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<u>AQ-6</u>	<p>Please provide publisher location details for 'Guyatt et al. (2002)'. <u>Chicago, IL</u></p>	<input type="checkbox"/>

# People living with facial disfigurement after having had noma disease: A systematic review of the literature

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## Abstract

Noma disease often results in impairment, morbidity and severe facial disfigurement. This article reports a systematic review of literatures published between 2006 and 2015 to establish existing knowledge about social stigma associated with facial disfigurements. Five databases were searched and 114 citations were screened, of which only 15 met the relevant criteria. Titles and abstracts of the retrieved articles were independently reviewed. The research was heterogeneous; therefore, overall synthesis using meta-analysis was inappropriate. It can be seen that the review demonstrates that facial disfigurements are far more complex than was previously thought.

## Keywords

facial disfigurements, health psychology, musculoskeletal disorders, noma disease, psychological support, stigma

## Introduction

Noma disease has a community health importance as acute noma often results in malnourished children, usually aged 1–4 years. Noma is seen in the remotest and poorest populations of the world (Bisseling et al., 2010). Cancrum oris, a form of noma disease, also known as oral–facial noma, is an acute, devastating, debilitating, gangrenous and ulcerative stomatitis commonly affecting the face. It destroys the hard and soft tissue of the mouth and face. The condition manifests itself in extreme circumstances of poverty whereby the victims or the survivors of the disease are seen with functionally impaired morbidity and severe facial disfigurement (Marck et al., 1998).

Stigma is an attribute which discredits a human being ‘from a whole and usual person to a tainted, discounted one’ (Goffman,

1963: 3) and it often occurs when individuals display ‘some attribute or characteristic that conveys a social identity that is devalued in a particular social context’ (Crocker et al., 1998: 505). Major and O’Brien (2005) discuss that people who are stigmatised have different marks, and they view that

Stigmatizing marks may be visible or invisible, controllable or uncontrollable, and linked to appearance (e.g., a physical deformity), behaviour (e.g., child abuser), or group membership (e.g., African American).

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Importantly, stigma is relationship- and context-specific; it does not reside in the person but in a social context. (p. 395)

Stigma is therefore a worldwide phenomenon and an inexorable human characteristic that takes different forms depending on the cultural and historical antecedents of affected societies.

In ancient times, the word 'stigma' was used to describe a scar resulting from a burn injury or a cut to identify an individual as a criminal or a slave (Davison and Neale, 1993, 2000). [AQ1] At a later time, the context was changed from the negative meaning to a more positive meaning, 'stigmata', which was used to identify wound marks which were reported to be observed on the bodies of holy people and saints (Leff and Warner, 2006; Whitehead et al., 2001). [AQ2] The authors also described stigma as differences that may be present in people's physical appearance, personality, sexual orientation, disability, illness, age and gender. However, today the term 'stigma' is seen as an attribute that echoes in pervasive social disapproval.

Social stigma is the intense denunciation of an individual or group of individuals on some social or cultural characteristics that are recognised and which serve to distinguish them from other members of the society (Benoit and Shumka, 2009). Indeed, social stigma has the potential to go beyond the stigmatised individual and affect their families and friends because of their relationship and closeness to them. Family members and friends are not the only targets that could be affected but also healthcare professionals, especially those working in psychiatric hospitals, the mental health sector and mental health services. Evidence shows that stigmatisation could negatively affect them and may consequently have an adverse effect on their productivity and the effectiveness of their services (Benoit and Shumka, 2009; Corrihan and Watson, 2002). Also, a stigma that is a result of mental illness may cause the person affected to have difficulty in expressing their feelings and experiences, making it harder to understand them. Thus, society distances itself from mentally ill

individuals or even isolates them far away from society. World Health Organization and World Psychiatric Association (2002) have issued a technical consensus statement, to fight against stigma and discrimination towards health. Recently, stigma has been increasingly becoming an important area of research among health and social science researchers who are interested in the wellbeing of the stigmatised or vulnerable population (Bos et al., 2013; Lang et al., 2013; Lebel et al., 2013; Richards et al., 2004).

Several pieces of evidence show that stigmatised populations suffer from a high burden of disease, illness and psychosocial problems compared to the general population, and they are also prone to have a higher mortality rate, poor quality of life, decreased life expectancy and diminished access to healthcare (Fingeret et al., 2012; Rumsey et al., 2004). This category of persons may include people with disability, low-income earners, older people, immigrants and refugees, homeless people and people with mental disabilities (Benoit and Shumka, 2009; Corrihan and Watson, 2002). A growing body of literature also found that individuals having facial disfigurement have problems of adjustment to their condition as well as social difficulty (Patrick et al., 2007).

Although there is a growing body of literature on stigmatisation experienced by individuals with facial disfigurement, with regard to noma disease, with its devastating end effect of facial disfigurement on survivors (Behanan et al., 2004; Marck, 2013), only a little is known about the stigma they encounter. Therefore, to explore the prejudices associated with facial disfigurement from a wider perspective, different forms of studies were included to examine the context of other types of facial disfigurement and the stigma imposed on the victims and makes an inferential conclusion with respect to noma patients who have similar challenges or circumstances. We therefore conducted a literature review to summarise reported factors

**Table 1.** Word groups used to identify relevant articles.

Question: What are the known factors associated with and social stigma among individuals with noma disease?			
Word group 1	Word group 2	Word group 3	Word group 4
'Noma disease*' OR	'Treatment' OR 'treat*' OR	'Stigma' OR	'quality of life'
'Cancrum oris' OR	'Group support*' OR	'Social stigma' OR	'Health need' OR
'Facial necrosis' OR	'Advocacy' OR	'Prejudice' OR	'improve' OR 'improvement' OR
'Facial difference*' OR	'Counsel*' OR	'Stigmat*' OR	'outcome' OR
'Facial disfigure*' OR	'Individual support*' OR	'Discriminate' OR	'results'
'Adult debilitating disease' OR	'Community support*' OR	'discriminat*' OR	'quality'
'Necrot* gingivitis' OR	Family support	'Social isolation' OR	
'Necrotising stomatitis' OR	'Surgery' OR	'Social disability' OR	
'Oral infect*' OR	'Therapy'	'Emotional	
'Facial disfigure*' OR		communication' OR	
'Facial mutilate*' OR		'Social functioning' OR	
'Facial contract*' OR		'Perceived stigmatisat*' OR	
'Facial paralysis' OR		'social exclusion'	
'Visible differ*' OR			

- Limits:
- Population/setting: Children, human, adult, human
- Publication date: 2006-2015
- Document/record type: peer-review article
- Language: English

All groups are combined using Boolean Operators.

associated with social stigma among individuals with facial disfigurement as a consequence of diseases, congenital malformation or accidents, and in what ways the consequences of the stigma can be ameliorated.

## Methods

In this study, we carried out a systematic review of published primary studies on the subject of social stigma among individuals with facial disfigurement. As Guyatt et al. (2002) point out, systematic reviews

deal with this problem by explicitly stating inclusion and exclusion criteria for evidence to be considered, conducting a comprehensive search for evidence, and summarising the results according to the explicit rules that include how effects may vary in different subgroups. (p. 180)

Several authors argue that a systematic review offers a distinct advantage of providing relevant collated evidence that assists in making sound healthcare decisions (Baker and Weeks, 2014; Lang, 2004). The Institute of Medicine (2011),

however, warns that reviewers might 'fail to acknowledge or address the risk of reporting biases, neglect to appraise the quality of individual studies included in the review, and (they) are subject to errors during the data extraction and analysis' (p. 82). [AQ3] In addition, there are always some elements of challenge for integrating disparate evidence from different studies to establish cohesive and acceptable evidence.

### Search strategy

To identify and highlight the primary research studies, the five major electronic databases (MEDLINE, EMBASE, CINAHL, AMED, and PsychInfo) were searched, focusing on studies related to psychosocial stigma and facial disfigurement. These five databases are considered the best-known search electronic databases, having international coverage of over 80 countries and about 40,000 journals indexed, alongside their vast resources capturing the fields of psychology, healthcare and related disciplines.

Following the formulation of research questions, a research strategy was planned.

Inclusion and exclusion criteria were set, and both free terms (text-words) and index-terms (thesauruses) were established, identified and searched from research topics and relevant databases. The search strategy to address this question involved searching these electronic databases in four groups (Table 1).

### *Screening strategy*

The retrieved literatures for this systematic review were screened in two phases. In phase 1, the titles and abstracts of the retrieved articles were independently reviewed by two authors to identify those studies likely to meet the inclusion criteria. At this point, some studies were excluded which did not meet the criteria of the study. In the phase 2, a scrutiny of the full text of the articles was done, more articles were excluded and the studies that passed the screening criteria were included in the study. Also, manual screening of articles through reference checklists of retrieved articles yielded one article. The flow diagram (Moher et al., 2009) was followed to present clear guidance on how the final 15 studies were identified and screened (Figure 1).

## **Results**

A total of 114 articles were retrieved using the electronic search method. Following the removal of duplicated materials, the database search revealed 104 citations. Initially, 79 citations were excluded based on title and abstract because they did not meet the inclusion criteria. A total of 25 eligible citations passed the inclusion into the review and were assessed as full texts. Six articles were excluded, and 19 citations were included. Following the assessment of the full text of the 19 articles, 5 articles were excluded, and 14 articles passed the full-text screening and were finally included for the review. Also, one article was retrieved from the reference checklist. Overall, 15 publications were included in this study. The main findings of the articles are set out in Table 2. The findings from the chosen studies were scrutinised by employing thematic analysis. According to

Ritchie et al. (2014), thematic analysis involves 'discovering, interpreting and reporting patterns and clusters of meaning within the data' (p. 27). In this study, as Joff (2012) suggests, we reviewed the included papers and identified important or recurrent themes, and the findings were then summarised under key thematic headings. Systematically, the themes were examined on their similarities, differences and contradictions, to answer the research question about social stigma towards individuals with facial disfigurement. This resulted in the creation of analytical themes. This process that was undertaken in a structured manner was in line with Gerrish and Lacey (2010). Four major themes emerged: (1) manifestations of social stigma, (2) consequences of stigma within the individual, (3) coping with the stigma and (4) positive social influences to coping. A description of key summaries of themes and sub-themes was provided (Figure 2).

### *Manifestations of social stigma*

This theme was made up of data segments that show how society or others perceive and treat people with disfigurement. These were based on the perspectives of third-party observers and members of society, close family members or parents of the individuals with a visible disfigurement as a consequence of a disease and individuals with the disfigurement themselves. The following sub-themes reported the categories identified from the data segments from the literature showing the manifestations of societal stigma towards people with visible disfigurement.

*Negative behaviour towards people with visible disfigurement.* Several studies (Masnari et al., 2013; Rahzani et al., 2009) reported that the individuals felt a lack of societal acceptance due to how people around them interacted with them or reacted to their disfigurement. These negative behaviours included active

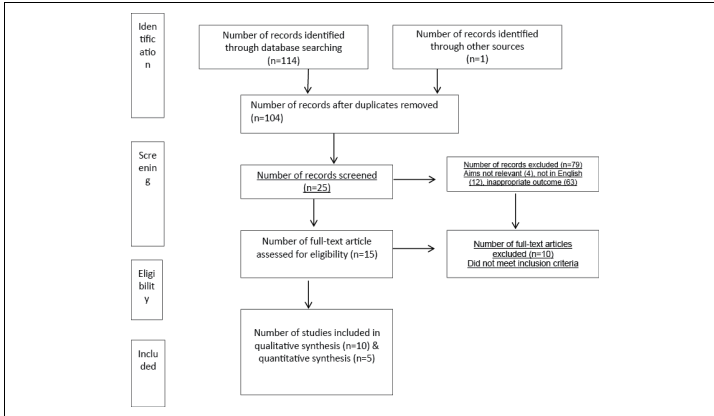


Figure 1. Flowchart of the process of identifying included studies.

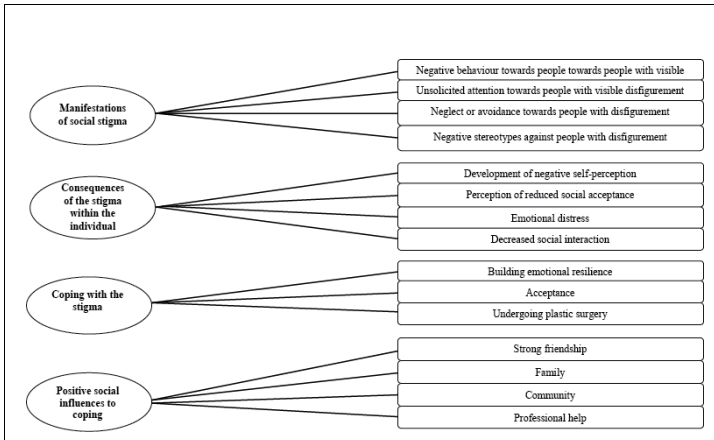


Figure 2. Descriptive themes





**Table 2. [AQ4] Articles and main findings.**

Study	Journal	Country/setting	Design/methods	Population group	Study results	Critical appraisal criteria (adapted from Smith et al. (2009))		Overall quality
						Qualitative	Quantitative	
Costa et al. (2014)	<i>Special Care in Dentistry</i>	Brazil	Qualitative, in-depth and semi-structured interview	General population	Facial disfigurements reported mostly by the consequences and impacts of the disease and surgical sequels	1, 2, 3, 4, 5, 6, 7, 8, 9, 10, 13, 14, 15	NA	Some gaps in methodology but overall convincing
Masrani et al. (2013)	<i>Body Image</i>	Switzerland	Quantitative, cross-sectional, questionnaire	School children	Children with facial difference perceived less favourably by unaffected population	NA	1, 2, 3, 5, 6, 7, 9, 10, 12, 13	Lack of methodological detail but plausible analysis
Stone and Wright (2013)	<i>Journal of Applied Social Psychology</i>	United Kingdom	Case-control	General population	Facial disfigurement or those described as wheelchair users received less favourable responses	NA	1, 2, 3, 4, 5, 6, 7, 8, 9, 10, 12, 13	Some gaps in methodology but overall convincing
Masrani et al. (2012)	<i>Journal of Plastic, Reconstructive &amp; Aesthetic Surgery</i>	Switzerland	Quantitative, cross-sectional, questionnaire	School children	Patient with a facial difference associated with higher levels of stigma	NA	1, 2, 3, 4, 5, 6, 7, 9, 10, 12, 13	Lack of methodological detail but plausible analysis
Marjike et al. (2012)	<i>Journal of Cranio-Maxillofacial Surgery</i>	Netherlands	Case-control	General population	Avoidance behaviour reported due to stigmatisation	NA	1, 2, 3, 4, 5, 6, 7, 9, 10, 12, 13	Some gaps in methodology but overall convincing
Gagnon (2012)	<i>International Journal of Nursing Studies</i>	Canada	Qualitative, semi-structured interviews	General population	Facial lipodatrophy reported vulnerable due to discrimination and stigma	1, 2, 3, 4, 7, 8, 9, 13, 14, 15	NA	Some gaps in methodology
Bogart et al. (2012)	<i>Journal of Health Psychology</i>	United states	Qualitative, focus group discussions	General population	Stigma due to facial disfigurements impact people's lives	1, 2, 3, 4, 6, 7, 9, 13, 14, 15	NA	Lack of methodological detail
Bonanno and Esmaili (2012)	<i>Clinical Journal of Oncology/Nursing</i>	United States	Qualitative, semi-structured interviews	Cancer patients	Interaction between patients with cancer who have facial disfigurement engenders stigma	1, 2, 3, 4, 7, 8, 9, 10, 12, 13, 15	NA	Some gaps in methodology
Roberts and Shute (2011)	<i>Clinical Child Psychology and Psychiatry</i>	Australia	Qualitative, semi-structured interviews	General population	Significant impact on young people's life	1, 2, 3, 4, 6, 7, 8, 9, 13, 14, 15	NA	Some gaps in methodology but overall convincing
Bonanno and Choi (2010)	<i>Health</i>	United States	Qualitative, semi-structured interviews/questionnaires	General population	Social consequences of facial disfigurement influenced mostly by the outcomes of interaction processes	1, 2, 3, 4, 7, 8, 9, 13, 15	NA	Lack of methodological detail
Feragen et al. (2010)	<i>Body Image</i>	Norway	Case-control	General population	Adolescents with a visible difference reported better functioning on all study variables compared to the comparison group	NA	1, 2, 3, 4, 5, 6, 7, 8, 9, 10, 12, 13	Lack of methodological detail but overall convincing
Prior and O'Dell (2009)	<i>Journal of Health Psychology</i>	United Kingdom	Qualitative, semi-structured interviews	General population	Psycho-social model of disfigurement associated with social world	1, 2, 3, 4, 6, 7, 8, 9, 12, 13, 15	NA	Lack of methodological detail
Rafzani et al. (2009)	<i>Qualitative Health</i>	Iran	Qualitative, face-to-face in-depth interviews	General population	Negative reactions associated with disfigured person	1, 2, 3, 4, 5, 6, 7, 8, 9, 10, 13, 15	NA	Lack of methodological detail
Hughes et al. (2009)	<i>Diversity in Health &amp; Care</i>	United Kingdom	Qualitative, focus group discussions	General population	People with disfigurements are more likely to be negatively evaluated	1, 2, 3, 4, 7, 9, 10, 12, 13, 14, 15	NA	Lack of methodological detail
Sour et al. (2008)	<i>American Journal of Tropical Medicine and Hygiene</i>	Loas	Qualitative, interviews	General population	Avoidance of social interaction associated with noma disease	2, 3, 4, 6, 7, 13	NA	Data do not support



actions that showed people's lack of acceptance towards individuals with a disfigurement. These actions ranged from joking and teasing – especially among children – that inflicted emotional harm on the individuals, to hurtful bullying that involved inflicting physical harm on the individuals. The extent of how the stigma was manifested in society had been reported to be felt by individuals as they grew up. However, these actions, or actual behaviour observed and experienced by people in society towards individuals with disfigurement, were not as common as the next manifestation of stigma.

*Unsolicited attention towards people with visible disfigurement.* There have been more reports (Bonanno and Choi, 2010; Hughes et al., 2009; Masnari et al., 2012; Rahzani et al., 2009; Roberts and Shute, 2011) of unsolicited attention towards individuals with disfigurement, rather than actual negative actions or behaviour that reflected the stigma towards individuals with a visible difference. This unsolicited attention included observations of staring at others, which was the most commonly reported manifestation. It also included social interactions that involved the disfigurement as the main topic. Although not particularly negative, such as teasing reported in the first category, these interactions typically involved pity or sympathy from others that could be viewed as either negative or positive by the individual, depending on his or her personality or outlook.

*Neglect or avoidance towards people with disfigurement.* As opposed to the first category, which involved active behaviour targeted at the individuals with visible differences, the third category under the manifestation of stigma in society was reported in the studies (Bonanno and Choi, 2010; Rahzani et al., 2009) as passive behaviour that could be either deliberate or not, that showed the individual that he or she was being avoided by people around him or her. Interestingly, it has been reported by some studies that some individuals felt less stigmatised when other people simply

neglected their disfigurement; therefore, this category gathered mixed responses since some felt negative about being avoided, and others felt better about their disfigurement if people just ignored them.

*Negative stereotypes against people with disfigurement.* Several studies (Bogart et al., 2012; Gagnon, 2012; Hughes et al., 2009) have reported that some people attached negative stereotypes or associations with people with disfigurement, as they were viewed as less capable, less attractive and less intelligent compared to their counterparts without disfigurement. Other people also often associated disfigurement with a deficiency, disorder or disease.

#### *Consequences of the stigma within the individual*

This theme involved the negative impacts of the experiences of stigma in society on the internal functioning, beliefs and perceptions of individuals with visible difference, which included psychological, emotional, cognitive and behavioural responses of individuals with disfigurement. As a result of their experiences with how society treats them, this involved the reported perspectives of the individuals with disfigurement themselves and their close family relatives, especially parents. This theme was made up of four sub-themes/categories, as discussed in the following subsections.

*Development of negative self-perception.* Some studies (Costa et al., 2014; Masnari et al., 2013; Rahzani et al., 2009; Roberts and Shute, 2011) have reported the negative impact of social stigma on the individual with disfigurement as having to do with the individuals developing a negative perception of themselves and seeing themselves as less beautiful and less attractive than their counterparts without the disfigurement. They became self-conscious and embarrassed by their physical appearance such that they developed low self-esteem, and they were uncomfortable with the way they looked to others.

*Perception of reduced social acceptance.* As opposed to the actual experiences that showed lack of social acceptance, different studies (Bogart et al., 2012; Bonanno and Esmaeli, 2012; Costa et al., 2014; Feragen et al., 2010; Gagnon, 2012; Rahzani et al., 2009) have reported that some people with disfigurement were likely to believe that society did not accept them, even without actual objective experience to support the belief. This impact on the perception of the individuals was reported to be the result of the individual's expectations of society's reaction to the disfigurement, rather than their practical experience in the society.

*Emotional distress.* This category included data segments from Prior and O'Dell (2009), Rahzani et al. (2009), Gagnon (2012) and Costa et al. (2014) considering the negative emotional impact on how society treats people with disfigurement, which included anxiety, feelings of emotional suffering, helplessness, anger, hatred, shame, embarrassment, sadness and feelings of being hopeless.

*Decreased social interaction.* Some studies (Bogart et al., 2012; Bonanno and Esmaeli, 2012; Costa et al., 2014; Masnari et al., 2013; Van den Elzen et al., 2012) reported that, as a result of the mentioned consequences of stigma such as negative perceptions of one's own appearance, perception of reduced societal acceptance and emotional distress, most individuals with disfigurement reported avoiding any social interaction at all for fear of being stigmatised or embarrassed by the way they look. As a result, especially among adolescents and children, they failed to develop the necessary social skills to function in society, which would lead to even bigger social problems and social anxiety that could persist until adulthood.

### *Coping with the stigma*

This theme involved the steps taken by individuals to cope with the negative impacts of social stigma towards people with disfigurement; this could be by the disfigured

individuals themselves or by their families. Moreover, this could also be internal steps of the individual or actual measures taken to come to terms with their current reality. There were three categories that were identified from the systematic literature review, namely, (1) building emotional resilience, (2) acceptance and (3) undergoing plastic surgery.

*Building emotional resilience.* It was reported that there were certain individuals who were naturally resilient against the stigma that they experienced, and they simply did not mind how others treated them as a result of the disfigurement that they had due to disease (Bogart et al., 2012; Masnari et al., 2012; Prior and O'Dell, 2009). The individuals with this resilient personality were simply more confident about themselves and did not let the opinions of others affect them. On the other hand, there were those who needed to develop the resilience necessary to survive the stigma imposed on them. It was reported that some families, especially where the person with disfigurement was still a child, worked together with the victims to jointly acquire the resilience to enable them to cope with the negative impacts of disfigurement.

*Acceptance.* Both Costa et al. (2014) and Hughes et al. (2009) reported that some individuals with acquired disfigurement as a result of a disease were more ready to accept their situation, particularly those with higher levels of spirituality and fatalism. They felt that what happened to them was their fate, and, therefore, they had to accept it. As a result of this disposition, they were more likely to accept their situation. Studies (Costa et al., 2014; Gagnon, 2012) reported that the solution for several individuals was to hide the disfigurement to avoid eliciting negative reactions from other people. In many cases, a permanent solution to hide the disfigurement was to undergo plastic surgery to lessen the severity of the appearance of the disfigurement.

### *Positive social influences to coping*

This theme included the different sources of social support that had been reported to help individuals with disfigurement to cope with their situation and be more resilient to the negative impacts of having a visible difference as a result of a disease. This theme was made up of four categories, namely, (1) strong friendships, (2) family, (3) community and (4) professional help.

**Strong friendship.** Positive social experiences brought about by close friendships had been reported by Roberts and Shute (2011), Bonanno and Esmali (2012) and Feragen et al. (2010), showing that strong friendship helped individuals, especially younger ones, with visible disfigurement to cope with the challenges of their situation. Strong friendships also provided affected individuals with social interactions that helped them become more resilient and accepting of themselves and their appearance. Friends were also reported to help in easing feelings of anxiety and helplessness by providing positive regard and trusting relationships.

**Family.** Just like strong friendships, familial support, especially parental support for younger individuals with disfigurement, offered unconditional social acceptance that helped the individual with disfigurement to feel more comfortable about their appearance. It had been reported by two studies, Prior and O'Dell (2009) and Gagnon (2012), that families with a strong relationship helped each other to be less susceptible to the negative stigma of society.

**Community.** Several studies (Bonanno and Esmali, 2012; Costa et al., 2014; Masnari et al., 2013; Prior and O'Dell, 2009; Roberts and Shute, 2011; Van den Elzen et al., 2012) outlined the importance of providing community awareness programmes to promote wider understanding of the disease that individuals suffered from and how to deal with people with disfigurement.

**Professional help.** This category included therapy from professionals as well as other training

programmes such as social skills development. These activities had been reported by some studies (Costa et al., 2014; Gagnon, 2012; Rahzani et al., 2009; Roberts and Shute, 2011) to provide the individual with disfigurement, a channel wherein he or she was understood and accepted, and it improved social interaction with others.

## Discussion and conclusion

In this article, we examined the existing knowledge about social stigma among people with facial disfigurement. This study added significantly to the literature on social stigma towards individuals with facial disfigurement; specifically, the study presented another index of the stigma affecting individuals with facial disfigurement that was not noticeably captured in the past. In addition, the study was used to show an amended model of interaction between people with disfigurement and society proposed by Bonanno and Choi (2010), which clearly demonstrated the pattern of interaction between facially disfigured individuals and the members of society.

The study further highlighted the manifestations of social stigma, its consequences on the individuals with the disfigurement, how they coped with it and the social influences that helped improve their chances of coping. As Bonanno and Esmali (2012) pointed out, three kinds of reactions were observed that facial disfigurement elicited from other people. The first one was intrusion, in which other people gave unwanted attention to the individual with disfigurement, such as staring, asking too many questions and feeling uncomfortable around the person with disfigurement. The second reaction was in the form of sympathy from other people. However, in the current research, these two forms of reaction have been categorised in one category under the manifestations of stigma theme and labelled as unsolicited attention coming from other people. The third response reported by Bonanno and Esmali (2012) was benign neglect, in which other people paid no attention to the disfigurement of the individual.

However, in this study, this category was also merged with actual neglect or avoidance, which was sometimes viewed as something negative by individuals with a disfigurement. The reason this category had recorded mixed reactions among the respondents of the previous studies is that some individuals had a personal preference for not getting any attention, while others preferred not to be ignored. Similarly, in this study was also recorded the existence of another category that manifested stigma against people with disfigurement, which was actual hurtful behaviour such as bullying, teasing and discrimination (Bogart et al., 2012; Stone and Wright, 2013).

This may be the result of the negative associations related to facial disfigurement, which was the fourth category in the manifestations of stigma theme. The findings of this study, therefore, showed that it would be inappropriate to suggest that benign neglect did not attract stigma, and that neglect should also be seen as a manifestation of stigma, thereby expanding the horizon on manifestations of stigma (Bonanno and Choi, 2010).

In fact, resilience was one of the most common categories observed in the literature. However, while the support of family and friends could influence resilience, it has been shown in much of the literature that the intrinsic motivation of people with disfigurement was strong to continue with their daily living, despite having the disfigurement as a result of the disease. Some codes that are related to intrinsic motivation included gratefulness, acceptance, coping, positivity, self-confidence and hardiness, among others. These were some of the intrinsic qualities of the participants with visible disfigurement, who were more likely to react more positively to the manifestations of stigma in society and less likely to develop depressive symptoms (Feragen et al., 2010).

Hughes et al. (2009) argued that it is a mental evolutionary process to associate disfigurement with a contagious disease. Other people associated disfigurement with negative personalities and physical incapacities, also leading them to act negatively towards the people with disfigurement (Bogart et al., 2012).

These associations may have included lack of communication and interaction skills, and being less intellectual and less capable. These negative mental associations and assumptions were often due to the lack of knowledge and understanding about the causes of disfigurement. It was these mental associations that often led to avoidance behaviour and negative harmful behaviour, such as bullying and teasing. It also led to discrimination, which could have detrimental effects on a person's career and source of income (Stone and Wright, 2013). However, as we explained earlier, it should not be ignored that for certain patients with disfigurement, too much sympathy and curiosity from other people resulted in unwanted attention, thereby creating a sense of stigma.

Another manifestation of social stigma was unsolicited attention, which included pity and sympathy towards the individual with disfigurement (Bogart et al., 2012; Hughes et al., 2009; Masnari et al., 2012). Although most people could mean well in their behaviour, some people with disfigurement, especially those who were trying to avoid getting noticed by avoiding social interaction, would not feel comfortable with such displays of sympathy. Thus, this was still viewed negatively by most individuals with disfigurement (Bogart et al., 2012). It has been reported that, in most instances, individuals with disfigurement were more likely to interpret a simple act by other people as something negative that resulted from having a facial disfigurement. As a consequence, these people tended to believe that the extent of social stigma was a lot bigger than it is understood. Similarly, it could be argued that stigma is not only imposed by society towards individuals with disfigurement as a result of a disease; rather, a significant part of it is imposed by the individuals on themselves as a result of their negative expectations of the society and negative perception of their physical appearance.

Our results reveal that one of the strongest predictors of a person with disfigurement being unhappy or depressed with the way society treated him or her was his or her own

satisfaction with their physical appearance, as confirmed by other studies (Masnari et al., 2013; Srour et al., 2008). It could be inferred that the way a person with disfigurement experienced the world and the society was profoundly influenced by their perception of oneself, and the way they may give meaning to the reality and experiences with interaction with other people was very subjective and based on individual differences of self-perception. Furthermore, this means that the extent to which social stigma was felt or experienced by individuals with disfigurement might depend on the subjective beliefs of the individual, as to how discriminating or stigmatising the society was. This belief regarding the society was also just a manifestation of how satisfied or confident they were in their appearance, even with the disfigurement. Thus, how they felt the stigma was also subjective, depending on how the individual made sense of or interpreted the behaviour of others towards them. Adolescents were also very prone to this problem due to their heightened self-consciousness during this stage. They were more likely to focus on how their peers perceived them and on being accepted. Thus, having a sudden facial disfigurement due to a disease would be very damaging to an adolescent, who would be most vulnerable to bullying and teasing from peers who were not aware of or educated about the repercussions of their actions (Bogart et al., 2012).

This study was not without some limitation, and it was acknowledged that this study be evaluated to be certain about the reliability and validity of the study conclusion. Hence, any limitation encountered in the research literature studied could also affect the validity of this study. Our study was not externally funded, and therefore time and resources were severe constraints. Nonetheless, careful consideration was given to selection of the material for the systematic literature review, on the reliability and validity of the methodologies employed.

Despite these limitations, the explanations presented here for establishing knowledge about social stigma among those people living

with facial disfigurement offered interesting insights for further research. The findings emphasised that healthcare and financial assistance in treating the disease might be available in some countries, but the support did not extend to addressing the facial disfigurement that usually resulted. There was a need for primary healthcare providers or policymakers to consider post-operation healthcare as a priority, since the emotional distress from the disease did not usually end after the illness was treated. Indeed, the significant challenge the patients faced after the treatment of the disease was their dwindling self-perception and the negative perception of society towards them because of the facial disfigurement they acquired due to the disease, therefore appropriate help or support, for example, physical therapy, psychological therapy and even plastic surgery for some cases should be provided. We further suggested that a two-tiered approach may be needed, namely, to educate the general public about facial disfigurement and to equip affected individuals with social skills and appropriate support at the community levels. Finally, in addition to the use of appropriate support, we strongly believe that the results of this study can not only prove beneficial to health and social care professionals in making informed decisions while dealing with individual cases of those affected but can also help in developing and implementing strategies to curtail the stigmatisation imposed on individuals with facial disfigurement, thus improving the quality of life of those affected groups.

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