meetings were highly effective in increasing knowledge and changing perceived community attitudes, and moderately effective in changing personal attitudes of community members and close contacts.

Stigma is a social process and while it mainly impacts the individual and their family, it originates from society's perceptions about persons with stigmatised conditions [40–42]. The origin of stigmatisation lies in the perception of persons affected by leprosy as 'different from the norm' [41]. It is therefore important to address the sources of stigma, i.e., to address public perceptions. We piloted two interventions: contextualised posters, and meetings with persons affected by leprosy, community members, persons affected by leprosy and influential people in the community (chapter 7). The primary aim of both interventions was to change perceptions (knowledge, attitudes, beliefs, emotions) regarding leprosy. We focused on changing the perception of leprosy, e.g., to make clear that leprosy can be prevented, disabilities can be prevented or managed, and that the prognosis of the disease is good. We also focused on changing the perception of persons affected by leprosy, e.g., indicating that persons affected by leprosy can lead a normal life, should not be discriminated against, and are just as valuable as anyone else. The secondary aim was to change behaviour. We expected that changing perceptions would change behavioural intention, which would ultimately lead to a change in behaviour also [43].

Since interventions are more likely to be successful if they are culture-specific and contextualised [44,45], we also contextualised our interventions. The content of the posters and meetings was based on the knowledge gaps, misconceptions, beliefs, and fears identified in chapters 3 and 4. Education – providing information about leprosy – was an important component of both interventions. Other aspects of the interventions were changing beliefs ('we can touch leprosy patients'), "normalisation" of persons affected by leprosy (stressing that they are no different from other community members), challenging fears (indicating that 'disabilities/ leprosy can be prevented' and that 'leprosy is curable') and providing calls to action ('immediately get [symptoms] checked at the health centre'). We found that the interventions were highly effective in increasing knowledge and changing perceived community attitudes, and moderately effective in changing personal attitudes of community members and close contacts (chapter 7). While we know that it is more difficult to change behaviour than to improve knowledge [46,47], we also know that changing perceptions, including improving knowledge, can lead to behaviour change [14,48,49]. We believe the effectiveness of the interventions could be further increased by combining them with other approaches such as facilitating contact between persons with and without leprosy [50–52].

Perceptions of leprosy are influenced by the media. The way people are portrayed in the media play a major role in defining and perpetuating public perceptions of those who are portrayed [53]. In other fields, such as cancer, HIV/AIDS and suicide prevention, the media has been found to both be a source of stigma as well as a medium to reduce stigma [54–56]. Views and attitudes of key influential people (or 'popular opinion leaders' or 'key players'), such as community leaders, religious leaders and health workers, can also influence how others in the community perceive leprosy [57]. Key influential people are "a minority of individuals who influence an exceptional number of their peers" [58], they are influential people in social networks, who are often perceived as trustworthy and are often at the centre of interactions [58,59]. Influential people play a vital role in shaping public opinion [58,59]. Targeting influential people and changing their perceptions, can influence public perceptions [58,59]. The information from the interventions can 'filter down' when influential people address leprosy at for example religious or community meetings [28]. Targeting influential people also increases the reach of interventions, given that these people are often at the centre of interactions [58,59]. We also included people working for the media (e.g., newspapers) and influential people, including teachers, heads of the village and religious leaders in the community meetings that were conducted (chapter 7). It would have been interesting if we could have reported on the impact of the community meetings on knowledge and perceptions of these influential people specifically, but they were not included as a separate target group in the evaluation of the interventions. The impact of interventions to improve perception on key influential people like heads of villages and religious leaders, and how their perceptions influence public perceptions, is an area for further research. In addition, while a few studies have explored the role of the media in the field of leprosy [28,60–62] and have found that social marketing campaigns on TV and radio can positively influence attitudes towards leprosy [61], the role the media can play in changing the perception of leprosy has not been studied sufficiently. Therefore, the role key influential people and the media can play in changing the perception of leprosy and reducing stigma is also an area for future research.

In conclusion, contextualised posters and community meetings are highly effective in increasing knowledge and changing perceived community attitudes, and moderately effective in changing personal attitudes. Addressing public perceptions is important, because stigma originates from public perceptions about persons who have stigmatised conditions. The role of key influential people in changing the perception of leprosy and reducing stigma is an area for future research.

Research question 4: What interventions have the potential to reduce the impact of leprosy at individual and family level?

Answer:

Interventions that focus on the entire family and that aim to strengthen resilience or social support and social connection, have the potential to reduce the impact of leprosy at individual and family level.

From other studies we know that there are several strategies and interventions that can mitigate the impact of stigma on persons affected by leprosy, including strengthening social support, psychological support and counselling, socio-economic empowerment, resilience and empowerment [49,63–67]. We conducted two proof of concept studies (chapters 8 and 10). One intervention was designed to strengthen individual and family resilience, the other to prevent and (self-)manage leprosy-related disabilities. Both interventions were based on exploratory studies (chapters 6 and 9), focused on the entire family, and were delivered over several monthly or weekly meetings. Key components of both interventions were strengthening (family) social support and connection, and improving self-perception.

We found that relationships with and social support from family members, friends and other persons affected by leprosy was a primary source of resilience for persons receiving services for leprosy in Porto Velho, Brazil (chapter 9). The importance of social support was also highlighted in chapter 6. Indeed, quality relationships and social support can help people regulate their emotions and cope with stress, can provide practical support, help solve problems, promote self-esteem and can foster resilience [63,68–71]. Family social support not only provides practical and emotional support in the context of discrimination [37,72], but can also enhance a sense of belonging or connectedness and group identification (feelings of inclusion that are threatened by stigma) for all family members. Feeling like you belong to a social group is a basic psychological need [71,73,74]. (Perceptions of) social isolation and loneliness have a major influence on mental wellbeing [71,75]. When faced with significant changes in life (such as being diagnosed with leprosy), wellbeing and adjustment are enhanced when people are able to maintain or acquire new social group memberships [76]. Family members can help meet this need, but this can also be met by friends, peers or religious or other groups. The importance of relationships with peers and the importance of religion and spirituality as sources of strength were identified in chapter 9 also. Other studies have also emphasised the importance of peer support and self-help groups; these groups have been found to improve social participation, restore dignity, and create a sense of belonging within the community [49,77–79]. Given that discrimination can have an impact on the whole family (chapters 5 and 6) and keeping in mind that families are interconnected, we believe that increasing (quality) social support and connection has the potential to reduce the impact of leprosy at individual and family level.

Another key component of the two interventions we piloted was that both improved the (self-) perception of persons affected by leprosy. Negative perceptions and stigma can threaten personal and social identity and self-esteem [40,80,81]. Persons affected by leprosy can for example be reduced from being a mother or a teacher to being ‘sick’ or a ‘leper’. Identity threat occurs when stressors that are harmful to one’s identity exceed an individual’s coping resources [81]. The interventions we piloted (chapters 8 and 10) also focused on strengthening coping resources: resilience, social support, and social connection. Several aspects of the interventions we piloted, such as socio-economic empowerment, management of disabilities, improving knowledge of leprosy and strengthening resilience, contributed to a positive self-perception and have likely also changed the way people in the community perceive persons affected by leprosy. Socio-economic empowerment (chapter 8) can change (self-)perception through increasing financial means, protecting against loss of social value, and promoting dignity, independence and social participation [52,65]. By restoring a person’s social life, socio-economic empowerment also improves the acceptability of persons affected by leprosy by their family and community members [82]. Disease management (chapter 8) also influences self- and disease perception, through minimising (the development of) visible impairments. Increasing knowledge of leprosy can help persons affected by leprosy to see themselves as cured and no longer infectious, rather than infectious and ill, and therefore change their self-perception (chapters 7, 8 and 10 and [49]). Finally, resilience reduces the impact of 'negative' perceptions and discrimination on persons affected, and alters someone's self-perception, through experiencing that one is able to handle and overcome difficulties.

In conclusion, strengthening family or group (social) support and connections, and improving self-perception through socio-economic empowerment, management of disabilities, knowledge of leprosy and strengthening resilience have the potential to reduce the impact of leprosy at individual and family level. It should be noted that while these interventions can the reduce the impact of leprosy at individual and family level, the interventions do not target the sources of stigma – public perceptions. As long as there is community stigma, efforts should be made to reduce the impact of stigma on persons affected also. Since we conducted pilot studies that included relatively small numbers of participants, the data should be interpreted with caution.

Experiences of stigma and the interaction of different social identities

People who experience leprosy-related stigma, often experience multiple overlapping inequalities in social relations, for example gender and socioeconomic status. These inequalities in social relations can exacerbate experiences of stigma. This is called 'intersectionality'. Intersectionality refers to the interaction of different social identities, that influence social relations [83–85]. Social identities such as 'race'/ethnicity, class, gender, sexuality, geography, ability, and age, are interrelated and shape one another [83–85]. Interactions between these different identities occur within systems and structures of power, such as laws, policies, governments, religious institutions and media [85]. This creates forms of privilege and oppression (inequalities) [85]. In the field of leprosy, (female) gender has been linked to more experiences of stigma. Multiple studies, including a systematic review, found women to be more affected by leprosy and its stigma than men [86–91]. This is reflected in our results from chapter 5 also, where women indicated that their feelings and desires were not valued, and that there was a difference between men and women when it comes to remaining faithful to their marriage. Another important adversity that intersects with and reinforces the experience of stigma is poverty. People who are poor often have a perceived lower social standing [92,93]. In addition, poverty can create barriers to accessing schooling/education, health care or social support [92,93]. This can in turn exacerbate the experience of marginalisation [93]. A study in Nepal, for example, found that the impact of stigma was less for persons affected by leprosy with a higher social status [94].

Intersectionality highlights the importance of understanding the social context of persons with stigmatised health conditions and emphasises the need for and importance of multi-level, multi-faceted, cross-cutting interventions. In India, there are many inequalities in social classes and interactions that can reinforce experiences of stigma. For example, people may be stigmatised or marginalised because of their caste or tribe, socio-economic class, religion, gender, sexual orientation, migrant status, because they have disabilities or certain diseases (e.g. leprosy), or because they are married women who are childless [95–100]. They deviate from what is considered ‘the ordinary’ [100]. These stigmas interact with and compound each other. Persons affected by leprosy may face multiple barriers because of their multiple identities [99]. At the same time these factors, such as social class or age, can mediate stigma [100]. Social structures and systems enforce discrimination and influence those who are most marginalised in society [99]. In India, the Government has mainly used policy changes to reduce stigmatisation and to protect people belonging to marginalised groups [95]. For example through the Scheduled Castes and The Scheduled Tribes Amendment Act 2018 [95], legislation to ensure equal rights for people with disabilities [98], a policy of reservation for Schedule Caste/Schedule Tribe in government jobs [95], and abolishment of the Criminal Tribe Act [97]. Many laws that discriminate against persons affected by leprosy have also been repealed, for example, the Leper’s Act [101], about segregation and institutionalisation of leprosy patient. It is unclear if these policy changes have resulted in changes in perceptions and behaviour towards persons affected by leprosy. It is important to take intersectionality into account, if we want to improve the lives of people who are marginalised, such as persons affected by leprosy may be.

Methodological issues

We used different study designs in the studies presented in this thesis. We conducted cross-sectional and pre/post intervention studies and used qualitative or mixed methods approaches. The studies described in chapters 7, 8 and 10 are pre/post intervention studies, the strengths and limitations of these studies are discussed below. In addition, in this section methodological considerations regarding intervention development and clinical relevance are discussed.

Pre/post intervention studies

We used pre/post intervention study designs with mixed methods for the development and evaluation of posters and community meetings to change perceptions of leprosy (chapter 7), a proof of concept family-based intervention for prevention and self-management of disabilities (chapter 8), and a proof of concept family-based intervention to strengthen resilience (chapter 10). We used two independent samples in chapter 7 and paired samples in chapters 8 and 10.

The proof of concepts studies (chapters 8 and 10) assessed the short-term effects of family-based interventions among a relatively small sample. Because of the non-random sample and relatively short follow-up time, effectiveness of the intervention could not be assessed. Another limitation is that we did not use a control group; this would have provided additional evidence. However, since we conducted proof of concept studies, we believe we have demonstrated the feasibility and potential of the family-based interventions. A strength of these studies is their mixed method approach that allowed for triangulation of the data. Further research using a randomised controlled design with a larger sample is needed to gather evidence of the effectiveness of the interventions. The strengths and limitations of the interventions described in chapter 7 are discussed below.

Intervention development and theoretical base

We believe perceptions and behaviour mutually influence each other (Figure 1) [102]. Behaviour theories generally assume that attitudes and beliefs influence behaviour, but often do not acknowledge that behaviour can also influence attitudes and beliefs. When designing interventions to change the perception of leprosy and to reduce leprosy-related stigma, it is important to take into account that perceptions and behaviour can mutually influence each other. For example, behaviour of persons affected by leprosy during “contact interventions” (e.g. giving a testimony) can change their (self-)perception, for example through increased self-confidence [103]. Changing someone’s experience with a person or group of people can also change his or her perceptions of this person or group of people [104]. This is again illustrated by the “contact interventions” that have been used to reduce negative attitudes and stigma, also in the field of leprosy. These events change people’s experience with persons affected by leprosy, which may result in changed perceptions [103]. Environmental factors, such as legislation or attitudes of others, can also influence perceptions – for example in the case of internalised stigma, in which people self-stigmatise themselves based on perceived discriminatory views of society [105].

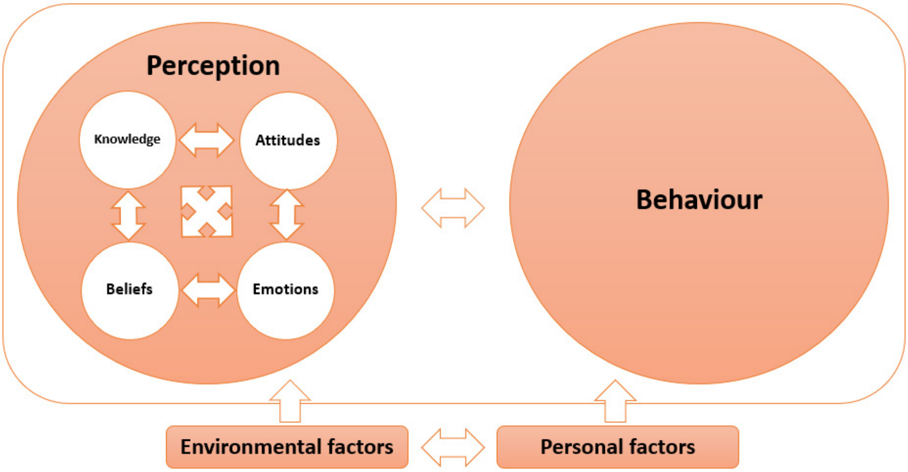

Figure 1. Components of perception and the relation between perception and behaviour.

Chapter 7 describes the development and evaluation of contextualised, relatively low-cost and easy to replicate interventions: posters and community meetings. We focused on addressing the drivers of negative attitudes we identified in chapter 3: poor knowledge of leprosy, (negative) local beliefs, and fear of contagion. We also prioritised knowledge gaps, such as cause, mode of transmission, symptoms and infectiousness of leprosy. The messages used 'clear and simple' language as much as possible, keeping in mind that the target group of our interventions had low literacy [106,107]. A strength is that the interventions were developed through collaborations and consultations between the target population, researchers, health workers, leprosy experts, communication experts and policymakers. This maximises the likelihood that the interventions fit with the target groups' needs and are acceptable, while promoting the uptake of the interventions by policymakers [108]. In addition, the posters were extensively pilot tested among the target groups. This resulted in an inventory of strategic locations to place the posters, and in changes in the language and images that were used. For example, a picture of Gandhi nursing a leprosy patient was replaced by another picture, because participants did not recognise Gandhi. We used the 6SQuID approach to develop the interventions [108].

Several approaches and frameworks exist to inform the development of behaviour change interventions. Well-known and commonly used examples include Intervention Mapping (IM) [109], PRECEDE-PROCEDE [110], the Behavior Change Wheel [111], the Medical Research Council (MRC) framework [112], the Theoretical Domains Framework (TDF) [113], the Knowledge-To-Action (KTA) process [114] and the 6SQuID approach [108]. While each framework has a slightly different focus and approach, the intervention development steps are similar. These steps broadly encompass: (1) analysing and understanding the problem, (2) defining the scientific core of the intervention, (3) designing and developing the intervention, (4) conducting an empirical optimisation, i.e. testing and refining the intervention, (5) designing the intervention evaluation, and (6) implementing and evaluating the intervention [115]. We used the 6SQuID approach for the development of the interventions in chapter 7, because it offers detailed and pragmatic guidelines on intervention development, with a population instead of an individual behaviour change focus [108].

The primary focus of our interventions was to change perceptions. For this reason, we did not use a behavioural change theory. Behaviour change approaches are especially relevant if behaviour needs changing on a large scale. The focus of our interventions was not primarily behaviour change, since only a small minority of people in a given community would have engaged in actual discriminatory behaviour. Regardless of its influence on behaviour, ‘perception of leprosy’ is a valid intervention target on its own. This is illustrated by a study in Bangladesh and West Bengal. In this study, being afraid of the negative perception and consequences of leprosy caused patients to delay seeking diagnosis and treatment, even though they had not experienced any discriminatory behaviour themselves, [116]. This illustrates the influence of perceptions alone, even when discriminatory behaviour is not experienced. However, while it was not the primary focus of our interventions, we anticipated that a change in perceptions would ultimately also change behaviour (Figure 1).

While we did not use behaviour (change) theories as a theoretical base in the development of our interventions in chapter 7, certain elements can be recognised in what we have done. Beliefs play an important role in theories that have been applied most often in the context of public health interventions, such as the Health Belief Model (HBM), the Theory of Planned Behaviour (TPB) and the Social Learning Theory (SLT) [117–120]. We focused our messages on improving knowledge and changing beliefs. In addition, from our understanding of the problem, we explored perceptions (knowledge, attitudes, beliefs, emotions) and noted the influence of perceptions on behaviour, and the role of personal and environmental factors on both perceptions and behaviour. This is reflected in the SLT that stresses that environmental, personal and behavioural elements all influence each other and determine behaviour change [121]. We also included elements of the HBM, for example, we stressed that leprosy is not highly contagious (perceived susceptibility), that disabilities can be prevented, that leprosy is curable and can be prevented (perceived severity and perceived benefits), and that medication is freely available (perceived barriers).

Therefore, although we did not use a behaviour (change) theory to develop the interventions, we did include some elements of such theories. In addition, theories are often used to understand behaviour [117,118] – we based the content of our interventions on a thorough understanding of local perceptions and aimed to challenge myths and fill knowledge gaps that were identified. While we described the perceptions, we aimed to change in detail (our primary objective), we defined the target behaviour quite broadly, e.g., 'more positive behaviour towards leprosy and persons affected by leprosy', 'no discrimination and exclusion'. Therefore, applying a behavioural change theory in our study might have resulted in clearer objectives for the target behaviour. However, a key challenge then is to assess changes in stigmatising behaviour. To our knowledge, there are no suitable measures to assess this. We would recommend that future studies explore meaningful ways to assess actual changes in behaviour and indicators of behaviour change, for example by asking persons affected by leprosy about their experiences at health facilities.

Clinical relevance

A challenge we encountered when evaluating the impact of the interventions in chapter 7, is that, while we found that the improvements in perception between Survey 1 and 2 were statistically significant, it was more challenging to assess whether the differences were also clinically relevant[1]. A way to determine whether changes or impact are interpreted as clinically relevant, is by using a cut-off point to classify respondents into groups that have an intuitive meaning to practitioners, such as 'negative attitude' and 'no negative attitude', or 'poor knowledge' and 'adequate knowledge'. Looking at the changes in proportion of respondents in each group pre and post intervention can help practitioners to interpret the findings and to estimate the magnitude of (meaningful) change or effect.

A novel feature of our study in chapter 7 is that we determined and used a cut-off point for negative personal attitudes on the SDS, which was not yet available. A cut-off point was already available for the EMIC-CSS [122]. However, while it is very useful, determining a cut-off point is also arbitrary. Itis a subjective point determined by the researcher. When developing a cut-off for classifying participants into those with a positive and those with a negative attitude based on the SDS score, we considered that the response scale items 'probably unwilling' and 'definitely unwilling' represent either fear or a negative attitude towards the person with the condition under study. Scoring several questions as 'probably unwilling' would indicate a negative attitude. We set the cut-off at '3 or more questions answered with 'probably unwilling (out of 7 in total)'. In addition, if

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[1] While the term that is often used is 'clinical relevance', this term does not only refer to the relevance or significance to clinical practice but can also refer to social or psychological relevance.

someone ‘scores’ a ‘definitely unwilling’ even only once, this already is a strong indication of fear or negative attitude. Therefore, when combined with a least one ‘probably unwilling’, we operationalised this as an overall ‘negative attitude’ classification. Our qualitative data also showed reflections of participants, indicating and endorsing changes in perceptions and behaviour.

Another way of determining clinical relevance is by calculating, for example, the minimal (clinically) important difference, using distribution or (ideally) anchor-based methods [123,124]. Anchor-based methods use an external criterion to operationalise minimal (clinically) important change, for example, participants self-reported global rating of perceived change [123]. It should be noted that this is also arbitrary: it depends on “the type of anchor, the definition of 'minimal importance' on the anchor, and on the baseline score which might be an indicator of severity of the disease” [123]. When it comes to stigma, it is very difficult if not impossible to use an anchor-based approach to determine which change should be considered sufficiently important (“better”) by persons affected. In the absence of an anchor-based method, we believe using a cut-off point (value) is a good way to express clinical relevance. This also allows for comparison across countries and conditions. Even though cut-off points are one of the best available methods in social science research to express clinical relevance and can help practitioners to interpret their findings, it should be noted that this remains an arbitrary method. There are no reference values for ‘normal’ and no gold standard for judging the relevance of a given measured change or difference in social science research, in the same way as normal cut-offs exist for e.g., temperature in the case of fever. Future research should therefore further explore ways to improve assessment of the clinical relevance of the impact of interventions.

Recommendations for policy, practice, and future research

Based on the results of the studies described in this thesis, we developed a set of recommendations for policy and practice:

- We found that psychological support in the early phases of diagnosis and treatment was a priority for persons affected by leprosy. We also found that leprosy can negatively influence marital and family life in several ways, some problems seemed to have been caused by misconceptions about leprosy and a lack of knowledge. We therefore recommend counselling and education of persons affected by leprosy *and* their family members at diagnosis, to help prevent some of the problems reported.
- This thesis has highlighted the importance of strengthening social support

and connections. Social support and connections can be strengthened at the family level, but friends, peers or religious or other groups can also play an important role. We recommend that support groups are established to reduce the impact of leprosy at individual and family level.

- We found that contextualised posters and community meetings were effective in changing community and personal attitudes of community members and close contacts towards leprosy. The effectiveness of these interventions in changing attitudes could likely be further improved by combining these interventions with other approaches, such as facilitating contact between persons with and without leprosy. We recommend that programme managers use multi-level and multi-faceted interventions to improve perceptions of leprosy.
- We strongly recommend working closely with the target population and local stakeholders, when developing interventions. This maximises the likelihood that the interventions fit with the target groups' needs and are acceptable, while promoting the uptake of the interventions by policymakers.

In addition, the results reported in this thesis identified the need for more research. Recommendations for further research include:

- To explore additional determinants of perceptions and knowledge, to get a better understanding of the underlying mechanisms behind perceptions and knowledge, and ultimately to change these perceptions.
- To explore how religion and religiosity influence resilience and perceptions of leprosy, and what role religiosity could play in strengthening resilience and changing perceptions.
- To explore what role key influential people, like heads of the village, religious leaders, and media personnel, can play in changing the perception of leprosy and in reducing stigma.
- To investigate the extent of the problems in marital and family life of persons affected by leprosy and their family members, using larger, random samples and control groups, and ways to ameliorate the situation.
- To further explore ways to improve assessment of the clinical relevance1 of the impact of interventions.
- To explore meaningful ways to measure changes in behaviour and indicators of behaviour change.

Conclusions

In this thesis we studied the perception of leprosy and interventions to change the perception of leprosy, and we explored interventions to reduce the impact of leprosy at individual and family level.

We found that leprosy can influence marital and family life in several ways. It was sometimes difficult to distinguish the problems arising due to the impact of leprosy, from those that might have occurred regardless of people's leprosy status. We found that many of the problems persons affected by leprosy experienced in their marital and family life likely result from a lack of knowledge about leprosy, insufficient access to (sexual) health education and negative perceptions of leprosy in the family and community. We showed that interventions that focus on the entire family and that aim to strengthen resilience or social support and social connection, have the potential to reduce the impact of leprosy at individual and family level. While these interventions can the reduce the impact of leprosy at individual and family level, they do not target the sources of stigma (public perceptions).

We developed two interventions to change public perceptions of leprosy, based on the knowledge gaps, misconceptions, beliefs and fears we identified. The two interventions we piloted (contextualised posters and community meetings) were effective in increasing knowledge and changing personal and perceived community attitudes. Addressing public perceptions is important, because stigma originates from public perceptions about persons who have stigmatised conditions. We found that knowledge of leprosy, education, knowing someone who is affected by leprosy and living area are determinants of perceptions of leprosy – but they can only explain little of the variability of perception. We identified three main drivers of negative attitudes: (1) poor knowledge and misconceptions about leprosy, (2) local beliefs about leprosy, and (3) fear of contagion. Interventions to change the perception of leprosy should target these drivers taking care to adapt these carefully to the local context.

References

1. Rao PS. Study on differences and similarities in the concept and origin of leprosy stigma in relation to other health-related stigma. Indian J Lepr 2010;82:117–21.
2. World Health Organization. WHO Expert Committee on leprosy: eighth report. Geneva: World Health Organization; 2012.
3. Duthie MS, Truman RW, Goto W, O'Donnell J, Hay MN, Spencer JS, et al. Insight toward early diagnosis of leprosy through analysis of the developing antibody responses of Mycobacterium leprae-infected armadillos. Clin Vaccine Immunol 2011;18:254–9.
4. Suzuki K, Akama T, Kawashima A, Yoshihara A, Yotsu RR, Ishii N. Current status of leprosy: epidemiology, basic science and clinical perspectives. J Dermatol 2012;39:121–9.
5. Van Brakel WH, Sihombing B, Djarir H, Beise K, Kusumawardhani L, Yulihane R, et al. Disability in people affected by leprosy: the role of impairment, activity, social participation, stigma and discrimination. Glob Health Action 2012;5:18394.
6. Tora A, Franklin H, Deribe K, Reda AA, Davey G. Extent of podoconiosis-related stigma in Wolaita Zone, Southern Ethiopia: a cross-sectional study. Springerplus 2014;3:647.
7. Zeldenryk LM, Gray M, Speare R, Gordon S, Melrose W. The Emerging Story of Disability Associated with Lymphatic Filariasis: A Critical Review 2011.
8. Navon L. Beggars, metaphors, and stigma: a missing link in the social history of leprosy. Soc Hist Med 1998;11:89–105.
9. Adhikari B, Kaehler N, Raut S, Marahatta SB, Ggyanwali K. Risk factors of stigma related to leprosy-A systematic review. J Manmohan Meml Inst Heal Sci 2013;1:3–11.
10. John AS, Rao OS. Awareness and attitudes towards leprosy in urban slums of Kolkata, India. Indian J Lepr 2009;81:135–40.
11. Marahatta SB, Amatya R, Adhikari S, Giri D, Lama S, Kaehler N, et al. Perceived stigma of leprosy among community members and health care providers in Lalitpur district of Nepal: A qualitative study. PLoS One 2018;13:e0209676.
12. Peters R, Mieras L, Subedi M, Apte H, Koesbardiati T, Banstola NL, et al. A single dose of rifampicin to prevent leprosy: qualitative analysis of perceptions of persons affected, contacts, community members and health professionals towards chemoprophylaxis and the impact on their attitudes in India, Nepal and Indonesia. Lepr Rev 2018;89:335–52.
13. Adhikari B, Shrestha K, Kaehler N, Raut S, Chapman SR. Community attitudes towards leprosy affected persons in Pokhara municipality of western Nepal. J Nepal Health Res Counc 2014.
14. van den Broek J, O'Donoghue J, Ishengoma A, Masao H, Mbega M. Evaluation of a sustained 7-year health education campaign on leprosy in Rufiji District, Tanzania. Lepr Rev 1998;69:57–74.
15. Singh R, Singh B, Mahato S. Community knowledge, attitude, and perceived stigma of leprosy amongst community members living in Dhanusha and Parsa districts of Southern Central Nepal. PLoS Negl Trop Dis 2019;13:e0007075.
16. Seshadri D, Khaitan BK, Khanna N, Sagar R. The tangled web: a study of knowledge and attitude towards leprosy from a tertiary care hospital in India. Indian J Lepr 2014:27–41.
17. van't Noordende AT, Korfage I, Lisam S, Arif MA, Kumar A, van Brakel WH. The role of perceptions and knowledge of leprosy in the elimination of leprosy: A baseline study in Fatehpur district, northern India. PLoS Negl Trop Dis 2019;13:e0007302.

18. Ebenso B, Newell J, Emmel N, Adeyemi G, Ola B. Changing stigmatisation of leprosy: an exploratory, qualitative life course study in Western Nigeria. BMJ Glob Heal 2019;4:e001250.
19. Sottie CA, Darkey J. Living with stigma: Voices from the Cured Lepers' village in Ghana. Soc Work Health Care 2019;58:151–65.
20. Sermrittirong S, Van Brakel W. Stigma in leprosy: concepts, causes and determinants. Lepr Rev 2014;85:36–47.
21. Tabah EN, Nsagha DS, Bissek A-CZ-K, Njamnshi TN, Njih IN-N, Pluschke G, et al. Community knowledge, perceptions and attitudes regarding leprosy in rural Cameroon: The case of Ekondotiti and Mbonge health districts in the South-west Region. PLoS Negl Trop Dis 2018;12:e0006233.
22. Ibikunle PO, Nwokeji SC. Assessment of stigma among people living with Hansen's disease in south-east Nigeria. Lepr Rev 2017;88:43–57.
23. Tekle-Haimanot R, Forsgren L, Gebre-Mariam A, Abebe M, Holmgren G, Heijbel J, et al. Attitudes of rural people in central Ethiopia towards leprosy and a brief comparison with observations on epilepsy. Lepr Rev 1992;63:157–68.
24. Hofstraat K, van Brakel WH. Social stigma towards neglected tropical diseases: a systematic review. Int Health 2016;8:i53–70.
25. de Stigter DH, Geus LD, Heynders ML. Leprosy: between acceptance and segregation. Community behaviour towards persons affected by leprosy in eastern Nepal. Lepr Rev 2000;71:492–8.
26. Raju MS, Reddy J V. Community attitude to divorce in leprosy. Indian J Lepr 1995;67:389–403.
27. Faatoese A, Sikaleti S, Priest P, Chambers S. Knowledge and attitudes to leprosy of Pacific People living in New Zealand. Lepr Rev 2016;87:368–77.
28. Brown W. Can social marketing approaches change community attitudes towards leprosy? Lepr Rev 2006;77:89–98.
29. Linton S. Claiming Disability: Knowledge and Identity. Claiming Disabil., New York University Press; 1998.
30. Rai SS, Irwanto I, Peters RM, Syurina E V, Putri AI, Mikhakhanova A, et al. Qualitative Exploration of Experiences and Consequences of Health-related Stigma among Indonesians with HIV, Leprosy, Schizophrenia and Diabetes. Kesmas Natl Public Heal J 2020;15:7–16.
31. Van Brakel WH. Measuring health-related stigma—a literature review. Psychol Health Med 2006;11:307–34.
32. Van Brakel WH, Cataldo J, Grover S, Kohrt BA, Nyblade L, Stockton M, et al. Out of the silos: identifying cross-cutting features of health-related stigma to advance measurement and intervention. BMC Med 2019;17:13.
33. Somar PMW, Waltz MM, van Brakel WH. The impact of leprosy on the mental wellbeing of leprosy-affected persons and their family members–a systematic review. Glob Ment Heal 2020;7.
34. Goffman E. Stigma and social identity 1963.
35. Dako-Gyeke M. Courtesy stigma: A concealed consternation among caregivers of people affected by leprosy. Soc Sci Med 2018;196:190–6.
36. Broderick CB. Understanding family process: Basics of family systems theory. Sage; 1993.
37. Walsh F. Family resilience: A framework for clinical practice. Fam Process 2003;42:1–18.

38. Dako-Gyeke M, Asampong E, Oduro R. Stigmatisation and discrimination: Experiences of people affected by leprosy in Southern Ghana. Lepr Rev 2017;88:58–74.
39. Roosta N, Black DS, Rea TH. A comparison of stigma among patients with leprosy in rural Tanzania and urban United States: a role for public health in dermatology. Int J Dermatol 2013;52:432–40.
40. Goffman E. Stigma: Notes on the management of spoiled identity. Simon and Schuster; 2009.
41. Weiss MG, Ramakrishna J, Somma D. Health-related stigma: rethinking concepts and interventions. Psychol Health Med 2006;11:277–87.
42. Link BG, Phelan JC. Conceptualizing stigma. Annu Rev Sociol 2001;27:363–85.
43. Fishbein M, Ajzen I. Belief, attitude, intention, and behavior: An introduction to theory and research. Addison-Wesley: Reading, MA, USA; 1975.
44. Opala J, Boillot F. Leprosy among the Limba: illness and healing in the context of world view. Soc Sci Med 1996;42:3–19.
45. Chen PC, Sim HC. The development of culture-specific health education packages to increase case-finding of leprosy in Sarawak. Southeast Asian J Trop Med Public Health 1986;17:427–32.
46. Bay JL, Mora HA, Sloboda DM, Morton SM, Vickers MH, Gluckman PD. Adolescent understanding of DOHaD concepts: a school-based intervention to support knowledge translation and behaviour change. J Dev Orig Health Dis 2012;3:469–82.
47. Barr RG, Barr M, Fujiwara T, Conway J, Catherine N, Brant R. Do educational materials change knowledge and behaviour about crying and shaken baby syndrome? A randomized controlled trial. Cmaj 2009;180:727–33.
48. Arole S, Premkumar R, Arole R, Maury M, Saunderson P. Social Stigma: A Comparative Qualitative Study of Integrated and Vertical Care Approaches to Leprosy. Lepr Rev 2002;73:186–96.
49. Lusli M, Peters R, van Brakel W, Zweekhorst M, Iancu S, Bunders J, et al. The impact of a rights-based counselling intervention to reduce stigma in people affected by leprosy in Indonesia. PLoS Negl Trop Dis 2016;10:e0005088.
50. Brown L, Macintyre K, Trujillo L. Interventions to reduce HIV/AIDS stigma: what have we learned? AIDS Educ Prev 2003;15:49.
51. Couture S, Penn D. Interpersonal contact and the stigma of mental illness: A review of the literature. J Ment Heal 2003;12:291–305.
52. Dadun D, Van Brakel WH, Peters RMH, Lusli M, Zweekhorst M, Bunders JGF, et al. Impact of socio-economic development, contact and peer counselling on stigma against persons affected by leprosy in Cirebon, Indonesia–a randomised controlled trial. Lepr Rev 2017;88:2–22.
53. Susman J. Disability, stigma and deviance. Soc Sci Med 1994;38:15–22.
54. Chapple A, Ziebland S, McPherson A. Stigma, shame, and blame experienced by patients with lung cancer: qualitative study. Bmj 2004;328:1470.
55. Bekalu MA, Eggermont S, Ramanadhan S, Viswanath K. Effect of media use on HIV-related stigma in sub-Saharan Africa: A Cross-Sectional Study. PLoS One 2014;9:e100467.
56. Niederkrotenthaler T, Reidenberg DJ, Till B, Gould MS. Increasing help-seeking and referrals for individuals at risk for suicide by decreasing stigma: the role of mass media. Am J Prev Med 2014;47:S235–43.
57. Rafferty J. Curing the stigma of leprosy. Lepr Rev 2005;76:119–26.

58. Watts DJ, Dodds PS. Influentials, networks, and public opinion formation. J Consum Res 2007;34:441–58.
59. Momtaz NJ, Aghaie A, Alizadeh S. Identifying opinion leaders for marketing by analyzing online social networks. Int J Virtual Communities Soc Netw 2011;3:19–34.
60. White C. Sociocultural considerations in the treatment of leprosy in Rio de Janeiro, Brazil. Lepr Rev 2002;73:356–65.
61. Williams PG, Dewapura D, Gunawardene P, Settinayake S. Social marketing to eliminate leprosy in Sri Lanka. Soc Mar Q 1998;4:27–31.
62. White C. Déjà vu: leprosy and immigration discourse in the twenty-first century United States. Lepr Rev 2010;81:17.
63. Cohen S. Psychosocial models of the role of social support in the etiology of physical disease. Heal Psychol 1988;7:269.
64. Enache R. Psychological counseling and developing resilience for young people with psychological trauma in childhood or adolescence. J Exp Psychother 2016;19:73.
65. Ebenso B, Fashona A, Ayuba M, Idah M, Adeyemi G, S-Fada S. Impact of socio-economic rehabilitation on leprosy stigma in Northern Nigeria: findings of a retrospective study. Asia Pacific Disabil Rehabil J 2007;18:98–119.
66. Shih M. Positive stigma: Examining resilience and empowerment in overcoming stigma. Ann Am Acad Pol Soc Sci 2004;591:175–85.
67. Corrigan PW, Rao D. On the self-stigma of mental illness: Stages, disclosure, and strategies for change. Can J Psychiatry 2012;57:464–9.
68. Earvolino-Ramirez M. Resilience: A Concept Analysis. Nurs. Forum, vol. 42, 2007, p. 73.
69. Gurung R, Sarason B, Sarason I. Close personal relationships and health outcomes: A key to the role of social support. Handb Pers Relationships Theory, Res Interv (2nd Ed) Chichester, UK Wiley 1997:547–73.
70. Werner EE. Risk, resilience, and recovery: Perspectives from the Kauai Longitudinal Study. Dev Psychopathol 1993;5:503–15.
71. Heinrich LM, Gullone E. The clinical significance of loneliness: A literature review. Clin Psychol Rev 2006;26:695–718.
72. Windle G. What is resilience? A review and concept analysis. Rev Clin Gerontol 2011;21:152–69.
73. Baumeister RF, Leary MR. The need to belong: desire for interpersonal attachments as a fundamental human motivation. Psychol Bull 1995;117:497.
74. Maslow AH. A theory of human motivation. Psychol Rev 1943;50:370.
75. Hawkley LC, Cacioppo JT. Loneliness matters: A theoretical and empirical review of consequences and mechanisms. Ann Behav Med 2010;40:218–27.
76. Haslam C, Haslam SA, Jetten J, Cruwys T, Steffens NK. Life Change, Social Identity, and Health. Annu Rev Psychol n.d.
77. Benbow C, Tamiru T. The experience of self-care groups with people affected by leprosy: ALERT, Ethiopia. Lepr Rev 2001;72:311–21.
78. Deepak S, Hansine PE, Braccini C. Self-care groups of leprosy-affected people in Mozambique. Lepr Rev 2013;84:283.

79. Ebenso J, Muyiwa LT, Ebenso BE. Self care groups and Ulcer prevention in Okegbala Nigeria. Lepr Rev 2009;80:187.
80. Scambler G. Health-related stigma. Sociol Health Illn 2009;31:441–55.
81. Major B, O'brien LT. The social psychology of stigma. Annu Rev Psychol 2005;56:393–421.
82. Rao VP, Rao IR, Palande DD. Socio-economic rehabilitation programmes of LEPRA India-methodology, results and application of needs based socio-economic evaluation. Lepr Rev 2000;71:466–71.
83. Collins PH, Bilge S. Intersectionality. John Wiley & Sons; 2020.
84. Nash JC. Re-thinking intersectionality. Fem Rev 2008;89:1–15.
85. Hankivsky O. Intersectionality 101. Inst Intersect Res Policy, SFU 2014:1–34.
86. Mankar MJ, Joshi SM, Velankar DH, Mhatre RK, Nalgundwar AN. A comparative study of the quality of life, knowledge, attitude and belief about leprosy disease among leprosy patients and community members in Shantivan Leprosy Rehabilitation centre, Nere, Maharashtra, India. J Glob Infect Dis 2011;3:378.
87. Singh GP. Psychosocial aspects of Hansen's disease (leprosy). Indian Dermatol Online J 2012;3:166.
88. Try L. Gendered experiences: marriage and the stigma of leprosy. Asia Pacific Disabil Rehabil J 2006;17:45–6.
89. Varkevisser CM, Lever P, Alubo O, Burathoki K, Idawani C, Moreira TMA, et al. Gender and leprosy: case studies in Indonesia, Nigeria, Nepal and Brazil. Lepr Rev 2009;80:65.
90. Vlassoff C, Khot S, Rao S. Double jeopardy: women and leprosy in India 1996.
91. Dijkstra JI, van Brakel WH, van Elteren M. Gender and leprosy-related stigma in endemic areas: A systematic review. Lepr Rev 2017;88:419–40.
92. Rai SS, Peters RMH, Syurina E V, Irwanto I, Naniche D, Zweekhorst MBM. Intersectionality and health-related stigma: insights from experiences of people living with stigmatized health conditions in Indonesia. Int J Equity Health 2020;19:1–15.
93. Jackson-Best F, Edwards N. Stigma and intersectionality: a systematic review of systematic reviews across HIV/AIDS, mental illness, and physical disability. BMC Public Health 2018;18:1–19.
94. Heijnders ML. The dynamics of stigma in leprosy. Int J Lepr 2004;72:437–47.
95. Bhanot D, Verma SK. Lived Experiences of the Indian Stigmatized Group in Reference to Socio-Political Empowerment: A Phenomenological Approach. Qual Rep 2020;25.
96. Adamson PC, Krupp K, Niranjankumar B, Freeman AH, Khan M, Madhivanan P. Are marginalized women being left behind? A population-based study of institutional deliveries in Karnataka, India. BMC Public Health 2012;12:1–6.
97. Gavhale SR. Social Stigma and Struggle for Life of Marginal Group in Modern India. Int J Res Soc Sci 2016;6:211–22.
98. Ghai A. Disabled women: An excluded agenda of Indian feminism. Hypatia 2002;17:49–66.
99. Khanday ZA, Akram M. Health status of marginalized groups in India. Int J Appl Sociol 2012;2:60–70.
100. Riessman CK. Stigma and everyday resistance practices: Childless women in South India. Gend Soc 2000;14:111–35.

101. World Health Organization. Report of the informal consultation on stopping discrimination and promotion inclusion of persons affected by Leprosy, New Delhi, 14-16 Nov 2017. World Health Organization. Regional Office for South-East Asia; 2018.
102. Wells GL, Petty RE. The effects of over head movements on persuasion: Compatibility and incompatibility of responses. Basic Appl Soc Psych 1980;1:219–30.
103. Peters RMH, Zweekhorst MBM, Bunders JFG, van Brakel WH. A cluster-randomized controlled intervention study to assess the effect of a contact intervention in reducing leprosy-related stigma in Indonesia. PLoS Negl Trop Dis 2015;9:e0004003.
104. Reibstein DJ, Lovelock CH, Dobson R de P. The direction of causality between perceptions, affect, and behavior: An application to travel behavior. J Consum Res 1980;6:370–6.
105. Weiss MG. Stigma and the social burden of neglected tropical diseases. PLoS Negl Trop Dis 2008;2:e237.
106. Seligman HK, Wallace AS, DeWalt DA, Schillinger D, Arnold CL, Shilliday BB, et al. Facilitating behavior change with low-literacy patient education materials. Am J Health Behav 2007;31:S69–78.
107. Jensen JD. Addressing health literacy in the design of health messages. Heal Commun Messag Des Theory Pract 2012:171–90.
108. Wight D, Wimbush E, Jepson R, Doi L. Six steps in quality intervention development (6SQuID). J Epidemiol Community Heal 2016;70:520–5.
109. Bartholomew LK, Parcel GS, Kok G, Gottlieb NH, Schaalma HC, Markham CC, et al. Planning health promotion programs: an intervention mapping approach. Jossey-Bass; 2006.
110. Crosby R, Noar SM. What is a planning model? An introduction to PRECEDE-PROCEED. J Public Health Dent 2011;71:S7–15.
111. Michie S, Van Stralen MM, West R. The behaviour change wheel: a new method for characterising and designing behaviour change interventions. Implement Sci 2011;6:1–12.
112. Craig P, Dieppe P, Macintyre S, Michie S, Nazareth I, Petticrew M. Developing and evaluating complex interventions: the new Medical Research Council guidance. Bmj 2008;337.
113. French SD, Green SE, O’Connor DA, McKenzie JE, Francis JJ, Michie S, et al. Developing theory-informed behaviour change interventions to implement evidence into practice: a systematic approach using the Theoretical Domains Framework. Implement Sci 2012;7:1–8.
114. Graham ID, Logan J, Harrison MB, Straus SE, Tetroe J, Caswell W, et al. Lost in knowledge translation: time for a map? J Contin Educ Health Prof 2006;26:13–24.
115. Araújo-Soares V, Hankonen N, Presseau J, Rodrigues A, Sniehotta FF. Developing behavior change interventions for self-management in chronic illness. Eur Psychol 2018.
116. Nicholls PG, Chhina N, Bro AK, Barkataki P, Kumar R, Withington SG, et al. Factors contributing to delay in diagnosis and start of treatment of leprosy: Analysis of help-seeking narratives in northern Bangladesh and in West Bengal, India. Lepr Rev 2005;76:35–47.
117. Weston D, Ip A, Amlôt R. Examining the application of behaviour change theories in the context of infectious disease outbreaks and emergency response: a review of reviews. BMC Public Health 2020;20:1–19.
118. Angus K, Cairns G, Purves R, Bryce S, MacDonald L, Gordon R. Systematic literature review to examine the evidence for the effectiveness of interventions that use theories and models of behaviour change: towards the prevention and control of communicable diseases. European Centre for Disease Prevention and Control; 2013.

119. McGlashan AJ, Finch CF. The extent to which behavioural and social sciences theories and models are used in sport injury prevention research. Sport Med 2010;40:841–58.
120. Davis R, Campbell R, Hildon Z, Hobbs L, Michie S. Theories of behaviour and behaviour change across the social and behavioural sciences: a scoping review. Health Psychol Rev 2015;9:323–44.
121. Bandura A, McClelland DC. Social learning theory. vol. 1. Englewood cliffs Prentice Hall; 1977.
122. Sermrittirong S, Van Brakel WH, Kraipui N, Traithip S, Bunders-Aelen JF. Comparing the perception of community members towards leprosy and tuberculosis stigmatization. Lepr Rev 2015;86:54–61.
123. de Vet HC, Terwee CB, Ostelo RW, Beckerman H, Knol DL, Bouter LM. Minimal changes in health status questionnaires: distinction between minimally detectable change and minimally important change. Health Qual Life Outcomes 2006;4:54.
124. Wright A, Hannon J, Hegedus EJ, Kavchak AE. Clinimetrics corner: a closer look at the minimal clinically important difference (MCID). J Man Manip Ther 2012;20:160–6.

Summary
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Summary

Leprosy, or Hansen's disease, is an infectious disease that is characterised by hypopigmented, anaesthetic skin patches. Without timely diagnosis and treatment, the disease can be progressive and lead to permanent impairments and disabilities. Many persons affected by leprosy experience negative consequences of their condition, such as social participation restrictions and mental distress. Family members may also experience public disapproval as a consequence of being associated with their affected family member. This is caused by the very widespread negative perceptions about and practices towards persons affected and their disease. Perception is an important driver of discrimination.

The overall aim of this thesis is to contribute to a better understanding of the perception of leprosy and to explore interventions to change the perception of leprosy at individual, family, and community level. The specific research questions addressed are:

1. What are determinants of perception and knowledge of leprosy in endemic communities?
2. How does leprosy impact marital and family life?
3. How effective are posters and community meetings in changing perception and knowledge of leprosy at community level?
4. What interventions have the potential to reduce the impact of leprosy at individual and family level?

This thesis consists of three parts. In part 1, three studies were conducted on the assessment of perception of leprosy. **Chapter 2** describes a cross-sectional mixed-methods study among persons affected by leprosy, close contacts, community members and health care workers in Fatehpur district, Uttar Pradesh, India. We found that only 12.5% of the participants had adequate knowledge of leprosy, while 22% had poor knowledge. Knowledge on cause (answered correctly by 10% of the participants), mode of transmission (5%) and symptoms of leprosy (16%) was especially poor. We also revealed high levels of stigma, fear of infection and desire to keep social distance towards persons affected by leprosy among community members. Finally, better knowledge of leprosy was associated with lower levels of social distance towards persons affected by leprosy.

Chapter 3 describes a cross-sectional mixed-methods study in which we examined the differences and commonalities in and determinants of perceptions of leprosy in endemic districts in India and Indonesia. We found that 88% of the participants in India and 90% of the participants in Indonesia had inadequate knowledge of

leprosy. In both countries, cause, mode of transmission, early symptoms and contagiousness of leprosy were least known, and treatment and treatability of leprosy were best known. In both countries, health workers knew most about leprosy and community members the highest levels of stigma. Local beliefs and misconceptions differed between countries, for instance in Indonesia leprosy is believed to run in the family for seven generations and in India leprosy is believed to be a result of karma. The determinants of leprosy knowledge and stigma as measured in this study explained very little of the variability in levels of knowledge and stigma. We did identify three main causes of stigma: (1) poor knowledge and misconceptions about leprosy, (2) local beliefs, and (3) fear of contagion.

In **Chapter 4** we described the Perception Study Toolkit (PST). The PST can help identify specific beliefs, knowledge gaps, misconceptions, and fears to inform community education and behaviour change interventions and can be used to monitor and evaluate such interventions. The toolkit consists of four measures: (1) a Communication Needs Assessment questionnaire, (2) a Knowledge Attitudes and Practices measures, (3) the EMIC community stigma scale and (4) the Social Distance Scale. It also comprises qualitative methods to investigate perception: the way people see leprosy, what they know about leprosy and their attitudes, beliefs and reported behaviour towards persons affected by leprosy. We argue that using a standard toolkit like the PST would enable assessment of the perception of leprosy or other NTDs that would allow comparison across projects and countries and the monitoring of changes over time.

Chapters 5 and 6 form part 2, that explores the impact of leprosy on marital and family life. **Chapter 5** reports the results of a cross-sectional study among three groups of women: (1) women affected by leprosy, (2) women not affected by leprosy, with visible impairments and (3) women that were 'able-bodied' (a control group) in Eastern Nepal. A total of 30 women across these three groups were interviewed about their marital and sexual relationship. Women affected by leprosy appeared to face additional problems with regard to their marital and sexual relationships as compared to women with physical disabilities and 'able-bodied' women. Women affected by leprosy experienced problems during treatment, such as not daring to have intercourse at all due to fear of contagion, experiencing more distance from close others, and husband and wife sleeping in separate beds. In addition, we found that some women affected by leprosy were abandoned or sexually abused by their husband. Some problems may have been caused or aggravated by the fact that almost half of the women did not know the cause and mode of transmission of their disease. In addition, we found that some of the problems women experienced in their marital life related to the fear of leprosy.

Chapter 6 describes a cross-sectional study among persons with visible impairments due to leprosy, podoconiosis and lymphatic filariasis, and their family members. We found that restrictions in social participation (isolation, not being invited to weddings, the houses of friends and coffee ceremonies and barriers to employment and education), reduced productivity and marginalisation were common for persons affected. In addition, difficulties in finding a spouse and divorce were common for persons affected. Many persons affected reported mental distress. While most people received social and physical support from their families, there were a few exceptions. In particular, persons with younger children seemed to lack social support. Family members also experienced discrimination, including difficulties finding a spouse. In addition, having to provide for their affected family member sometimes caused stress, school dropouts and an additional workload. Financial problems and loss of livelihood were reported by almost all participants.

In part 3, consisting of chapters 7, 8, 9 and 10, we explored interventions to change perceptions about leprosy at community level and interventions to reduce the impact of leprosy at individual and family level. **Chapter 7** describes a pre/post intervention study in which we developed and evaluated posters and community meetings to change perceptions of leprosy in two leprosy-endemic districts in India. We used the Perception Study Toolkit described in chapter 4 to evaluate the effect of the interventions. We found that the interventions were highly effective in improving knowledge and reducing community stigma, and a moderately effective in improving personal attitudes. We revealed that the more posters participants had seen, the better their knowledge of leprosy, and the lower their stigma scores. Findings from this study suggest that contextualised posters and community meetings are effective in increasing leprosy-related knowledge and changing the perception of leprosy in Fatehpur and Chandauli districts, India.

Chapter 8 reports findings from a quasi-experimental mixed-methods (proof of concept) study in Ethiopia. We developed and evaluated a family-based intervention for prevention and self-management of disabilities due to leprosy, podoconiosis and lymphatic filariasis. The family-based intervention was developed based on the results of an exploratory study (chapter 6) and consisted of self-management of disabilities, awareness raising and socio-economic empowerment. The intervention was delivered during up to eight monthly group meetings. We found no significant reduction of eye and hand problems or increase of activity levels after the intervention. However, the number of foot and leg impairments, leg and foot circumference, and number of acute attacks all significantly decreased at follow-up compared to baseline. In addition, family quality of life scores significantly improved from 76.9 to 84.1 for persons affected by leprosy and from 67.4 at baseline to 89.9 at follow-up for family members ($p<0.001$). Stigma levels significantly decreased from 24.0 at baseline to 16.7 at follow-up ($p<0.001$). Findings from this proof of concept study suggest that the family-based intervention had a very positive effect on impairments and self-management of disabilities, family quality of life and stigma.

Chapter 9 explores key sources of strength and resilience for persons receiving services for leprosy in Porto Velho, Brazil. Persons affected by leprosy and healthcare providers were interviewed. While a few individuals affected were provided with formal psychological support in the early phases of their treatment, we found that many participants noted the importance of providing such support at this time. This study revealed that relationships with and social support from family members, friends and others affected by leprosy was a primary source of resilience. A key context for building resilience was through the peer-level sharing and engagement experienced in self-care and support groups. Participants also emphasised the importance of providing appropriate information about leprosy and the importance of beliefs and spirituality.

In **chapter 10** we used a quasi-experimental, before-after study design with a mixed-methods approach to evaluate a 10-week family-based intervention, designed to strengthen the resilience of individuals and families by enhancing their protective abilities and capacity to overcome adversity. The study was conducted in two sites, urban areas in Telangana state, and in rural areas in Odisha state, India. Persons affected by leprosy and their family members were included. We found a statistically significant increase in resilience scores for persons affected and family members from Odisha state. This improvement was maintained at a six-month follow-up. There was no increase in resilience scores post-intervention among participants from Telangana state. Quality of life scores were significantly higher at follow-up for persons affected in both states, and for family members in Odisha state. All participants described benefitting from participating in the programme. Participants especially appreciated the social dimensions of the intervention. This pilot study showed that the intervention was feasible and has the potential to improve resilience and quality of life of persons affected by leprosy and their family members.

Finally, **chapter 11** contains answers to the research questions, an overall discussion, and a number of recommendations for future research and practice.

Recommendations for practice:

- We found that psychological support in the early phases of diagnosis and treatment was a priority for persons affected by leprosy. We also found that leprosy can negatively influence marital and family life in several ways, some problems seemed to have been caused by misconceptions about leprosy and a lack of knowledge. We therefore recommend counselling and education of persons affected by leprosy *and* their family members at diagnosis, to help prevent some of the problems reported.
- This thesis has highlighted the importance of strengthening social support and connections. Social support and connections can be strengthened at the family level, but friends, peers or religious or other groups can also play an important role. We recommend that support groups are established to reduce the impact of leprosy at individual and family level.
- We found that contextualised posters and community meetings were effective in changing community and personal attitudes of community members and close contacts towards leprosy. The effectiveness of these interventions in changing attitudes could likely be further improved by combining these interventions with other approaches, such as facilitating contact between persons with and without leprosy. We recommend that programme managers use multi-level and multi-faceted interventions to improve perceptions of leprosy.
- We strongly recommend working closely with the target population and local stakeholders, when developing interventions. This maximises the likelihood that the interventions fit with the target groups' needs and are acceptable, while promoting the uptake of the interventions by policymakers.

Recommendations for further research:

- To explore additional determinants of perceptions and knowledge, to get a better understanding of the underlying mechanisms behind perceptions and knowledge, and ultimately to change these perceptions.
- To explore how religion and religiosity influence resilience and perceptions of leprosy, and what role religiosity could play in strengthening resilience and changing perceptions.
- To explore what role key influential people, like heads of the village, religious leaders, and media personnel, can play in changing the perception of leprosy and in reducing stigma.
- To investigate the extent of the problems in marital and family life of persons affected by leprosy and their family members, using larger, random samples and control groups, and ways to ameliorate the situation.
- To further explore ways to improve assessment of the clinical relevance1 of the impact of interventions.
- To explore meaningful ways to measure changes in behaviour and indicators of behaviour change.

Samenvatting

Lepra, of de ziekte van Hansen, is een infectieziekte die wordt gekenmerkt door verkleurde, gevoelloze vlekken op de huid. Zonder tijdige diagnose en behandeling kan de ziekte progressief zijn en tot blijvende beperkingen en en tot blijvende lichamelijke beperkingen leiden. Veel mensen met lepra ondervinden hier negatieve gevolgen van, zoals beperkingen op het gebied van sociale participatie en psychische problemen. Ook familieleden kunnen afkeuring van anderen ervaren als gevolg van de omgang met hun familielid met lepra. Dit wordt veroorzaakt door de zeer wijdverspreide negatieve percepties over en praktijken ten aanzien van mensen met de ziekte. Perceptie is een belangrijke aanjager van discriminatie.

Het doel van dit proefschrift is om bij te dragen aan een beter begrip van de perceptie van lepra en om interventies te onderzoeken om de perceptie van lepra op individueel-, gezins- en gemeenschapsniveau te veranderen. De specifieke onderzoeksvragen die aan de orde komen zijn:

1. Wat zijn determinanten van perceptie en kennis van lepra in endemische gemeenschappen?
2. Welke invloed heeft lepra op het huwelijk en gezinsleven?
3. Hoe effectief zijn posters en bijeenkomsten in het veranderen van perceptie en kennis van lepra op gemeenschapsniveau?
4. Welke interventies hebben het potentieel om de impact van lepra op individueel en gezinsniveau te verminderen?

Dit proefschrift bestaat uit drie delen. In deel 1 zijn drie onderzoeken uitgevoerd naar de meting van de perceptie van lepra. **Hoofdstuk 2** beschrijft een cross-sectionele studie met verschillende methoden onder personen met lepra, nauwe contacten, leden van de gemeenschap en gezondheidswerkers in het district Fatehpur, Uttar Pradesh, India. We ontdekten dat slechts 12,5% van de participanten voldoende kennis had over lepra, terwijl 22% slechte kennis had. Vooral de kennis over de oorzaak (correct beantwoord door 10% van de participanten), de wijze van overdracht (5%) en de symptomen van lepra (16%) was slecht. We onthulden ook hoge niveaus van stigmatisering, angst voor infectie en de wens om sociale afstand te bewaren tot personen met lepra onder leden van de gemeenschap. Ten slotte werd betere kennis van lepra geassocieerd met minder sociale afstand tot mensen met lepra.

Hoofdstuk 3 beschrijft een cross-sectionele studie met verschillende methoden waarin we de verschillen en overeenkomsten in en determinanten van percepties van lepra in endemische districten in India en Indonesië onderzochten. We ontdekten dat 88% van de participanten in India en 90% van de participanten in Indonesië onvoldoende kennis hadden van lepra. In beide landen was kennis over oorzaak, wijze van overdracht, vroege symptomen en besmettelijkheid van lepra het laagst, en was kennis over de behandeling en behandelbaarheid van lepra het hoogst. In beide landen wisten gezondheidswerkers het meest van lepra en hadden leden van de gemeenschap de meeste stigmatiserende opvattingen over mensen met lepra. Lokale opvattingen en misvattingen verschilden van land tot land, in Indonesië bijvoorbeeld wordt aangenomen dat lepra zeven generaties lang in de familie voorkomt en in India wordt aangenomen dat lepra het gevolg is van karma. De determinanten van kennis over lepra en stigmatisering zoals gemeten in deze studie verklaarden zeer weinig van de variabiliteit in kennisniveaus en stigmatisering. We identificeerden drie hoofdoorzaken van stigma: (1) gebrekkige kennis en misvattingen over lepra, (2) lokale overtuigingen en (3) angst voor besmetting.

In **hoofdstuk 4** hebben we de Perceptie Studie Toolkit (PST) beschreven. De PST kan helpen bij het identificeren van overtuigingen, hiaten in kennis, misvattingen en angsten om educatief materiaal en gedragsveranderingsinterventies te ontwikkelen en kan worden gebruikt om dergelijke interventies te monitoren en evalueren. De toolkit bestaat uit vier instrumenten: (1) een vragenlijst om communicatie behoeften te inventariseren, (2) een Knowledge, Attitudes and Practices meetinstrument, (3) de EMIC-gemeenschapsstigmaschaal en (4) de Social Distance Scale. Het omvat ook kwalitatieve methoden om perceptie te onderzoeken: de manier waarop mensen naar lepra kijken, wat ze weten over lepra en hun attitudes, overtuigingen en gerapporteerd gedrag ten opzichte van mensen met lepra. We stellen dat het gebruik van een standaardtoolkit zoals de PST het mogelijk zou maken om de perceptie van lepra of andere verwaarloosde tropische ziekten te beoordelen, zodat projecten en landen kunnen worden vergeleken en veranderingen in de tijd kunnen worden gevolgd.

De hoofdstukken 5 en 6 vormen samen deel 2, waarin de impact van lepra op het huwelijks- en gezinsleven wordt onderzocht. **Hoofdstuk 5** rapporteert de resultaten van een cross-sectioneel onderzoek onder drie groepen vrouwen: (1) vrouwen met lepra, (2) vrouwen zonder lepra, met zichtbare beperkingen en (3) vrouwen zonder beperkingen (een controlegroep) in Oost-Nepal. In totaal werden 30 vrouwen uit deze drie groepen geïnterviewd over hun huwelijk en seksuele relatie. Vrouwen met lepra bleken met extra problemen te kampen met betrekking tot hun huwelijk en seksuele relaties, in vergelijking met vrouwen met een lichamelijke beperking en vrouwen uit de controlegroep. Vrouwen met lepra ondervonden problemen tijdens de behandeling, zoals geen geslachtsgemeenschap durven hebben uit angst voor besmetting, meer afstand tot naasten, en het in aparte bedden slapen van man en vrouw. Bovendien ontdekten we dat sommige vrouwen met lepra door hun echtgenoot in de steek werden gelaten of seksueel werden misbruikt. Sommige problemen kunnen zijn veroorzaakt of verergerd doordat bijna de helft van de vrouwen met lepra de oorzaak en wijze van overdracht van hun ziekte niet kende. Bovendien ontdekten we dat sommige van de problemen die vrouwen in hun huwelijk ondervonden, verband hielden met de angst voor lepra.

Hoofdstuk 6 beschrijft een cross-sectionele studie onder personen met zichtbare beperkingen als gevolg van lepra, podoconiose en elefantiase, en hun familieleden. We ontdekten dat beperkingen in sociale participatie (isolatie, niet worden uitgenodigd voor bruiloften, de huizen van vrienden en koffieceremonies, en barrières in werk en onderwijs), verminderde productiviteit en marginalisatie veel voorkwamen bij de personen met zichtbare beperkingen. Bovendien kwamen moeilijkheden bij het vinden van een echtgenoot en echtscheiding veel voor bij de personen met zichtbare beperkingen. Veel van deze mensen gaven aan psychische onrust te voelen. Hoewel de meeste mensen sociale en fysieke ondersteuning kregen van hun familie, waren er een paar uitzonderingen. Vooral personen met jongere kinderen leken geen sociale steun te hebben. Familieleden ondervonden ook discriminatie, waaronder moeilijkheden bij het vinden van een echtgenoot. Bovendien veroorzaakte het moeten zorgen voor hun getroffen gezinslid soms stress, schooluitval en extra werkdruk. Financiële problemen en verlies van inkomsten werden door bijna alle participanten gemeld.

In deel 3 van het proefschrift, bestaande uit de hoofdstukken 7, 8, 9 en 10, hebben we interventies onderzocht om de perceptie van lepra op gemeenschapsniveau te veranderen en interventies om de impact van lepra op individueel- en gezinsniveau te verminderen. **Hoofdstuk 7** beschrijft een voor/na-interventiestudie waarin we posters ontwikkelden en bijeenkomsten organiseerden en evalueerden om de perceptie van lepra in twee lepra-endemische districten in India te veranderen. We gebruikten de Perceptie Studie Toolkit beschreven in hoofdstuk 4 om het effect van de interventies te evalueren. We ontdekten dat de interventies zeer effectief waren in het verbeteren van kennis en het verminderen van gemeenschapsstigma, en matig effectief waren in het verbeteren van persoonlijke attitudes. We onthulden dat hoe meer posters de participanten hadden gezien, hoe beter hun kennis van lepra en hoe lager hun stigmascores. Bevindingen uit deze studie suggereren dat gecontextualiseerde posters en bijeenkomsten effectief zijn bij het vergroten van lepra-gerelateerde kennis en het veranderen van de perceptie van lepra in de districten Fatehpur en Chandauli, India.

Hoofdstuk 8 rapporteert bevindingen van een quasi-experimentele (*proof of concept*) studie met verschillende methoden in Ethiopië. We ontwikkelden en evalueerden een gezinsgerichte interventie voor preventie en zelfmanagement van lichamelijke beperkingen als gevolg van lepra, podoconiose en elefantiase. De interventie was gebaseerd op de resultaten van een verkennend onderzoek (hoofdstuk 6) en bestond uit zelfmanagement van lichamelijke beperkingen, het creëren van bewustzijn en sociaaleconomische empowerment. De interventie bestond uit tot acht maandelijkse groepsbijeenkomsten. We vonden geen significante vermindering van oog- en handproblemen of toename van activiteitsniveaus na de interventie. Het aantal voet- en beenbeperkingen, de been- en voetomtrek en het aantal acute aanvallen nam echter allemaal significant af bij *follow-up* in vergelijking met de nulmeting. Bovendien verbeterden de scores voor de kwaliteit van leven van het gezin significant van 76,9 naar 84,1 voor personen met lepra en van 67,4 naar 89,9 voor gezinsleden ($p<0,001$). De stigmaniveaus namen significant af van 24,0 bij de nulmeting tot 16,7 bij follow-up ($p<0,001$). Bevindingen uit deze studie suggereren dat de gezinsgebaseerde interventie een zeer positief effect had op lichamelijke beperkingen en zelfmanagement van lichamelijke beperkingen, gezinskwaliteit van leven en stigma.

Hoofdstuk 9 onderzoekt de belangrijkste bronnen van (veer)kracht voor personen die onder (na)behandeling voor lepra zijn in Porto Velho, Brazilië. Mensen met lepra en zorgverleners werden geïnterviewd. Hoewel een paar mensen met lepra in de vroege fasen van hun behandeling daadwerkelijk formele psychologische ondersteuning kregen, vonden we dat veel participanten het belang van dergelijke ondersteuning tijdens deze vroege fasen benadrukten. Uit dit onderzoek bleek dat relaties met en sociale steun van familieleden, vrienden en anderen met lepra de voornaamste bron van veerkracht waren. Een belangrijke plek om veerkracht te versterken, was tijdens het delen met en participeren in zelfzorg en steungroepen. De participanten benadrukten ook het belang van het verstrekken van passende informatie over lepra en het belang van geloof en spiritualiteit.

In **hoofdstuk 10** gebruikten we een quasi-experimenteel, voor/na-interventiestudie met verschillende methoden om een 10 weken durende gezinsgebaseerde interventie te evalueren, ontworpen om de veerkracht van individuen en gezinnen te versterken, door hun beschermende capaciteiten en vermogen om met tegenslag om te gaan te verbeteren. Het onderzoek werd uitgevoerd op twee locaties: stedelijke gebieden in de staat Telangana en op het platteland in de staat Odisha, India. Personen met lepra en hun familieleden werden geïncludeerd. We vonden een statistisch significante toename in veerkrachtscores voor mensen met lepra en familieleden uit de staat Odisha. Deze verbetering was ook na zes maanden *follow-up* nog aanwezig. Er was geen toename in veerkrachtscores na de interventie onder participanten uit de staat Telangana. De scores voor kwaliteit van leven waren significant hoger bij follow-up mensen met lepra in beide staten, en voor familieleden in de staat Odisha. Alle participanten gaven aan iets aan deelname aan de interventie gehad te hebben. De participanten konden met name de sociale dimensies van de interventie waardeerden. Deze pilotstudie toonde aan dat de interventie uitvoerbaar is en het potentieel heeft om de veerkracht en kwaliteit van leven van mensen met lepra en hun familieleden te verbeteren.

Ten slotte bevat **hoofdstuk 11** antwoorden op de onderzoeksvragen, een algemene discussie en een aantal aanbevelingen voor toekomstig onderzoek en praktijk.

Aanbevelingen voor de praktijk:

- We ontdekten dat psychologische ondersteuning in de vroege fasen van diagnose en behandeling een prioriteit was voor mensen met lepra. We ontdekten ook dat lepra het huwelijk en het gezinsleven op verschillende manieren negatief kan beïnvloeden. Sommige problemen leken te zijn veroorzaakt door misvattingen over lepra en een gebrek aan kennis over lepra. We raden daarom aan om mensen met lepra *en* hun familieleden bij de diagnose te begeleiden en voor te lichten, om enkele van de gemelde problemen te helpen voorkomen.
- Dit proefschrift heeft het belang benadrukt van het versterken van sociale steun en verbinding. Sociale steun en verbindingen kunnen op gezinsniveau worden versterkt, maar ook vrienden, leeftijdsgenoten of religieuze of andere groepen kunnen hierin een belangrijke rol spelen. We raden aan om steungroepen op te richten om de impact van lepra op individueel- en gezinsniveau te verminderen.
- We ontdekten dat contextualiseerde posters en bijeenkomsten effectief waren in het veranderen van de gemeenschaps- en persoonlijke attitudes van dorpsgenoten van mensen met lepra. De effectiviteit van deze interventies bij het veranderen van attitudes zou waarschijnlijk verder kunnen worden verbeterd door deze interventies te combineren met andere benaderingen, zoals het faciliteren van contact tussen personen met en zonder lepra. We raden programmamanagers aan om *multi-level* en veelzijdige interventies te gebruiken om de perceptie van lepra te veranderen.
- We raden sterk aan om nauw samen te werken met de doelgroep en lokale stakeholders bij het ontwikkelen van interventies. Dit maximaliseert de kans dat de interventies aansluiten bij de behoeften van de doelgroepen en acceptabel zijn, bovendien bevorderd dit de acceptatie van de interventies door beleidsmakers.

Aanbevelingen voor verder onderzoek:

- Het onderzoeken van aanvullende determinanten van percepties en kennis, om zo meer inzicht te krijgen in de onderliggende mechanismen achter percepties en kennis, om zo uiteindelijk deze percepties te kunnen veranderen.
- Onderzoeken hoe religie en religiositeit veerkracht en percepties van lepra beïnvloeden, en welke rol religiositeit zou kunnen spelen bij het versterken van veerkracht en het veranderen van percepties.
- Onderzoeken welke rol invloedrijke mensen, zoals dorpshoofden, religieuze leiders en mediapersoneel, kunnen spelen bij het veranderen van de perceptie van lepra en bij het verminderen van stigmatisering.
- Het onderzoeken van de omvang van de problemen in het huwelijks- en gezinsleven van mensen met lepra en hun gezinsleden, door meer en willekeurig geselecteerde participanten en controlegroepen te includeren, en het onderzoeken van manieren om de situatie te verbeteren.
- Verder zoeken naar manieren om de beoordeling van de klinische relevantie van de impact van interventies te verbeteren.
- Het onderzoeken naar manieren om gedragsveranderingen te meten en het onderzoeken van indicatoren om gedrag te meten.

Dankwoord

Dit proefschrift was er niet geweest zonder de steun en het vertrouwen van velen, dank daarvoor! Er zijn een aantal mensen die ik in het bijzonder wil bedanken.

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About the author

Anna van ‘t Noordende was born on December 28, 1990, in Den Helder and grew up on the island of Texel. In 2009 she received her VWO diploma at OSG de Hogeberg in Den Burg, Texel. After graduation she travelled through Australia and New Zealand for seven months. From 2010 to 2013 she studied Lifestyle Informatics (Artificial Intelligence), during which she also took extra courses mainly in the field of psychology and anthropology, at the VU University in Amsterdam. She obtained her Bachelor’s degree in 2013. In 2015 she obtained her Master’s degree in Management, Policy Analysis and Entrepreneurship in the Health and Life Sciences (MPA), with specialization International Public Health, at the VU University. During this time, she conducted two research internships, one for NLR in Nepal and one for the Neglected Tropical Disease Non-Governmental Development Organisation Network in Brazil. This was also when she became acquainted with leprosy research and practice for the first time. After graduation, from 2015 to 2017, she worked for the International Federation of Anti-Leprosy Associations (ILEP). In 2017, she also became involved with Disability Studies in Nederland, for which she continues to work several hours a week as a researcher. In June 2017 she started as Programme Support and Research Officer, a combination of programme support responsibilities (50%) and a PhD-assignment (50%), at NLR’s PEP++ project. While the PEP++ project aims to test the efficacy of an enhanced chemoprophylaxis regimen for the prevention of leprosy, and to stop the transmission of leprosy in endemic areas in India, Indonesia and Brazil, her research work focused on the perception of leprosy. During this time, she also (co-)initiated and worked on collaborative Leprosy Research Initiative (LRI) funded research projects in leprosy. This thesis is the synthesis of her scientific work.

Publications

This thesis:

1. **van ‘t Noordende, A. T.,** Korfage, I., Lisam, S., Arif, M. A., Kumar, A., & van Brakel, W. H. (2019). The role of perceptions and knowledge of leprosy in the elimination of leprosy: A baseline study in Fatehpur district, northern India. *PLoS Neglected Tropical Diseases*, *13*(4), e0007302.
2. **van ‘t Noordende, A. T**., Lisam, S., Ruthindartri, P., Sadiq, A., Singh, V., Arifin, M., van Brakel, W. H. & Korfage, I. J. (2021). Leprosy perceptions and knowledge in endemic districts in India and Indonesia: Differences and commonalities. *PLoS Neglected Tropical Diseases*, *15*(1), e0009031.
3. **van ‘t Noordende, A. T.**, van Brakel, W.H. (2021). Towards a cross-neglected tropical disease perception study toolkit: a prototype toolkit developed in the field of leprosy. *Leprosy Review*, *92*(2), 170-181.
4. **van ‘t Noordende, A. T**., van Brakel, W. H., Banstola, N., & Dhakal, K. P. (2016). The impact of leprosy on marital relationships and sexual health among married women in eastern Nepal. *Journal of Tropical Medicine*, *2016,* 88-96.
5. **van ‘t Noordende, A. T**.*, Aycheh, M. W.*, & Schippers, A. (2020). The impact of leprosy, podoconiosis and lymphatic filariasis on family quality of life: a qualitative study in northwest Ethiopia. *PLoS Neglected Tropical Diseases*, *14*(3), e0008173.
6. **van ‘t Noordende, A. T**., Lisam, S., Singh, V., Sadiq, A., Agarwal, A., Hinders, D., Richardus, J. H., van Brakel, W. H. & Korfage, I. J. (2021). Changing perception and improving knowledge of leprosy: an intervention study in Uttar Pradesh, India. *PLoS Neglected Tropical Diseases 15*(8), e0009654.
7. **van ‘t Noordende, A. T**., Wubie Aycheh, M., Tadesse, T., Hagens, T., Haverkort, E., & Schippers, A. P. (2021). A family-based intervention for prevention and self-management of disabilities due to leprosy, podoconiosis and lymphatic filariasis in Ethiopia: A proof of concept study. *PLoS Neglected Tropical Diseases*, *15*(2), e0009167.
8. **van ‘t Noordende, A. T**., Bakirtzief da Silva Pereira, Z., & Kuipers, P. (2021). Key sources of strength and resilience for persons receiving services for Hansen's disease (leprosy) in Porto Velho, Brazil: What can we learn for service development? *International Health,* ihab001.
9. **van ‘t Noordende, A. T**. Bakirtzief da Silva Pereira, Z., Biswas, P., Ilyas, M., Krishnan, V., Parasa, J. & Kuipers, P. (2021). Strengthening individual and family resilience against leprosy-related discrimination: a pilot intervention study. *PLoS Neglected Tropical Diseases*, *15*(4), e0009329.

Other peer-reviewed scientific publications:

10. **van ‘t Noordende, A. T**., Kuiper, H., Ramos, A. N., Mieras, L. F., Barbosa, J. C., Pessoa, S. M., Souza, E. A., Fernandes, T. A., Hinders, D. C., Praciano, M. M. A., & van Brakel, W. H. (2016). Towards a toolkit for cross-neglected tropical disease morbidity and disability assessment. *International Health*, *8* (suppl_1), i71-i81.
11. **van ‘t Noordende, A. T**., Lacey Krylova, V., Duck, M., & Kuipers, P. (2017). Focusing anti-discrimination efforts in areas of most relevance to people affected by leprosy. *Leprosy Review*, *88*(3), 410-415.
12. **van ‘t Noordende, A. T**., Hinders, D., Tiwari, A., Richardus, J. H., & van Brakel, W. (2019). A leprosy elimination investment case: proceedings of an expert consultation. *Leprosy Review*, *90*(1), 124-127.
13. **van ‘t Noordende, A. T**., Kuipers, P., & Pereira, Z. B. D. (2019). Strengthening personal and family resilience: a literature review for the leprosy context. *Leprosy Review*, *90*(1), 88-104.
14. **van ‘t Noordende, A. T**.*, Aycheh, M. W.*, & Schippers, A. P. (2020). An exploration of family quality of life in persons with leprosy-, lymphatic filariasis–and podoconiosis-related disabilities and their family members in Ethiopia. *Transactions of The Royal Society of Tropical Medicine and Hygiene*, *114*(12), 1003-1012.
15. Dahiru, T., Iliyasu, Z., Mande, A. T., **van ‘t Noordende, A. T**., Aliyug, M. H. Emotions, relationships, and behavior: A qualitative study into experiences of stigma among persons affected by leprosy in northern Nigeria. (Submitted)
16. Dahiru, T., Iliyasu, Z., Mande, A. T., **van ‘t Noordende, A. T.**, Aliyug, M. H. Community perspectives on leprosy and related stigma in northern Nigeria: A qualitative study. (Submitted)
17. Meis, M. J., **van ‘t Noordende, A. T.**, Mieras, L., Banstola, N., Dhakal, K. P., Essink, D. R., van Brakel, W. H. The impact of leprosy and physical disability on marital and sexual relationships of married Nepali men. (Submitted)
18. **van ’t Noordende, A. T.,** Aycheh, M. W., Moges, N. A., Tadesse, T., Schippers, A. P. A family-based intervention for prevention and self-management of disabilities due to leprosy, podoconiosis and lymphatic filariasis versus usual care in Ethiopia: study protocol for a cluster-randomised controlled trial. (Submitted)
19. Susanto, D. F. P.*, **van ’t Noordende, A. T.***, Septian, E. R., van Brakel, W. H., Peters, R. M. H., Irwanto, I. The influence of leprosy and other disabilities on marital relationships and sexual health among married women in Indonesia: a qualitative study into experiences and coping. (Submitted)

Other publications:

20. **van ‘t Noordende, A. T.,** Mangeard-Lourme, J., Bakirtzief da Silva Pereira, Z., Shrubsole, G., Augustine, V., Kunju, J. P., Gwaikolo, W., & Warne, G. (2020). ILEP/NNN. Guides on Stigma and Mental Wellbeing. Guide 2. How to reduce the impact of stigma. International Federation of Anti-Leprosy Associations and Neglected Tropical Disease NGO Network: *https://www.infontd.org/toolkits/stigma-guides/guide-2-how-reduce-impact-stigma*
21. **van ‘t Noordende, A. T.*,** & Broekkamp, H.* (July 2020). Why we need to focus on stigma and discrimination — 5 lessons from the NTD field. Devex: *https://www.devex.com/news/opinion-why-we-need-to-focus-on-stigma-and-discrimination-5-lessons-from-the-ntd-field-97676*
22. **van ‘t Noordende, A. T.*,** & Geutjens, R.* (October 2020). We are all responsible for addressing Hansen's disease-related stigma. InfoHansen: *https://en.infohansen.org/blog/we-are-all-responsible*

** Shared first author.*

PhD Portfolio

Name PhD student:	A.T. van ‘t Noordende	**PhD period:**	2018-2022
Erasmus MC Department:	Public Health	**Promotor:**	Prof. dr. J.H. Richardus
Research School:	NIHES	**Supervisors:**	Dr. I.J. Korfage Dr. W.H. van Brakel

1. PhD training	**Year**	**ECTS**
Courses at the Netherlands Institute for Public Health (NIHES)		
ESP38 Conceptual Foundation Epidemiologic Study Design	2017	0.7
ESP03 Introduction to Data-analysis	2017	0.7
ESP09 Regression Analysis	2017	1.4
ESP66 Logistic Regression	2018	1.4
ESP11 Methods of Public Health Research	2018	0.7
HS02c Public Health Research: Intervention Development and Evaluation	2019	1.9
Other courses		
Doelrealisatie (Goede Doelen Nederland & MDFnl)	2017	0.9
Good Clinical Practice (ICH GCP) (Whitehall Training, virtual)	2018	0.5
Business safety and security training (Expat Preventive)	2018	0.6
Cursus personeelsvertegenwoordiging (Goede Doelen Nederland & MDFnl)	2018	0.3
Advanced female traveller security training (Expat Preventive)	2018	0.3
Introduction in Advocacy and Policy Influencing (API) (MDF)	2019	0.1
Cursus presenteren (Speechen.nl)	2020	0.9
Foundations of Public Health Practice: Behaviour& Behaviour Change (Imperial College London, virtual)	2020	0.6
Projectmanagement en Projectmatig werken (projectmanagement-training.nl)	2021	0.9
Seminars and workshops		
Leprosy elimination investment case meeting, Amsterdam, the Netherlands	2017	0.3
Mapping workshop, Amsterdam, the Netherlands	2017	0.3
NLR Key Priority Programme 1 workshop, Utrecht, the Netherlands	2017	0.6
3rd International Disability Studies Conference 'The Art of Belonging', Amsterdam, the Netherlands	2017	0.3
Population at Risk meeting, Amsterdam, the Netherlands	2018	0.3
Leprosy Research Initiative Spring meeting, Veenendaal, the Netherlands	2018, 2019, 2021	0.6

1. PhD training	Year	ECTS
NLR Key Priority Programme 4 workshop, Utrecht, the Netherlands	2018	0.6
Neglected Tropical Disease NGO Network (NNN) Temporary Expert Group on the revision of the stigma and mental wellbeing guidelines, working meeting, Utrecht, the Netherlands	2018	0.9
Community Education and Behaviour Change workshop, Delhi, India	2018	0.6
Community Education and Behaviour Change workshop, Surabaya, Indonesia	2019	0.9
PEP++ Scientific Steering Committee meeting, Varanasi, India	2019	0.9
PEP++ Scientific Steering Committee meeting, Manila, the Philippines	2019	0.3
Mapping workshop, Fortaleza, Brazil	2019	0.6
Community Education and Behaviour Change workshop, Fortaleza, Brazil	2019	0.6
Working meeting on resilience intervention, Hyderabad, India	2020	0.9
Erasmus MC, University Medical Center Rotterdam, Digital PhD day, virtual	2021	0.2
Conferences		
3rd International Conference Disability Studies 'The Art of Belonging', Amsterdam, the Netherlands	2017	0.3
Worldviews and health-related stigma, VUB Brussels Humanities, Sciences & Engineering Campus, Brussels, Belgium	2018	0.3
20th International Leprosy Congress, Manila, the Philippines	2019	0.9
Neglected Tropical Disease NGO Network (NNN) conference, virtual	2020	0.3
International Federation of Anti-Leprosy Associations (ILEP) conference, virtual	2020	0.3
COR-NTD meeting, virtual	2020	0.2
Presentations		
ILEP Panel of Men and Women Affected by Leprosy, meeting, Würzburg, Germany.	2017	1
PEP++ Community Education and Behaviour Change workshop, Delhi, India.	2018	2
NLR Key Priority Programme 4 workshop, Utrecht, the Netherlands.	2018	1
NNN Temporary Expert Group on the revision of the stigma and mental wellbeing guidelines, Utrecht, the Netherlands.	2018	1
PEP++ Community Education and Behaviour Change workshop, Surabaya, Indonesia.	2019	2
PEP++ Scientific Steering Committee meeting, Varanasi, India.	2019	2
20th International Leprosy Congress, Manila, the Philippines.	2019	4
PEP++ Community Education and Behaviour Change workshop, Fortaleza, Brazil.	2019	2
The Leprosy Research Initiative (LRI) Spring Meeting, virtual.	2021	1

1. PhD training	Year	ECTS
Supervising students		
VU University Amsterdam, two Master students, research internship supervision	2019-2021	4
VU University Amsterdam, Master student, thesis supervision	2018	2
Leiden University, Bachelor student, thesis supervision	2021	2
Other		
Peer review of scientific articles for Leprosy Review, Journal of HIV and AIDS, Global Health Action and PLOS NTD	2017-2021	1
Member of the staff council/personeelvertegenwoordiging	2018-2020	4
Member of working Group Global Partnership for Zero Leprosy (GPZL) on stigma	2018	0.5
Member of the ILEP/NNN Temporary Expert Group on Stigma, Discrimination, and Mental Well-being (convenor of module 2 of the revision of the stigma guidelines)	2018-2020	6
Grant acquisition, Leprosy Research Initiative	2017-2021	4

Total ECTS: 62.6.