



Social inclusion, care and belonging of children with spina bifida: perspectives from Uganda

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Sociale inclusie, zorg, betrokken- en geborgenheid van kinderen met spina bifida:
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To all the children with spina bifida who participated in this study



Foreword

In May 2007 my employer (AVSI) introduced me to Pierre Mertens, president of the International Federation of Spina Bifida and Hydrocephalus (IF) at the time, who asked me to attend a session at the International Spina Bifida and Hydrocephalus Conference in Kampala, Uganda. I had met children with spina bifida and hydrocephalus in northern Uganda, where AVSI had set up a referral system for children requiring neurosurgery from the Gulu orthopaedic workshop and rehabilitation centre to CURE hospital in Mbale. A partnership between AVSI, my employer, and IF then started, and AVSI hosted the IF Uganda office. I was asked to supervise the staff in the IF Uganda office and implementation of the program in Uganda.

My meetings with children with disabilities and their families spurred my interest in carrying out research and furthering my professional development as a psychologist, anthropologist, and development program advisor. With the help of colleagues and friends I explored the possibilities of pursuing a PhD in disability studies whilst living and working in Uganda. This thesis is the results of a 6 year journey, in which I learned a lot about research, academic writing, politics, belonging, resilience and hope.

I met, spoke, and played with children and families who often struggled economically, and socially. Some were surprised I wanted to speak to them. Through my work and research, there were a few children I met regularly. Ruth was one of them. She was born with spina bifida and developed hydrocephalus shortly after birth. Her father left her mother after she had given birth to her. Ruth, her mother, and aunt live in a room of 3x4 meters in a slum area of Kampala and have a monthly household income of less than a hundred euros. Ruth's mother is a teacher; every day she carries Ruth on her back to school. Ruth enjoys school, and she loves singing.

The traditional greeting of welcome in Ruth's home and many others I have entered in the central region in the past few years is '*karibu, nsanyuse okubalaba*'. There are similar greetings in the languages of the other regions, and all mean: 'Welcome, I am happy to see you.' When leaving one does not say goodbye, but says '*nkomawo*': I am coming back.

Uganda's languages and context show how we are because of others, and how interdependency can give us a different sense of belonging, not related to autonomy and rights but through being part of and practising humanity.

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Chapter 1 Introduction



1.1 African Childhood Disability Studies

1.1.1 Historical perspectives of disability in sub-Saharan Africa

Perceptions of impairment and disability are social phenomena subjected to “substantive temporal, cultural and situational variation” (Ingstad & Whyte, 1995). To understand disability in a particular society attention needs to be paid to geographical, historical and cultural contexts, and social, economic and cultural disparities (Albrecht, DeVlieger, & Van Hove, 2008). E.g. in cultures with an interdependent focus, acceptance in the group are a significant factor contributing to quality of life, more than in independent or individualistic oriented cultures (Schalock, 1997). Belonging to a family is a central and key concept in African societies (Bigombe & Khadiagala, 2003; Chataika & McKenzie, 2013; Guyer, 1981; Malinowski, 1929).

Social-anthropological and historical studies describing cultural descriptions and belief systems of disability in Africa show a complex range of cultural concepts e.g. the child being “cursed”, “bringing misfortune” or being a “gift” (Braathen & Ingstad, 2006; Franzen, 1990; Miles, 2002; Wright, 1960). These cultural concepts derive from beliefs about the causes of disability in East Africa which include animistic beliefs in which disabilities are considered punishments for bad deeds or the result of witchcraft exercised by other people (Hartley, Ojwang, Baguwemu, Ddamulira, & Chavuta, 2005; Ogechi & Ruto, 2002), religious fatalism beliefs in which disability results as an act of God’s will (DeVlieger, 1998; Kisanji, 1995), and medical determinist beliefs in which modern medicine are believed to cause disabilities (Harknett, 1996). Multiple categories of beliefs about cause, such as suggesting a medical explanation in addition to traditional animism are not uncommon (Ingstad & Whyte, 1995).

Most East African languages do not include words that allow ‘disabled’ to be directly translated from English, but define disabilities relating to the body part that does not function normally (Ogechi & Ruto, 2002). The body and words about the body are prominent because a healthy body is important to daily life in local communities (Ingstad & Whyte, 1995). Attitudes towards people with disabilities in East Africa have both positive and negative aspects and are often linked to beliefs about the causes of disability (Ingstad, 1995; Miles, 2006). In many proverbs, kindness towards individuals with disabilities and punishment for negative attitudes or actions are evoked (DeVlieger, 1994). Whilst families and communities care for children with disabilities, children and family members appear to be less accepted into the community if the individual cannot contribute economically or is unable to take part in culturally important rituals (Ingstad & Whyte, 1995).

1.2 Theoretical framework for disability studies in sub-Saharan Africa

As a theoretical framework this study uses Reindal’s social relational model of disability. We look at the child in the context of the biological and physiological realities of his or her body, and personal and social limitations as experienced due to reduced functioning on one side, and material and cultural social barriers and the experience of social barriers that are discriminating and restraining on the other hand (Reindal, 2008). This model is significant in looking at children with spina bifida, who might have been ‘repaired’ physically through available treatment, but are still living with the consequences of their illness, physically, mentally, and socially. Their disability is often approached

medically, with an attempt to cure their impairment or restore 'normal' bodily functioning. The social model of disability which we follow in this study aims at full integration of persons with disabilities in society (DeVlieger, 1998) taking into account the bodily experiences (Thomas, 2004).

Social inclusion, care, and belonging are the key concepts of this study. The dominant discourse on social inclusion (or exclusion) in sub-Saharan Africa is focused on poverty, marginalisation, vulnerability and sustainable development (WHO, 2008). We look at social inclusion as the interaction between interpersonal relationships and community participation, considering individual, interpersonal, organizational, community, and socio-political factors (Simplican et al, 2014). Social inclusion is linked to the sense of agency and capacity to choose whether to participate or not (Baumgartner & Burns, 2013). It is not about social integration, as Brown (2002) described: "It is absolutely not our job to fit into mainstream society. Rather [...] mainstream society needs to figure out how we fit in, but how we can be of benefit exactly the way we are."

In this study we approach care in terms of 'being concerned about', 'looked after', and 'protected' as societal responses which go beyond medical and rehabilitative care (WHO, 2001). Rather than looking at achieving independence, we focus on interdependence in care: "Inclusion entails recognition of our universal oneness and interdependence. Inclusion is recognizing that we are one even though we are not the same" (Asante, 2002).

To belong is "to feel attached, to feel valued, and to have a sense of insidership and proximity to 'majority' people, activities, networks and spaces" (Hall, 2010). The sense of belonging is key to and interrelated with interpersonal relationships and community participation (Simplican et al, 2014).

In sub-Saharan Africa Disability Studies have largely focused on adults with and caregivers of children with disabilities, and have primarily been conducted in South Africa. Children's narratives are absent in most disability literature (Curran & Runswick-Cole, 2014). Like in other low resource settings, these studies have argued for valuing diverse understandings and a different discourse (Grech, 2009; Meekosha, 2011; Seligman & Darling, 2009; Whyte, 1995). Chataika and McKenzie (2013) suggest African Childhood Disability Studies look at family, cultural conceptions of disability, poverty, and the notion of '*ubuntu*' ("I am because we are" or "humanity to others") as central concepts (Chataika & McKenzie, 2013; Oppenheim, 2010).

As persons with disabilities experience the world with their impaired body (Campbell, 2009; Hughes, 2007), there is need to understand the impaired body and the implications of the permanent presence of the impairment (Hughes & Paterson, 1997). In the next section an overview of the medical, rehabilitative, and social aspects affecting daily life of children with spina bifida is provided. This is followed by a section with a description of the Ugandan context, looking at history, culture and meaning, relating the implications of the permanent presence of the impairment, political systems and cultural beliefs, taking into account the concept of ontoformativity: "the understanding of social dynamics in bodies as a form of global social embodiment" (Meekosha & Soldatic, 2011).

1.2 Spina bifida

1.2.1 Spina bifida and hydrocephalus

Spina bifida is a Neural Tube Defect (NTD), a congenital disability, whereby the spinal cord and vertebrae do not form completely and the neural tube fails to develop normally (Northrup & Volcik, 2000). The intake of folic acid through supplements and food fortification before and during pregnancy can reduce the incidence of NTDs (Czeizel, 2000; Wilson et al., 2003). Countries fortifying flour with at least folic acid see an average 46% reduction of NTD prevalence (Pachón, Kancherla, Handforth, Tyler, & Bauwens, 2013).

There are three main types of spina bifida: occulta, cystica meningocele and cystica myelomeningocele. Spina bifida occulta is a very mild and common form and very rarely causes disability. Spina bifida meningocele and myelomeningocele have visible signs of a sac or cyst covered by a thin layer of skin. In meningocele the sac contains tissues which cover the spinal cord (meninges) and cerebro-spinal fluid. The nerves are not usually badly damaged and are able to function. In myelomeningocele the cyst does not only contain tissue and cerebro-spinal fluid but also nerves and part of the spinal cord. The spinal cord is damaged or not properly developed. As a result, there is always some paralysis and loss of sensation below the damaged region. The amount of functional impairment depends very much on where the lesion is and the amount of nerve damage involved (IFSBH, 2014). Most children with spina bifida have some degree of paralysis, which affects mobility as well as bowel and bladder control (Northrup & Volcik, 2000) (Verpoorten & Buyse, 2008). Between 70 and 90% of children with spina bifida develop hydrocephalus. In hydrocephalus the natural circulation of Cerebrospinal fluid (CSF) in the brain is obstructed and fluid accumulates (IFSBH, 2014). The excess fluid presses on the brain causing damage to the surrounding tissue. In babies and infants where the skull is still soft, the head enlarges (CURE, 2008).

1.2.2 Prevalence and incidence

The prevalence at birth of spina bifida in Europe, Northern America and Australia has decreased due to the increased use of and fortification of food with folic acid, which can prevent spina bifida, and antenatal screening resulting in early termination of pregnancies with NTDs (Frey & Hauser, 2003). As well mortality associated with the condition has fallen due to improved perinatal and postnatal care (Thompson, 2009). Nevertheless the worldwide incidence of spina bifida is still between 0.17 and 6.39 per 1000 live births (Bowman, Boshnjaku, & McLone, 2009; Kinasha & Manji, 2002; Msamati, Igbigbi, & Chisi, 2000; Shaer, Chescheir, & Schulkin, 2007).

Incidence and prevalence rates in low income countries such as Uganda may be higher due to inadequate folate consumption by pregnant women, lack of or inadequate pre-natal care (Miles, 2002), absence of secondary prevention services (Frey & Hauser, 2003), and higher exposure to environmental risk factors such as dioxins (Safi, Joyeux, & Chalouhi, 2012) and fumonisins intake (Hendricks, 1999; Marasas et al., 2004; Wild & Gong, 2010). No national data are available in Uganda. Warf et al estimate that 1,400 children are born with spina bifida in Uganda annually (Warf, Wright, & Kulkarni, 2011). Sixty-six per cent of the children with spina bifida develop hydrocephalus (Warf & Campbell, 2008).

1.2.3 Surgical and rehabilitative treatment

Most children born with spina bifida need surgery to close the exposed tissues and to prevent infections. Surgical closure of myelomeningocele is usually performed in the early postnatal period. Delay in surgical intervention has been associated with higher incidence of preoperative rupture of the myelomeningocele and neurodevelopmental delay. Some children will need subsequent surgeries to manage problems with the feet, hips, or spine (IFSBH, 2014).

Children with progressive hydrocephalus need placement of a ventriculo-peritoneal (VP) shunt or endoscopic third ventriculostomy (ETV) to drain cerebral spinal fluid and prevent secondary impairments (IFSBH, 2014; Thompson, 2009; Warf, 2005). A shunt is simply a drain that diverts or shunts the accumulated CSF from the obstructed drainage pathways and returns it to the bloodstream (CURE, 2008). Shunts are sensitive to infection and malfunctioning (Hoppe-Hirsch, 1998; Hunt, 1999; Radmanesh, 2009), and are related to epilepsy (Bourgeois, 1999; Kulkarni, 2004). Shunt-dependency is more dangerous when urgent access to neurosurgical care is not available, such as in low income countries. ETV is an alternative primary treatment for hydrocephalus and alternative for malfunctioning and infected shunts. It creates a natural bypass for the CSF (O'Brien, 2005). Warf et al (2005) demonstrated that the majority of children with hydrocephalus can avoid shunt-dependency when treated with ETV (Warf, 2005).

Mobility challenges and incontinence require the use of assistive devices and continence management (Abresch, McDonald, Widman, McGinnis, & Hickey, 2007; Andren & Grimby, 2000; Danielsson et al., 2008; Jansen, Blokland, de Jong, Greving, & Poenaru, 2009). Children with spina bifida benefit from physio- and occupational therapy to prepare for (assisted) sitting, walking, and participation in daily activities. Children with more extensive paralysis will often require a wheelchair, while others may be able to use crutches, braces, or walking frames. Children with spina bifida are prone to pressure sores and need regular skin checks to prevent these (Lindsay, 2014).

Most children with spina bifida have bowel and bladder problems. Clean Intermittent Catheterization (CIC) and bowel management techniques are used to keep the child dry and clean. In CIC a catheter is put into the bladder through the urethra and removed when the bladder is empty. Bowel management techniques include the use of enemas or suppositories, or high bowel washout (IFSBH, 2014). If CIC is started early children with spina bifida can be dry by school-going age (Jong et al, 2008; Verpoorten & Buyse, 2008).

1.2.4 Cognitive and social functioning

The majority of children with spina bifida and hydrocephalus have intelligence in the normal range, tending to be in the low normal range. Research in high income countries suggests that children and adolescents with spina bifida are at risk for difficulties in attention (not sustained attention or ADHD) (Burmeister et al, 2005; Fletcher et al 2005; Rose & Holmbeck, 2007; Swartwout et al, 2008), short-term memory (Vacha & Adams, 2003), prospective memory, immediate and delayed episodic memory (Dennis et al, 2007), nonverbal learning disabilities (Rist et al, 2007), language comprehension and discourse (Huber et al, 2005; Barnes et al, 2007; Pike, Swank, Taylor, Landry, & Barnes, 2013), and approximate and standardized arithmetic measures (Raghubar et al., 2015). Dennis and Barnes (2010) reviewed the cognitive phenotype of spina bifida and emphasize the need

to make a distinction between the functional assets and deficits in timing, attention, movement, perception, language, literacy, and numeracy of children with spina bifida (Dennis, 2010).

Persons with spina bifida often face social skills and inclusion challenges (Wyszynski, 2006). Regardless whether persons with spina bifida can ambulate or use wheelchairs, low social integration and economic self-sufficiency scores are prevalent (Dicianno et al, 2009). In Kenya social isolation was high in children with spina bifida (van 't Veer et al, 2008).

1.2.5 Family, care, and parental stress

In a review of the social, psychological, and economic burden of having a child with spina bifida, Rofail et al found that activities of daily living, work, parental responsibilities, confidence, feelings and emotions, mental health, stress, psychological adjustment, relationships, social support, and finances were impacted in caregivers of children with spina bifida (Rofail, Maguire, Kissner, Colligs, & Abetz-Webb, 2013). Families of children with neuro-disabilities including spina bifida in Kenya (van't Veer et al., 2008), Malawi (Paget, Mallewa, Chinguo, Mahebere-Chirambo, & Gladstone, 2015), and South Africa (Coomer, 2013), struggle with the financial implications and social barriers towards care and support for their child. In the absence or very limited availability of health care and social services for children with disabilities, families in sub Saharan Africa are often their main source of care and protection for children (Guyer, 1981; Miles, 2002).

Research on family functioning and psychosocial adjustment of families of children with spina bifida in high income countries support a resilience–disruption view of family functioning, whereby the presence of a child with spina bifida disrupts normative family functioning at first, but after a period of time families adapt and exhibit considerable resilience (Holmbeck et al., 1997; Vermaes, Gerris, & Janssens, 2007). Stress levels of parents of children with spina bifida are higher than parents of typically developing children (Holmbeck et al., 1997; Kanaheswari, Razak, Chandran, & Ong, 2011; Ong, Norshireen, & Chandran, 2011; Wallander, Pitt, & Mellins, 1990). Caregivers of children with disabilities in both high and low income countries often felt that they do not have sufficient time to cope with household tasks, and feel isolated (Gona, Mung'ala-Odera, Newton, & Hartley, 2011; Hartley, Ojwang, Baguwema, Ddamulira, & Chavuta, 2004). Adjustment and siblings relations in families with a child with spina bifida in high income countries argue for supporting positive siblings interactions, as satisfaction with family functioning was associated with warmth and conflict in the sibling relationship (Bellin, Bentley, & Sawin, 2009; Bellin & Rice, 2009).

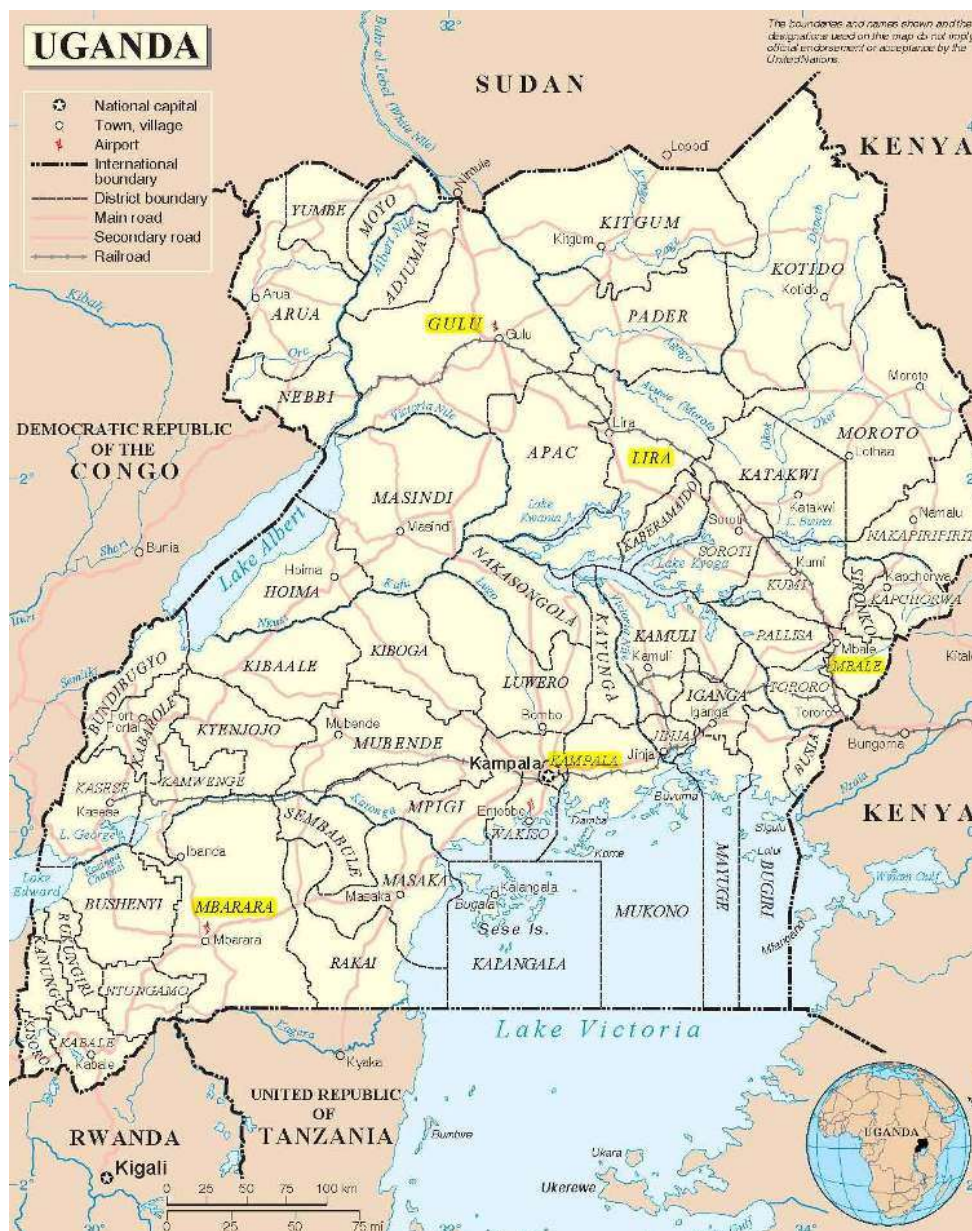
1.3 Study setting

1.3.1 Study area

This study was conducted in Uganda, a land locked country of 241,039 square kilometers and a population of 34.8 million of which 56.7% is under 18 years of age. Uganda borders Kenya to the east, Tanzania to the south, Rwanda to the southwest, the Democratic Republic of Congo to the west, and South Sudan to the north. The country is administratively divided into 112 districts. The male to female sex ratio is 95:100 and the average household size is 6.2 (UBOS, 2014). The economy is predominantly agricultural, with the majority of the population dependent on subsistence farming and light agro-based industries (UDHS 2011). Uganda has 41 tribes and over 17 local languages. The

official language is English. The largest tribe are the Baganda with 17.3% followed by the Banyankore representing 7.8% of the population (UBOS, 2002). Participant recruitment was conducted in the northern, eastern, central, and western regions of Uganda. Each region is the home to different tribes, with their own language and cultural beliefs and practices. Kampala, the capital city in the central region, has a multi-ethnic population. The central, eastern, and western tribes have in common that they are Bantu tribes, while the northern tribes are Nilotic. In the northern region, the majority of the people were displaced during the 22-year conflict between the Government of Uganda and the Lord's Resistance Army (Annan, 2008). Since 2006, relative peace allowed people to return home and start rebuilding their communities (Annan, 2008). The other regions have enjoyed relative peace since 1986, apart from occasional border conflicts with neighbouring countries.

Map 1. Study sites Gulu, Kampala, Lira, Mbale, Mbarara and Kampala in Uganda



1.3.2 Disability in Uganda

The Uganda Bureau of Statistics estimates 12.5% of the population has a disability, which translates into 2.5 million children living with some form of disability in Uganda (UBOS, 2014).

In most languages spoken in Uganda there is no single word that translates into the English word 'disabled', however all descriptions combine the notion of physical limitation and powerlessness (Lwanga-Ntale, 2003). The definition of disability in Ugandan laws is not harmonized. The Persons with Disabilities (PWD) Act 2006 defines disability as "a substantial functional limitation of daily life activities caused by physical, mental or sensory impairment and environmental barriers resulting in limited participation". By recognizing that disability is the result of the interaction between impairment and external barriers, the PWD Act aligns the legal definition of disability in the Ugandan law to that enshrined in the Convention on the Rights of Persons with Disabilities (CRPD), which Uganda ratified in 2008 (NUDIPU, 2013) (Enable, 2015). However unlike the CRPD, the PWD Act requires that disability be substantive, which has implications for disability rights at a practical level (Ojok & Wormnæs, 2013).

National policies relevant to disability and inclusion in Uganda include the Uganda National Institute of Special Education Act (1995), the Council for Children Act (1996), Council for Disability Act (2003), the Orphans and Vulnerable Children Policy (2004), the National Council for Disability Act and Policy on Disability (2006), and the Equal Opportunities Act (2007). Uganda has a strong Disability Movement and national Disabled Persons Organizations, such as the National Union of Disabled Persons of Uganda (NUDIPU), and the Uganda Society for Disabled Children (USDC). Uganda adopted Community Based Rehabilitation as a health service strategy for reaching more persons with disabilities in 1990 (NUDIPU, 2007). A special needs education department was created in the Ministry of Education in 1973 (USDC, 2003). Persons with disabilities are represented in the Uganda Constituent Assembly, local governance structure, and in parliament (Government of Uganda, 2006). They are included in the development of government plans, e.g. the Ugandan Government National Development Plan 2010/11-2014/5, which integrated vulnerable groups including Persons with Disabilities in her development planning and implementation. This study specifically relates to the National Development Plan's 4th objective "increasing access to quality social services"; and sections 565 higher education, objective 1, strategy 5 "promote applied research and publications", and objective 2, strategy 1 "provide integrated promotive, preventative, curative, and rehabilitative services that have been proven effective" (Government of Uganda, 2010).

Despite the laws and policies being in place, implementation remains lacking because of limited budget resource allocation and limited attention paid to children with disabilities. Community based rehabilitation services are implemented by rehabilitation centers funded by non- governmental organizations and private health facilities. Discriminatory attitudes and behaviour, a gap in implementation of the regulatory framework, lack of coordination between government and civil society, and a fragmented programmatic approach together challenge the implementation of the CRPD (UNICEF, 2014).

1.3.3 Health and rehabilitative services

There are wide disparities in health status across Uganda, closely linked to underlying socio-economic, gender and geographical disparities. The life expectancy of Ugandans at birth is 57 for males and 61 years for females (WHO, 2013). The infant mortality rate stands at 54 per 1000 live births, and maternal mortality rate is 438 per 100,000 live births (UBOS, 2014). Maternal and perinatal conditions contribute to high mortality. The total fertility rate is approximately 6.2 births per woman (WHO, 2013). Uganda has a national Health Policy 2010-2020 and a Health Sector Strategic and Investment Plan 2010/11-2014/15 to guide the strategic focus for the health sector (Government of Uganda, 2011). The resources available for health are less than required to deliver the Uganda National Minimum Health Care Package. Households continue to carry a heavy burden with a high out of pocket expenditure on health. Gaps in human resources for health, medicine and supplies stock outs, and poor planning and management pose a challenge for effective service delivery (WHO, 2013).

National health care services are organized in national, referral, and district hospitals, and higher and lower level health centers providing health care within the various districts. Specialized health and rehabilitative services for children with spina bifida are limited. At the time of this study the initial surgery (closure of the spine) for children with spina bifida was only available in two public government funded hospitals (Mulago National Referral Hospital in Kampala and Mbarara Regional Referral Hospital), and one private specialized neuro-paediatric hospital (CURE Children's Hospital) in Mbale, eastern Uganda. Placement of VP-shunts for children with progressive hydrocephalus was offered at Mulago and CURE hospitals, whilst ETV was only available at CURE hospital. All hospitals offered general physiotherapy services. In north, west, and central Uganda three rehabilitation centers, funded by international donors and charities offered specialized occupational therapy, physiotherapy, continence management, and social support services for children with spina bifida and their families.

Traditional health systems co-exist with the biomedical national health system. The traditional system involves herbalists, spirit diviners, and traditional birth attendants. Earlier studies on AIDS education, counselling, and treatment of mental illness have suggested collaboration and involvement of traditional healers in biomedical care (King et al, 1997; Ovuga et al, 1999; Abbo et al, 2009). The biomedical services for children with spina bifida outlined above did not collaborate with traditional healers. Occupational therapists of the rehabilitation centers did mention they had approached traditional healers to refer children with spina bifida for surgery and rehabilitative care.

1.3.4 Inclusive education

In 1997 Uganda endorsed the Universal Primary Education (UPE) policy, which aims to provide free primary education for all children in Uganda (Government of Uganda, 1997). In line with the Education For All (EFA) initiative the target group of children with disabilities expanded to including orphans, traumatised children, HIV positive children and others with special needs. A new Basic Education Policy for Disadvantaged Children was endorsed in 2002, which increased the demand for training of staff at all levels (DANIDA, 2005). The Ministry of Education has an inclusive education policy in draft for inclusion of children with special needs in primary schools (Nyende, 2012). Despite

these policies being in place implementation of Universal Primary Education (UPE) and inclusive education remains limited (Ejuu, 2016). According to a UNICEF report only 9% of children with disabilities in Uganda attended school and only 6% completed primary school and proceeded with secondary education between 2009 to 2011 (UNICEF, 2014).

1.3.5 Socio-economic factors

Persons with disabilities typically live in poorer than average households, and have lower educational attainment (Emmett, 2006; Filmer, 2008; Lwanga-Ntale, 2003). The average monthly income of households in Uganda is 156 US dollars (UBOS, 2014). Social services in forms of grants or allowances for families with children with disabilities are limited. Disabled Persons Organisations can apply for economic development grants at district level, and some have started village saving and loan associations to improve their household income. Unfortunately parent support groups are not considered as Disabled Persons Organisations (DPOs) and are not eligible for the few grants available. Some families have benefited from national programs such as the National Agricultural Advisory Services Programme (NAADS) program to improve their socio-economic status (Government of Uganda, 2014). However individual funding to make adjustments in the living and school environments to improve inclusion is not available in the country.

1.4 Study Questions

The objective of this study is to present a situation analysis on inclusion of children with spina bifida in Uganda, evaluate the impact and relevance of rehabilitative and social services provided to the children and their parents, and provide recommendations for future interventions and research. Originally the study aimed to study quality of life in relation to neurosurgery and rehabilitative services. However the concept of quality of life did not fit the cultural context and terminology; quality of life assessment scales - even if adjusted - were not well understood nor described the actual reality of the children in our study population. The number of children who did complete quality of life scales was too small to conduct meaningful comparison analysis. Through observations and interviews social inclusion, care and belonging were identified as key concepts of the children's lives.

The specific questions the studies aimed to answer are:

- a) What is the knowledge, attitude about, and perception of children with spina bifida in Uganda?
- b) Which health and rehabilitative care services do children with spina bifida and their parents access?
- c) Which factors promote social inclusion and belonging of children with spina bifida?
- d) What is the role of the family in social inclusion, care, and belonging of children with spina bifida?

1.5 Methods

1.5.1 Ethical approval

Ethical approval and research clearance for this study were obtained from Ghent University, Belgium, the Uganda Virus Research Institute (UVRI), and the Uganda National Council for Science and Technology (UNCST). Informed consent was obtained from all parents and teachers, and assent from children and siblings of 8 years and above where possible. Consent forms were translated into the local languages and discussed and agreed with the participants. Annual reports were submitted to, and approvals for continuation of the study received from UVRI and UNCST between 2011 and 2015.

1.5.2 Study population

With permission from CURE children's hospital of Uganda, the researcher reviewed the records of 3,192 children with spina bifida and/or hydrocephalus treated in CURE between 2000 and 2009 in Uganda. Initially the plan was to select a random sample from this group. The inclusion criteria were a) presence of spina bifida (myelomeningocele type) with or without hydrocephalus b) minimum age at enrolment 4 years, maximum 14 years (typical pre-primary and primary school age) c) medical records and contact details or location description of the home of the child are available at partner organizations d) child and parent live in one of the 16 selected districts and speak one of the following languages: English, Luganda, Ruyankole, Acholi, or Langi. Selection based on the impairment (a) and age criteria (b) resulted into identification of 2,005 files of children with spina bifida aged 4 to 14 years. Of these 359 lived in one of the 16 selected districts, and spoke the selected languages (d). Only 32 of them had phone contacts, and 98 had a location description of their home (c). It was impossible to trace children without location details as there is no national registry or mapping system in place. Efforts were made to explore recruitment of children through Mulago national referral hospital in Kampala, but no database or information was available on children who accessed care for spina bifida and hydrocephalus. Therefore the initial sample selection method was abandoned, and the following recruitment method was applied.

In Mbarara, Kampala, and Mbale non-governmental organizations (NGO) partners of CURE where CURE holds (bi-) monthly clinics were requested to list the children in their programs meeting the inclusion criteria, and inform the children and parents about the study and invite them to participate.

In Gulu and Lira where no follow up system or registry of the children was in place at the time of this study, but CURE did carry out outreach clinics at the hospitals, radio announcements were aired to inform parents about the study, and invited parents with children with spina bifida aged 4 to 14 to attend the next CURE outreach clinic.

Of the 98 children who had village and sub-county indications on their CURE file, 26 were traced using local and community structures: 12 in the central region, 10 in the eastern, and 2 in the western region). In Gulu and Lira none of the children with CURE file records could be traced, as people were displaced in camps at the time of registration at CURE, but had returned to their villages of origin after the conflict ended.

In total parents of 243 children with spina bifida and hydrocephalus expressed interest in participating. Parents were informed about the study and participation criteria. After establishing eligibility and interest to participate from the child and parent, consent (and assent where applicable) was obtained. In total 178 children and parents, and 35 siblings participated. Depending on availability and time assessments and interviews were carried out immediately or an appointment was made. Contact details and directions to the home locations of participants were registered to allow for follow up visits and interviews in the home setting.

During the study four children of the study population died. All had spina bifida and hydrocephalus and had a shunt placed early in life. One child died from malaria. The other three parents reported symptoms of shunt failure, e.g. convulsions, high fever, and headaches. In two cases parents did not seek medical treatment early enough, and informed staff of the rehabilitation centres and researchers about the symptoms after the children had died. In one case the parent did seek assistance and was referred to CURE hospital immediately, but unfortunately the child died on the way to the hospital.

Random samples of pregnant women, and health workers, and purposeful selection of teachers, and policy makers were carried out for the health services, education, and prevention study parts. In total 394 pregnant women attending ANC, 35 health workers, 30 teachers, and 9 policy makers were interviewed.

1.5.3 Study methods

A knowledge, attitude, and perception analysis (KAP) was carried out to understand the specific context and cultural interpretation of, and health seeking behaviour, and perception of services provided for children with spina bifida. Baseline demographic characteristics were collected through interviews with children and parents, and file review, and included sex, age, tribe, religion, number of children in the family, household size, socio-economic status, education level, etc.

In total 178 semi-structured interviews, 248 observations, and 6 focus groups discussions were held with children, parents, local, traditional, and religious leaders, staff of hospital and rehabilitation centres, teachers and community members. The interviews and focus group discussions included questions on the knowledge of causes, presentation, and treatment for spina bifida (and hydrocephalus), as well as the attitude and perception of society towards children with disabilities including spina bifida, and related practices. Descriptive case studies using photos and video material were made of children and parents participating in the study to illustrate their sense of belonging, and (barriers to) inclusion.

To understand which services the children and their parents received, medical, rehabilitation, and social files review was carried out for each of the children at CURE hospital and partner organizations. Structured interviews with 12 neurosurgeons, health workers, physiotherapists, occupational therapists, and social workers were held to understand the type of services provided to each child. In total 394 pregnant women, and 35 antenatal care workers were interviewed to understand folic acid intake.

To measure motor function, observations of gross motor skills were registered, and the Daily tasks subscale of the Vineland Adaptive Behavioural Skills (VABS) was administered (Sparrow, Balla, Cicchetti, & Doll, 1984). Continence information was collected of all children.

To measure cognitive functioning appropriate assessment batteries for children with spina bifida were selected through a literature review taking cultural aspects, prior use, validation, and translations in Luganda into consideration. A cognitive test battery with selected sub-tests relevant to our study population was compiled and administered to 133 children with spina bifida and 35 siblings.

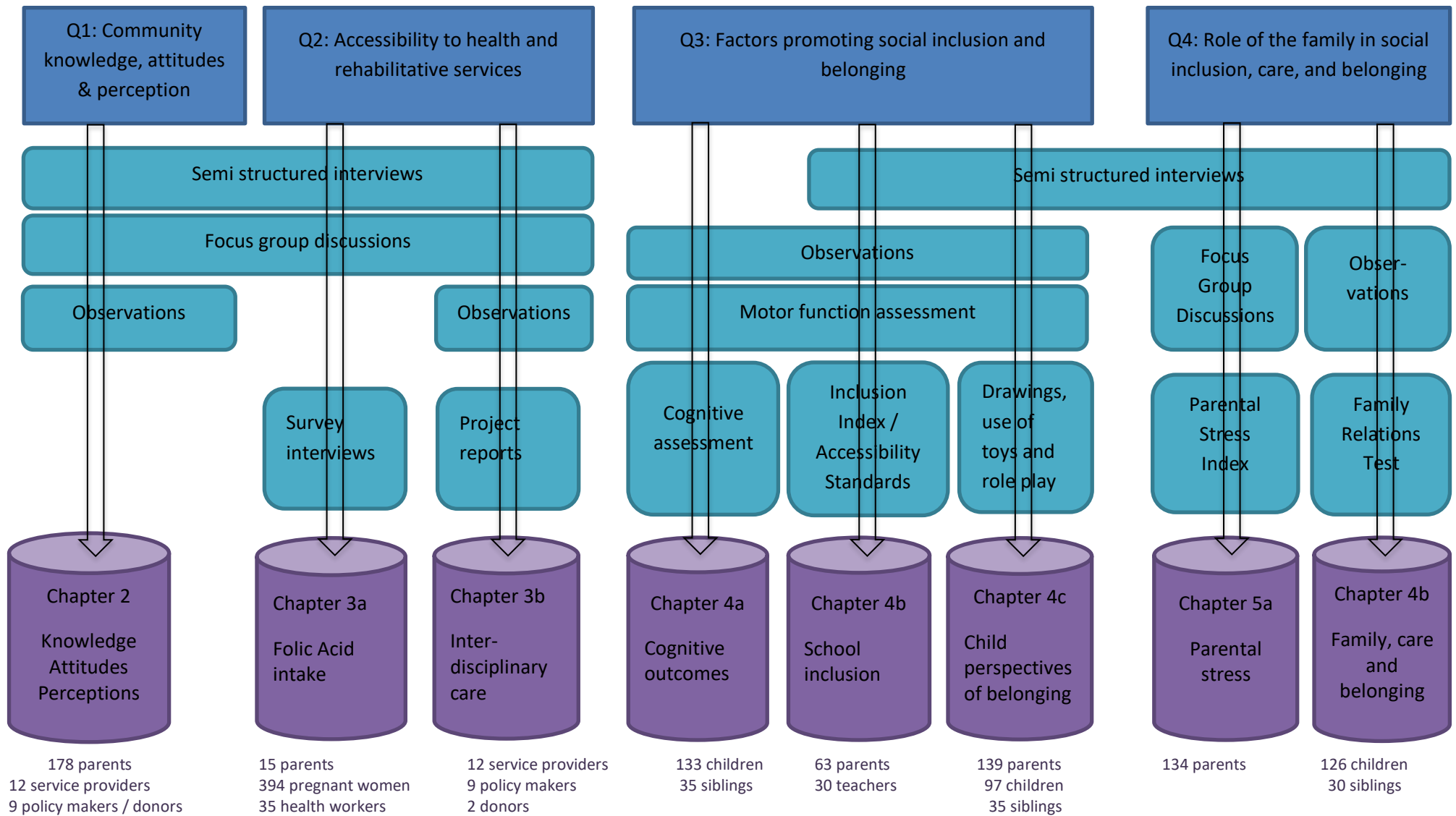
The Index for Inclusion (Booth & Ainscow, 2002) was used as a guideline to measure the inclusion levels and attitudes of school administrations and teachers in 30 schools. Assessments of the school environment included interviews and observations of 30 children, 30 teachers, and 63 parents. To assess accessibility the Ugandan National Accessibility Standards were used (UNAPD & Government of Uganda, 2010).

To investigate parent child interaction, parental distress, and perceived family relationships, the Parenting Stress Index short form (PSI-SF) was administered to 134 parents. In total 4 focus group discussions with parents were organized to collect information on causes and types of parental distress, type and perceived level of support from relatives, friends, community members, and organizations.

To understand belonging in the family, and the role of the family in care the Family Relations Test (FRT) was adapted used in semi-structured interviews with children with spina bifida and their siblings. In total 126 children with spina bifida and 30 siblings completed the FRT.

All questions and assessment scales were translated in the local languages, and pre-tested before administering to the whole study population. In total three research assistants to carry out assessments and interviews in 5 locations were recruited. They were assisted by one translator in the northern, and two translators in the western region. The research assistants spoke at least one of the local languages spoken by the majority of the selected children in the region where they conducted interviews. The research assistants were trained on the study protocol, informed consent process, confidentiality, and administration of the study tools, and referral of children to other services. They received clearance for participation in the study by the President's Office as per national study protocol guidelines. The staff of CURE Children's hospital of Uganda in Mbale, Katalamwa Cheshire Home in Kampala, Gulu Regional Rehabilitation Centre/AVSI Foundation in Gulu, Lira Hospital in Lira, and Organised Useful Rehabilitation Services for PWD in Mbarara received a briefing about the study.

1.5.4 Study Design



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Chapter 2 Knowledge, Attitudes, and Practices



2.1 Community Knowledge, Beliefs, Attitudes, and Practices Towards Children with Spina Bifida and Hydrocephalus in Uganda¹

Abstract

This article describes findings of a qualitative study on knowledge, beliefs, attitudes, and practices towards children with spina bifida and hydrocephalus in four regions of Uganda.

Focus group discussions and semi-structured interviews were held with parents of children with spina bifida and hydrocephalus, policy makers, and service providers.

Our findings describe how negative knowledge, beliefs, attitudes, and practices create barriers to treatment and inclusion of children with spina bifida and hydrocephalus and their parents in Uganda. It shows how knowledge, beliefs, attitudes, and practices evolve over time, resemble and differ in the various regions, and become more conducive towards accessing treatment and achieving inclusion.

Sensitisation and early intervention including parents and service providers in dissemination of knowledge, and rehabilitative care to set the trend for positive change and support, as well as longitudinal studies of children with spina bifida and hydrocephalus and their parents are recommended.

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Introduction

Interpretations of Disability in Sub-Saharan Africa

To understand disability in a particular society one needs to pay attention to geographical, historical and cultural contexts, and social, economic and cultural disparities in a society (Albrecht, Seelman, & Bury, 2001). Anthropological and social studies have provided explanatory models through participant observation, and interviews with key informants, including community members and local leaders on attitudes and perceptions of persons with disabilities (PWD) in various African countries. More quantitative survey-based studies on knowledge, attitudes, and practices (KAP) are common in public health research in Sub-Saharan Africa. Some of the HIV-related KAP research has specifically focused on PWD (Hanass-Hancock, 2009; Lefevre-Chaponniere, 2010).

In most African societies, disability is associated with supernatural forces (Munyi, 2012). However, large differences exist between societies in which PWD are seen as bringers of good luck (Wright, 1960) and in others evil (Franzen, 1990). In the Democratic Republic of Congo, persons are categorised in both human and non-human; children with disabilities belong to the category of 'non-human', an area believed to bring misfortune (Devlieger, 1998). Ingstad and Whyte (1995) explain how perceptions of 'impairment' and 'normality' are social phenomena that are not easily defined, and are subject to substantive temporal, cultural and situational variation (Ingstad & Whyte, 1995). Differences are not tight by country boundaries, but rather by different interpretations of cultures within these countries, and the type of impairment (Munyi, 2012; (Mezue & Eze, 1992). Visible deformities seem to be more often associated with negative attitudes and practices than non-visible ones (Adeloye, 1971).

Spina Bifida and Hydrocephalus

Spina bifida is a Neural Tube Defect (NTD), a congenital disability, whereby the spinal cord and vertebrae do not form completely and the neural tube fails to develop normally. This usually causes some degree of paralysis, which affects mobility as well as bowel and bladder control (Northrup & Volcik, 2000). Depending on the height of the lesion, persons with spina bifida are either able to walk with or without aids or they have to use a wheelchair. Ninety percent of persons with spina bifida have urinary and fecal incontinence (Verpoorten & Buyse, 2008).

Sixty six percent of children with spina bifida in low income countries also develop hydrocephalus (Warf et al., 2009). Hydrocephalus is an abnormal expansion of ventricles within the brain caused by an imbalance between production and absorption of cerebrospinal fluid (CSF). Hydrocephalus also occurs without spina bifida, as a result of an obstruction caused by tumors, infections or a cerebral hemorrhage. Surgery is often needed for children with progressive hydrocephalus to prevent secondary impairments. Most children born with spina bifida also need surgery to close the back to prevent infections (March of Dimes, 2006).

In high income countries, the prevalence of spina bifida at birth has decreased to 0.5–1.000 per live births (Shaer et al., 2007) due to the use of folic acid through fortification of staple food or the daily intake of supplements (Miles, 2002), and antenatal screening, resulting in termination of pregnancies (Frey & Hauser, 2003). The prevalence of infantile hydrocephalus is 0.48, and for hydrocephalus associated with spina bifida 0.18 (Persson, Anderson, Wiklund, & Uvebrant, 2007). The incidence of spina bifida in low incomes countries is higher: 2.61/1000 live births in Tanzania (Kinasha & Manji, 2002), and 7/1000 deliveries in Nigeria (Airede, 1992). The main causes of the high incidence of infantile hydrocephalus (hydrocephalus without spina bifida) are the high incidence of neonatal infection and poor treatment of postnatal meningitis (Warf & Campbell, 2008).

Children with spina bifida and hydrocephalus often are in need of lifelong care. Key to success is to provide community-based rehabilitation services in addition to clinic-based surgery and rehabilitation (Warf, Wright, et al., 2011) and involving parents of children with spina bifida and hydrocephalus (Mertens & Bannink, 2012).

This article describes knowledge, beliefs, attitudes, and practices towards children with spina bifida and hydrocephalus in Uganda.

Methods

In Uganda, neurosurgery for spina bifida and hydrocephalus is offered in the national referral hospital in the capital city Kampala in central Uganda, and a specialised neurosurgical children's hospital in eastern Uganda. Specialised rehabilitation services including physio-, and occupational therapy, and social support through parent support groups and counseling are offered in 4 locations in the central, eastern, northern, and south western regions. This study aimed to cover these four regions and appraise regional similarities and differences.

Ethical approval and research clearance were obtained from Ghent University, Belgium, the Uganda Virus Research Institute, and the Uganda National Council for Science and Technology.

Focus group discussions and semi structured interviews were held with parents of children with spina bifida and hydrocephalus, policy makers, and service providers in between June 2011 and July 2012 (Table 1). During the same period participant observation was carried out to collect additional information and check findings during clinic days and home visits.

To ensure parents' voices are heard, separate focus group discussions with parents were held. Parents from the existing east, west, central, and northern regions' parents support groups for spina bifida and hydrocephalus were requested to participate in four different focus group discussions.

One focus group discussion was held with health, education, human rights, and social protection service providers from different regions. One focus group discussion was organised for disability policy makers, and donors active in policy making and programming for children with disabilities.

Key informant interviews were also held with five service providers involved in the medical, rehabilitative and social care for children with spina bifida and hydrocephalus, a member of parliament for persons with disabilities, and a staff member of the International Federation for Spina Bifida and Hydrocephalus. Observations were carried out in all five clinics during review days specifically for children with spina bifida and hydrocephalus.

In total, 178 parents of children with spina bifida and/or hydrocephalus between 4 and 14 years of age were interviewed. Consent was obtained from all participants. While the majority of the interviews took place at the clinic, 32 of the interviews took place at the homes of the children.

Observations were conducted of 248 parents and children with spina bifida and hydrocephalus during outreach clinics in the five geographical areas, during parent support group meetings, and for 32 in the home setting.

The focus groups were conducted to guide the development of the semi structured interview guidelines for in-depth discussion. The interviews and focus group discussions with parents were held in the local language of the area, and a translator was hired and trained for each area to assist in conducting the interviews and focus group discussions, and observations. The other focus groups and interviews were carried out in the English language.

The focus groups were audio recorded and transcribed verbatim (with the assistance of the translator for the parents' focus groups). The semi-structured interviews were not audio recorded, but answers were written out during the interviews and entered into a database after completion. Observations were written down, and checked with the findings from the interviews and focus groups.

The focus group and interview data were analysed by doing a line-by-line analysis of the manuscripts. In this process themes were identified representing knowledge, attitudes, and practices towards children with spina bifida and hydrocephalus. Observations were used to support these themes, and quotes registered during observations were matched to the relevant themes. The outcomes were grouped in tables by region. The answers of the parents and children's interviews were grouped and analysed per thematic area using thematic analysis (Howitt, 2010).

Results

Following thematic analysis, the results of the focus group discussions and interviews were grouped in the following themes:

1. Knowledge, beliefs, attitudes, and practices of community members
2. Regional references and specificities
3. Parent's experiences in communities

Table 2 outlines the knowledge, beliefs, attitudes, and practices of community members.

Knowledge and Beliefs

The most common community beliefs were that the defects are caused by witchcraft and curses, bad luck, use of family planning methods, divine intervention, and genetic problems in the family, which are believed to affect the baby before birth. A few believe the impairments are caused by intense physical exercise during pregnancy, and contact with sacrificial places before birth. The few respondents who recognised the contribution of the lack of folic acid indirectly by mentioning not following medical prescription and malnutrition during pregnancy as causes of spina bifida, were all health workers.

Some of the respondents believed spina bifida and hydrocephalus were caused by breach birth, and delayed or prolonged labour. Post-natal causes mentioned were convulsions in early childhood (for hydrocephalus), infections in early childhood, use of traditional herbs, and again divine intervention and curses.

“When you produce a child with a disability others say you are bewitched.” (Mother of a child with spina bifida, south western region)

“Parents do not know what happens to them when they give birth to a child like that. Many think it is caused by family planning due to ignorance.” (Policy maker, central region)

Overall, children with spina bifida and hydrocephalus are perceived as ‘useless’ children, unable to contribute to society. Community members do not believe they can live and function till they witness some children actually develop and go to school.

“Everyone told me my child will die. Now she is 7 years old and goes to school. They are all surprised.” (Mother of an 8-year-old girl with spina bifida and hydrocephalus, eastern region)

“They say that when my child reaches 15 [years of age], she will die.” (Mother of a 4-year-old girl with hydrocephalus, northern region)

Referral for appropriate treatment appears a big challenge for most parents, as knowledge is limited and health workers may equally be ignorant.

“Health workers keep the same beliefs as the community they live in, despite medical education, so they do not refer.” (Neurosurgeon, central region)

“No one gave me advice. They did not know what the child was suffering from. Even at the hospital the medical personnel did not know what the problem was. I had to spend 3 weeks there as they examined him.” (Mother of a 7-year-old girl with spina bifida and hydrocephalus, south western region)

Attitudes

Community attitudes have changed over time in some areas when community members have seen children recover from surgery and progress with rehabilitation and parental care. Initially children with spina bifida and hydrocephalus are seen as children who are dying, not worth investing in. Common statements made are “do not take this child to the hospital because it’s already dead”, and “do not pay school fees for this child, it is a waste of money”. Mothers are often blamed for producing a child with spina bifida and hydrocephalus. The general belief is that children with spina bifida and hydrocephalus are useless, can not contribute to society and are only a burden to their parents. For these reasons, parents are often pitied. Parents are also feared in some communities, as they are believed to have given birth to a ‘demon’. Children and parents are then excluded from community life. Some, however, believe that what has been given by God needs to be taken care of, and try to be supportive to parents. In a few cases communities believe that the child has special powers, and as a result treat the child well out of fear.

Practices

Resulting from the knowledge and attitudes outlined above, various similar practices were registered in all four regions. In all regions, parents are encouraged to take their child to the witchdoctor for rituals to ‘undo’ the curse, and thereby cure the impairments.

Children and their parents are often neglected and discriminated. Often incontinence is a reason for stigmatisation, as children cannot control their urine and faeces and are “smelling bad”.

“Our neighbours’ children don’t want to play with my child, they say she smells and she urinates everywhere like a baby.” (Mother of a 5-year-old girl with spina bifida, eastern region)

“Our children are normally excluded from government programs. One time government was registering children under 5 years. When they reached my home they refused to register my son with spina bifida.” (Mother of a 4-year-old boy with spina bifida and hydrocephalus, central region)

Out of shame and lack of support children are hidden in the houses, and locked up when parents need to go out to dig or to the market.

“I used to carry my son on my back, but now he is too heavy, he cannot walk, I do not have a househelp, I have to leave him home alone when I go to the garden.” (Mother of a 4-year-old son with spina bifida and hydrocephalus, northern region)

As children are believed to be 'dead', life giving practices such as breastfeeding are discouraged by community members.

"Some people would tell me not to breast feed the child so that she can die." (Mother of an 8-year-old son with spina bifida, eastern region)

Referral to health care and rehabilitation only takes place if there is any knowledge about this service, and if parents are encouraged by other parents, health workers, or community members, and have sufficient funds to cover the transport and treatment costs. When a child develops and grows older, some communities become more supportive to the parents, and parents regain hope.

Regional Differences

Overall regional differences in attitudes and practices are mainly observed in the northern region, while in the central, east, and south western regions similar observations were recorded. Each region in this study is the home to different tribes, with their own language, and cultural beliefs and practices. The central, east and western tribes have in common that they are Bantu tribes, while the northern tribe is Nilotic. In the northern region, the majority of the population was displaced during the 22-year conflict between the Government of Uganda and the Lord's Resistance Army (LRA). Since 2006, relative peace allowed people to return home and start rebuilding their communities. The other regions have enjoyed relative peace—apart from occasional border conflicts with neighbouring countries—since 1986.

In the eastern region, some particular beliefs about the causes of spina bifida and hydrocephalus were found. E.g. marrying someone who has another religion, attending a burial of a child with hydrocephalus pre-natally, or having the child fall on a rough or dirty surface at birth were believed to cause spina bifida or hydrocephalus. It was also believed that postnatally hydrocephalus could develop if a child would "over-cry", or because a child has been named after the maternal instead of paternal clan (as is the custom).

The difference in attitudes is based on the fear of contamination in the north, and belief that the parent and communities should be 'relieved' from the burden as this child will not contribute to the household; something which is absent in the other areas. The related practice in the northern region is to bring the child to the river. The child is tied on the mother's back with a cloth, which is commonly used to carry children. While the mother stands in the river, she loosens the wrapping cloth, and the child 'accidentally' falls off her back. This would usually happen within the first three months after birth or after a child's head started swelling, and would be reported as an 'accident' in which the child drowned:

"A mother with a child like that will tie it on her back, walk to the river, and untie the child so that it is taken by the water." (Service provider, northern region)

Some of the respondents mentioned that this is an old practice, no longer in place. However, others disputed this, and said mothers still 'untie' their child if born with an obvious physical disability. Some respondents related it to the 'burden' a mother could not face during displacement in camps, but others said this practice was already in place before the war started. The practice of actively killing an infant with a disability is not practiced in other regions, however, neglect to the extent that the child may die is practiced in other regions.

Amongst the respondents, the service providers believed the incidence of spina bifida and hydrocephalus may be higher in the north due to the conflict and related poverty, which resulted into poor feeding and lack of health care during this period. No statistics are available on the prevalence of spina bifida and hydrocephalus in Uganda.

Nevertheless, in each region there were one or two parents who mentioned that the child was seen as a human being with divine powers, and is treated with great respect.

Table 3 describes the different local words to describe children with disabilities and spina bifida and hydrocephalus across the four regions derived from interviews and observations.

In the central region a child with a disability is commonly referred to as *'omwana teyesobola'*—a child who can not manage himself or herself. In case of dysfunction of the lower limbs, *'omulema'* meaning lame is used. Sometimes *'kisirani'* is used, referring to a child who was born this way because one of the parents (usually the mother) was cursed. In the east, north, and west, a word for 'children who are not normal' is used to refer to a child with a disability: *'baana ba baleme'* in the east, *'langolo'* in the north, and *'ekimuga'* in the west.

Descriptions of a child with spina bifida and hydrocephalus vary in reference to the physical appearance of the child, and their 'abnormality', and the believed causes of the impairments. For example, in the central and western regions, *'ekizimba mu mugongo'*, a child with a swelling on the back; *'owacwekire orukizi'*, the one who has a split spine; and *'omwana w'ekizimba kyahamugongo'*, a child with a swelling at the back are used for children with spina bifida. In the north no specific word is used to describe children with spina bifida, however the word *'langwece'* is used to refer to incontinence, literally 'smelling of urine'. To describe hydrocephalus, reference is made in all regions to the enlarged head *'nakalanga'* and *'omwana ow'omutwe omunene'* in the central, *'baana be gimitwe migali'* in the east, *'wiye dit'* in the north, and *'omwaana w'orutwe'* in the south west. In the central region children are sometimes called *'kasepiki'* (saucepan), referring to the size of their heads (as big as a saucepan).

Explanatory descriptions vary from *'balango'*, twin in the central region for spina bifida, and *'ekyonziira'*, a child whose condition is due to the influence of evil spirits for hydrocephalus, to *'babaana be kamakanga'*, mysterious children, *'babaana be kimisambwa'*, demonic children or children of evil spirits, and *'babaana ba family'*, family planning children for both impairments. The word twin in the central region is used as people think the swelling on the back signifies another body: a twin who never developed fully.

In the northern region additional last names are given to the children with an abnormality: *'Ojok'* for male and *'Ajok'* for female. The names are not specific to spina bifida but refer to an abnormality or omen seen by the parent, literally meaning 'touched by evil'. Opinions differ about whether this is only negative or also refers to spiritual powers.

Not only children, but also parents are referred to as the parent of a child with a disability in all regions.

"The people who nickname the children do the same to the parents as well. E.g. whenever I am passing [people in the village] they call me 'Nyinaw Ekimuga', meaning that woman gave birth to an abnormal child." (Mother of a 4-year-old daughter with spina bifida, south western region)

Parent's Experiences

In total, 178 parents of children with spina bifida and hydrocephalus with an average age of 7.6 years (min 4, max 14), were interviewed from four different regions in the country (57 central, 35 eastern, 57 northern, 29 western region). The majority were mothers (73.6%), followed by fathers (12.9%), grandmothers (7.9%), and others.

Personal Experiences

Most parents say to have felt disappointed and worried at the birth of their child. Common statements made by parents expressing their worries and disappointment and search for explanations just after the birth of their child were "I had wanted a healthy child", "I did not know

what to do”, “nobody wants a disabled child”, “I thought I was being punished by God”, and “I thought my child was bewitched”. Many took their children to the witchdoctor as advised by relatives and community members to undo the “curse” on their child. Parents report that after trying and failing with this option, some started looking for biomedical care.

The majority of parents express that the disappointment and worrying reduced after having received medical and rehabilitative care, and support from other parents. Many parents mentioned to feel supported by the “parents support groups”, self-help groups created by the parents who attend the specialised care centers in the various regions with support from the International Federation for Spina Bifida and Hydrocephalus. Most expressed a loving and caring attitude towards their child: “I love my child”, “the love we have for this child is more than that we have for other children”, and “we treat and love the child like our other children”.

“When she was born, we did not think she would survive. Now she goes to school and is one of the best of her class. She speaks on public events about her disability and encourages other children. We are proud of her.” (Father of 14-year-old girl with spina bifida, south western region)

“I thought I was the only one with a child like mine, but I discovered there are many, and I was encouraged by other parents when I joined the parents support group. They taught me how to look after my child and said I should not hide the child, but show it to others so that they can know my child can do things too, and is like their children. Now I take her with me everywhere I go.” (Mother of a 4-year-old daughter with spina bifida, south western region)

However a number of parents remained worried about community stigma and opportunities for their child “nobody accepts her/him”. Passive acceptance was also found, reflected in statements like “my child was given by God, I have to look after her, I have no choice” and “I have learned how to look after this child, no one else will”.

Family

Most of the parents said they felt supported by their families when they gave birth to a child with spina bifida (60%). A third of the parents reported to have been treated badly (34%) by their families, while the remaining felt some had been good to them, while others mistreated them.

Those who said they are supported by their families expressed this by saying “we are treated well. They see it as something normal”, and reference was sometimes made to health care workers in the family: “there are health workers in our family who knew about the condition.”

The 16% of parents who said they were treated well by some, and mistreated by others said “my family treats me well except for my husbands’ family which rejected the child”, and “we are loved by some. Others blame me for not doing any rituals for the child because they believe that the rituals would heal her.”

The third of the parents who reported to be mistreated were all mothers and said “they treated me badly. They said I produced a snake”, “they did not see my child for a long time. I hid her but when they eventually saw her they talked a lot and even discouraged me from breastfeeding her”, “My husband and his family told me to throw him in water or kill him. Even my own children ran away from me”, “They never liked my child. Some even refused to carry him up to today.”

The mothers, who reported to be mistreated, said it was mainly their in-laws who treated them badly: “my husband and his clan neglected me. He said “this kind of disability is not there in our family. When I gave birth my child came with club feet. His clan started despising me. I am now staying with my parents. When I call him for financial support he says it is none of his business”.

With time, the situation changed for some parents. They received more support when relatives saw the child survived and developed: “some people told my husband to send me away but he was firm. Now they have changed their attitude”. Parents reported that even if they had informed their family about the possible treatment and rehabilitation of their child, relatives tended to only be more supportive when they actually noticed improvement in the development of the child: “they did not believe me when I said there is treatment for my child, they only started being supportive when I had gone for treatment many times, and my child could now walk using crutches”, and “we were stigmatized by our family but after my son was operated in Mbale and he grew and started talking, they now treat us well.”

Community

Parents report various reactions in their communities when their child was born. While 55% of the parents said they were treated fairly well: “They are good to us except that they always ask a lot of questions”, the other 45% reported community members called them names, gossiped about them, said that their child would die, and rejected them.

Those who reported to be treated badly said: “people discourage me from taking the child to hospital. They say that I am wasting money. If they were me, they would leave the child to die”, “many people ran away from me. They did not even want me to go to their homes. They would lock themselves in their houses and refused to buy things from me for fear that they were taking bad luck from me”, “My child has been sick but people told me to leave her to die”.

Currently the percentage of parents who feel their child is treated well by their communities is 63%: “they said that they had never seen a child like mine. However they have stopped talking and they are now good to us”, “they are good to us. Sometimes they give me money for treatment”.

The percentage of parents who report current calling names, gossiping, and other negative attitudes from community members is 27%: “they call her big head”, “children beat and abuse him”. Ten percent says currently there is a mix with both positive and negative reactions: “Adults do not abuse him. It is children who abuse him because of the size of his head and club feet.”

Health Facilities

Parents report that in most cases they are treated well at hospitals and health centres, within the limitations of the Ugandan health system. A few said to be rejected by the health workers, discouraged from taking care of their child, or did not receive any care as the health worker was anxious about providing treatment for a ‘special’ case: “they fear to inject her at the local clinic. They only give tablets and advise me to take her back to CURE [specialized neurosurgical hospital] in Mbale.”

All parents said to be treated well at the specialized neurosurgical hospital in the country and its partner organisation who offer rehabilitative care for children with spina bifida and hydrocephalus: “the people are so nice to us, I would not know what to do without them”, “they [the staff from the specialized rehabilitation center] have explained me everything about my child, now I know it is not my fault, and my child can grow. I also meet other parents here, they encourage me.”

While grateful for the reduced rates applied in these facilities, parents did report difficulty in footing the bills for the specialised health care and assistive devices required for their children.

School

Of the parents interviewed, 60% of children were going to school. A number of parents had difficulties finding a school where their child would be accepted: “I went to 5 schools, the first 4

rejected him because of incontinence”, “they are good to her although in the beginning I had problems finding a good school for her”. While the majority is treated well in schools now, many were bullied initially: “she was bullied in the beginning but now the children are good after explaining to the teacher”. Parents report that when they talked to the teachers and explained the disability of their child, a lot improved: “they are good. I talked to them about her condition”, “they treat her well except that I have to pay a little extra so that they take care”.

However, approximately 14% of the parents with children in school still complain of mistreatment by other children and teachers, and 3% report that their child dropped out due to bullying and lack of understanding by the teachers: “when she started school children abused her that she has a big head and she refused to go back”, “some discriminate and isolate him especially when he wets himself”, “his classmates bully him and teachers do not allow him to play out of fear that he will fall down.”

Future

Parents would like their children to be treated like any other child, with love, respect and care: “We would like to be loved and treated well”. They feel community sensitisation is necessary to reduce people’s negative attitudes and bad behaviors such as calling names, blaming and discouraging the parents: “people should know that it was not my fault to have such a child”, “people should stop laughing at us. My son should be treated like any other child”. Parents say their children, if doing well, are the best testimony for others to believe it is worth investing in children with spina bifida and hydrocephalus. However, they do feel there is lack of commitment and budget at policy level to implement inclusion on the ground: “government should put more efforts in health care and education, if the general services do not improve, people never have time and money to look after our children with special needs.”

Discussion

This study describes how negative knowledge, beliefs, attitudes, and practices create barriers to treatment and inclusion of children with spina bifida and hydrocephalus and their parents in Uganda. It shows how knowledge, beliefs, attitudes, and practices evolve over time, and become more conducive towards accessing treatment and achieving inclusion. It provides recommendations to promote early intervention, and facilitate the creation of more positive knowledge, beliefs, attitudes, and practices systems.

Knowledge and Beliefs

Our findings show communities generally believe spina bifida and hydrocephalus are caused by witchcraft, bad luck, and the use of family planning methods. While witchcraft, curses, and bad luck have been cited in other studies as believed causes of spina bifida and hydrocephalus (Franzen, 1990; Masasa, Irwin-Carruthers, & Faure, 2005), the use of family planning seems a more recent belief. Use of contraception is discouraged by some churches in Uganda, and negative attitudes exist against its use. The explanation of having used ‘family planning’ is a more medical discourse but still with a divine connotation, blaming the mother for using medical methods which are against ‘God’s will’.

Words to describe a child with spina bifida and hydrocephalus in the local language vary based on the various languages spoken, but all refer to the physical appearance of the child, their ‘abnormality’, and the believed causes of the impairments.

Attitudes

After birth of a child with spina bifida and hydrocephalus community attitudes are largely negative. Parents are pitied, and discouraged from taking care of the child, as they are “already dead” or “dying”. This is in line with findings from South Africa where community members said to pity a child which always needs to be helped (Masasa et al., 2005). In some areas when community members have seen children recover from surgery and progress with rehabilitation and parental care, attitudes change and become more positive. Possibly when seen that treatment is available, this allows for a collective recognition of the impairment, if there is treatment the person with the impairment is recognised as a human being and can be removed from the ‘dead’ category.

Practices

Following the negative attitudes discrimination and neglect are common practices in communities. In the northern region parents are encouraged to kill the child by letting the child ‘accidentally’ fall off one’s back on untying the commonly used cloth with which babies are carried on the back. However when informed and acknowledging that some of the children do not die, and thrive, more supportive practices are established. Parents play the key role in making this happen, which corresponds with findings of Miles (2006) on how in each society some families are able to overcome considerable challenges to ignore the cultural expectation to kill the child, and try their best to enhance life chances of their child.

Some of the parents in Uganda described duality in support, whereby relatives may be supportive, but also do not agree that a parent does not perform the required cultural rituals, which are believed to cure the child. In all regions we found that parents were encouraged to conduct rituals with a witchdoctor to cure the child initially. Many parents tried this, and after failing to receive the expected cure, would start looking for biomedical care. Similar searches for cure have been described in India: “parents of children with disabilities are convinced that their offspring could be cured if they can remove the effects of ‘God’s punishment’ by prayers. They pay money to faith healers and on conducting worship rituals” (Lakhan & Sharma, 2010, p.114). Stroeken (2010) explains witchcraft as a moral discourse in which the responsibility for having a child with a disability could be placed with outside powers, taking away the dilemma and responsibility of the parent.

Gender

In our study negative gender attitudes and practices towards mothers of children with spina bifida and hydrocephalus were observed. In various cases the mother was blamed for giving birth to a child with a disability and was abandoned by her husband and/or mistreated by her in laws.

Kisanji’s (1995) study of attitudes towards persons with visual, hearing, physical, intellectual and behavioural impairments from Liberia, Tanzania, Zambia and DRC, emphasises that community attitudes “reflect fairness and equal opportunities for all community members including those with impairments” (Kisanji, 1995, p.5). Similar observations were made by Edgell and Stanfield (1972) on care for children with disabilities in Uganda: “On the whole the uncomplicated severely subnormal child is cared for by the extended Ugandan family and the threat of rejection and abandonment arises only where there is either totally unacceptable antisocial behaviour or no family support – for example, the unmarried or deserted mother” (Edgell & Stanfield, 1972, p.551). As these study and observations were made decade(s) ago, we do believe that with urbanisation and leaving behind traditional village structures, this has become more challenging for parents in this time. Nevertheless, we found room for positive change in attitudes once the

child has developed beyond what was (s)he initially expected to, and of fathers who stood by their wives and supported their child.

Regional variety

Regional differences in attitudes and practices are mainly observed in the northern region, while in the central, east, and south western regions similar observations were recorded. The difference in attitudes is based on the belief that contact with a person with a disability such as spina bifida or hydrocephalus (as well as epilepsy among others) could infect you with the same in the north, something which appears to be uncommon in the other areas. The practice related to these beliefs and attitudes is 'bringing the child to the river'. While this is not actively practised in other regions, denial of feeding and neglect to the extent that the child may die due to malnutrition or untreated infections is also practised in other regions. In exceptional cases children with spina bifida and hydrocephalus are seen as a human being with divine powers, and are treated with great respect. These exceptions show how knowledge, beliefs, attitudes, and practices differ within communities.

Parents' voices

Parents' experiences show a shift in attitudes, changing from disappointment and worry at the birth of their child, to more positive loving and caring attitudes after having received medical and rehabilitative care, and support from other parents. The shift in caring follows the shifting beliefs from being a 'curse' to a 'God given' child, making caring for this child not just a parental obligation but a divine vocation. This is in line with the search of a cure, initially with witchdoctors, and the later received bio-medical care from faith based hospitals and rehabilitation centers. Families and communities are overall supportive when a parent has 'proven' that with their investment in their child, the child can develop and be like any other child. However, support to enable parents to go through this process of surgery, and rehabilitative care, is lacking for almost half of the parents, leaving them in a rather isolated and challenging situation.

After acceptance by (some of) the family and community members, challenges remain for their children with spina bifida and hydrocephalus to access education and health services. While general health services are usually accessible, some children are turned away due to lack of knowledge about the impairments. There may be low awareness about the impairments and possibilities for referral in health facilities. While specialised facilities are often far, they are well equipped and staff is caring. Nationally 94.5% of children aged 4 to 14 are school going in Uganda (Government of Uganda, 2013), in our findings only 60% were going to school, and many faced challenges of being accepted and included. While enrollment may be low due to inclusion challenges, dropout rates may be higher due to bullying of children with spina bifida and hydrocephalus.

Parents point out that community sensitisation is necessary to reduce people's negative attitudes and practices, as well as commitment of government to invest in children with disabilities. By showing other that their children do well, they believe others may start believing too that it is worth investing in children with spina bifida and hydrocephalus.

Recommendations

Our study is a unique description of the voices and experiences of Ugandan parents of children with spina bifida. It argues for measurement of especially attitudes and practices at different points in time. Effect studies only describing attitudes and outcomes at one point in time, do not take into account the dynamics attitudes and practices are subject to (Gupta & Singhal, 2004).

We argue that when studying knowledge, attitudes, and practices, evolution needs to be added. Longitudinal studies measuring attitudes and practices at different points in time, with exposure to a child with spina bifida and/or hydrocephalus will give a more realistic and enabling description of inclusion in the society. In non-longitudinal studies researchers can systematically ask about evolution of knowledge, attitudes and practices over time.

The findings of our study show the importance of early intervention and sensitisation: when a community is informed about the impairments, beliefs about the causes may change and stress the contribution of parents in acquiring the impairment less. This could help in communities being more open and supportive rather than ejective towards parents and children. Health workers, teachers, and other service providers need to be sensitised about the impairments and referral paths, and encourage parents to seek treatment and promote inclusion of their child. When informed and encouraged, parents and children are more likely to seek treatment, resulting in a better prognosis, and ultimately have a better chance to be accepted and included in their communities as the attitudes and practices towards a child change. It should however be noted that knowledge about the impairments in itself may not change the attitudes, even if parents informed their relatives and community members, changes in attitudes were mostly reported after the child actually visibly developed. Once there is an observation on quality of life, and future prospect, community members are willing to invest and give children with SBH a chance. Our findings support the methodology of the International Federation of Spina Bifida and Hydrocephalus Spina Bifida and Hydrocephalus Interdisciplinary care Program (SHIP), in which service providers from different disciplines collaborate and put parents and children at the centre, not as recipients, but active participants in the process of care.

Parents play an important role in actively engaging their community to see their child, and not be afraid of the child. Parent support groups can help parents to encourage each other and feel strong in fighting for this in their communities.

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Table 1. Overview of study methods and respondents.

Method	Parents of children with SBH	Service providers	Policy makers and donors
Focus group discussion	31	12	7
Semi structured interview	178	5	2
Observation	248	5	0

Table 2. Knowledge, beliefs, attitudes, and practices about spina bifida and hydrocephalus.

<p>Knowledge and beliefs</p> <p>Community knowledge and beliefs about causes of spina bifida and hydrocephalus and possible prevention</p>	<p>Pre-natal</p> <ul style="list-style-type: none"> • Witchcraft and curses • Bad luck • Use of family planning methods • Challenge given by God • Genetic problem in the family • Intense physical exercise during pregnancy • When one does not follow medical prescriptions given at the hospital • Walking, sitting or passing tombs or sacrificial places when pregnant • Malnutrition during pregnancy • Women who have mourned children who have died of hydrocephalus, will give birth to a child with hydrocephalus (Eastern region only) • Marrying someone who does not share the same faith (Eastern region only) <p>Peri-natal</p> <ul style="list-style-type: none"> • Breach birth • Delayed or prolonged labour • When a child falls on a rough or dirty surface at birth (East only) <p>Post-natal</p> <ul style="list-style-type: none"> • Convulsions in early childhood (for hydrocephalus) • Infections in early childhood • Use of traditional herbs • The devil at work • Curses • Over-crying of a child (for hydrocephalus) (East only) • Naming a child after the maternal clan (East only) • Accidents and war (North only)
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<p>Community knowledge and beliefs about prognosis of children with spina bifida and hydrocephalus</p>	<ul style="list-style-type: none"> • Drugs for epilepsy (North only) <p>By avoiding the above one could prevent having a child with spina bifida or hydrocephalus.</p> <p>Prognosis and treatment</p> <ul style="list-style-type: none"> • Child will only get worse and die, will not be able to function. • There is no treatment unless the curse is taken away or God performs a miracle • Child will infect others, should be isolated or killed (North only) • A few heard of possibilities for surgery and rehabilitation services
<p>Community attitudes towards children with spina bifida and hydrocephalus</p>	<ul style="list-style-type: none"> • Say it is the mothers' fault • Feel that the children are useless, cannot contribute to society, are a burden to the parent • Pity parents and child • Believe it is not worth investing in children who cannot achieve anything and advise parents accordingly at birth; if they see the child is developing, they sometimes become supportive • Parents have produced a 'demon' • Say that is what God has given has to be taken care of • Fear contamination and therefore exclude children and parents (North only) • Say the child should not be kept in the family, and should be destroyed (North only) • Say the child has special powers (mentioned by 5 respondents)
<p>Practices in communities towards children with spina bifida and hydrocephalus</p>	<ul style="list-style-type: none"> • Exclude children and parents from community life (complain of smell / incontinence) • Take child to the witch doctor or traditional healers for ceremonies to undo the curse • Neglect the child • Hide child in the villages • Lock child in the house or on chain when parent needs to go out • Throw the child in the river (North only) • Referral to health care and rehabilitation only if known and encouraged by other parents, health workers, or community members • If child is growing and develops more than expected, community tends to be more supportive to parents, and parents regain hope.

Table 3. Local words used to refer to children with a disability, and specifically SBH.

Word	Central Uganda (language: Luganda)	Eastern Uganda (language: Lugishu)	Northern Uganda (language: Luo – Acholi dialect)	South Western Uganda (language: Ruyankole)
Child with a disability	<p><i>'Omwana teyesobola'</i> – a child who cannot manage him/herself. In case of dysfunction of the lower limbs, <i>'omulema'</i> - lame. No specific naming of the child by parents.</p> <p><i>'Kisirani'</i>- child born because one of the parents (usually the mother) was cursed</p>	<p><i>'Baana ba baleme'</i> – children who are not normal.</p> <p>No specific naming of the child by parents.</p>	<p><i>'Langolo'</i> (not normal). In addition children are given <i>'Ojok'</i> (Kranzer et al.) or <i>'Ajok'</i> (female) as their last names by their parents, meaning the one that was touched by an evil spirit.</p>	<p><i>'Ekimuga'</i>, someone who is not normal in the way he/she looks or does things. No specific naming of the child by parents.</p>
Child with spina bifida	<p><i>'Ekizimba mu mugongo'</i> - child with a swelling on the back</p> <p><i>'Balongo'</i> – twin, people think that the swelling on the back was due to the fact that they were supposed to be twins and the second did not develop properly hence the swelling.</p>	<p><i>'Babaana be kamakanga'</i>– Mysterious children</p> <p><i>'Babaana be kimisambwa'</i> – Demonic children or children of evil spirits</p> <p><i>'Babaana ba family'</i> – Family planning children</p>	<p>No specific word, but those who are incontinent are called <i>'langwece'</i>, which means smelling of urine.</p>	<p><i>'Owacwekire orukizi'</i> - the one who has a split spine;</p> <p><i>'Omwana w'ekizimba kyahamugongo'</i> - a child with a swelling at the back</p>
Child with hydrocephalus	<p><i>'Nakalanga'</i>- child with big head</p> <p><i>'Ekyonziira'</i> - a child whose condition is caused by evil spirits</p> <p><i>'Kasepiki'</i>- saucepan. School going children with hydrocephalus report that other children use this to say that their heads are as big as a saucepan.</p> <p><i>'Omwana ow'omutwe omunene'</i> - child with a big head</p>	<p><i>'Baana be gimitwe migali'</i>- Children with big heads</p>	<p><i>'Wiye dit'</i> (big head)</p>	<p><i>'Omwana w'orutwe'</i> (child with a big head)</p>

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Chapter 3 Health and Rehabilitative Services



3.1 Prevention of spina bifida: folic acid intake during pregnancy in northern Uganda²

Abstract

The intake of folic acid before conception and during the first trimester of pregnancy can prevent spina bifida. This paper describes folic acid intake in women in Gulu district in northern Uganda. Structured interviews were held with 394 women attending antenatal care (ANC), 15 mothers of children with spina bifida, and 35 health workers in 2012 and 2013. SPSS16 was used for data analysis.

1/4 mothers of children with spina bifida took folic acid during late pregnancy, none preconception. None had knowledge about folic acid and spina bifida prevention. 33.5% of women attending ANC had ever heard about spina bifida, 1% knew folic acid intake can prevent spina bifida. 42.4% took folic acid supplements in late pregnancy, 8.1% during the first trimester, none preconception. All women said to have eaten food rich in folic acid. None were aware about fortified foods. 7% of health workers understood the importance of early folic acid intake. All health workers recommended folic acid intake to women attending ANC. 20% of the health workers and 25% of the women said folic acid supplements are not always available.

Folic acid intake is limited in northern Uganda. This is attributed to limited education and understanding of women and health workers about the importance of early folic acid intake, late presentation of women at ANC, poor supply chain and dilapidated health services caused by war and poverty. A combination of food fortification, sensitization of health workers, women, and improving folic acid supply is recommended.

² Based on: Bannink, F., Larok, R., Bauwens, L., Kirabira P., and van Hove, G. (2015) Prevention of spina bifida: folic acid intake during pregnancy in northern Uganda. *The Pan African Medical Journal* 20: 90. DOI: 10.11604/pamj.2015.20.90.533.

Background

Spina Bifida is a neural tube defect (NTD) caused by a fault in the development of the central nervous system in the first 25 days of the pregnancy (Northrup & Volcik, 2000). The worldwide incidence varies between 0.17 and 6.39 per 1000 live births (Bowman et al., 2009; Kinasha & Manji, 2002; Msamati et al., 2000; Shaer et al., 2007). An estimated 1,400 children are born with spina bifida in Uganda annually (Warf, Wright, et al., 2011). Incidence and prevalence rates in Uganda are probably higher due to absence of folate consumption by pregnant women, lack of pre-natal care (Miles, 2006b), absence of secondary prevention (Frey & Hauser, 2003), higher exposure to environmental risk factors such as dioxins (Safi et al., 2012) and fumonisins intake (Hendricks, 1999; Marasas et al., 2004; Wild & Gong, 2010), and high birth rates (Warf, Wright, et al., 2011).

Folic acid (also referred to as folate, folacin, Vitamin B 9, pteroylglutamic acid), delivered through supplementation or fortification (or other strategies) prevents the first occurrence of NTDs (Czeizel, 2000; Wilson et al., 2003) as well as the recurrence of NTDs in families with previous NTD-affected pregnancy (MRC Vitamin Study Research Group, 1991). The World Health Organization (WHO) recommends that all women of childbearing age consume 400 µg of folic acid daily and that women with pregnancies previously affected by NTDs consume 5000 µg of FA daily (Pena-Rosas, De-Regil, Dowswell, & Viteri, 2012). Women should consume these amounts in the peri-conceptual period as it takes 8 weeks to reach the optimal level of serum folate (WHO, 2012). Supplements are an effective way to prevent spina bifida, but there is low compliance (Brough, Rees, Crawford, & Dorman, 2009; R. M. Nilsen et al., 2006). Countries fortifying the flour with at least folic acid see an average 46% reduction of NTD prevalence (Pachón et al., 2013).

In Uganda only 17% of pregnant women attend antenatal care (ANC) before the fourth month of pregnancy (Government of Uganda, 2011). Of those who do attend ANC women are 5.5 months pregnant (median) when they come for their first visit. The Ugandan Government advises intake of folic acid following WHO recommendations (Government of Uganda, 2010). In a study in rural Uganda 13.2% of women attending ANC took folic acid during pregnancy (Mbule, Byaruhanga, Kabahenda, & Lubowa, 2013).

The health indicators for the northern region are worse compared to the rest of the country (Government of Uganda, 2011), partly attributed to conflict between the Government of Uganda and the Lord's Resistance Army (1986 – 2006), which displaced an estimated 2 million people into internally displaced people (IDP) camps (Annan, 2008). Lack of human resources, limited knowledge, shortages of medical supplies and inadequate infrastructure hamper implementation of quality health services (Ahoua et al, 2010; AVSI Foundation & UNICEF Uganda, 2011). The study area, Gulu district had 410,673 inhabitants with 15,992 deliveries, 33% of pregnant women attending 4 ANC visits, and 72% of staff positions filled in health facilities in 2012 (AVSI Foundation & Gulu District Health Office, 2012).

Uganda passed a national legislation to require fortification of wheat and maize flours with iron, zinc, folic acid and other B vitamins in 2011. The legislation requires fortification of white and brown wheat flours and maize products produced in Uganda as well as those imported to Uganda. Whilst some flour producers have started fortifying their products, not all are compliant (Food Fortification Initiative, 2012).

Methods

Quantitative and qualitative descriptive cross-sectional survey methods were employed to collect data on folic acid intake and understanding of its effect on the prevention of spina bifida in Gulu district. In total 15 mothers of children with spina bifida were asked about folic acid intake during their pregnancy. In addition 394 women in reproductive age group (15-49 years) attending ANC were surveyed. As well 35 health workers, working at ANC at government health facilities were interviewed. The 394 women and 35 health workers were selected by simple random sampling at antenatal clinics. The mothers of children with spina bifida were purposefully selected from a mobile specialist review clinic for children with neurological conditions in Gulu. The data collection tools included semi structured questionnaires and interviews. Data was entered and analyzed using SPSS16.

Results

Study participants had a median age of 29 years, the majority was married, Christian, completed primary school, and had 3 to 4 children. Table 1 shows the demographic characteristics of 15 mothers of children with spina bifida, 394 women in reproductive age attending antenatal clinics, and 35 health workers.

Of the 15 mothers of children with spina bifida interviewed, only a fourth took folic acid supplements during pregnancy, and none of them took these in the first semester (table 2). All mothers of children with spina bifida had never heard about spina bifida before they gave birth to their child, and had not known about the preventative effect of folic acid. All said they ate vegetables rich in folic acid such as spinach and avocado before and during pregnancy, but not on a daily basis. Intake varied during the year depending on whether they were dependent on food handouts during their time in the internally displaced people camps, and/or the season and availability. None had heard about food fortification before having a child with spina bifida.

Of the women attending ANC, 65.8% lived within 2 kilometers of the health facility (259), 23.4% (92) within 5kms, and 11% (43) beyond 5kms. 50.5% (197) took folic acid, none before pregnancy, and only 8.1% (32) during the first trimester of pregnancy (table 2). In total 33.5% (132/394) of the women attending ANC had ever heard about spina bifida. Only 1% (2) knew about the preventative effect of folic acid intake. Women who had knowledge about spina bifida were more likely to take folic acid than those who did not (table 3, $\chi^2=26.24$, $p<0.001$).

All women said they ate vegetables rich in folic acid such as spinach, okra, beans, lentils and avocado regularly. Intake varied during the year depending on the season and availability. None of the women had heard about food fortification or recognized the signs on available food products. Of the women taking folic acid, 75.5% (149/197) said folic acid (in 5mg tablets) was available at the health facility during their visits. The other 24.5% (46) said sometimes stock outs occurred. Those who had access to folic acid at the health facility were more likely to take it ($\chi^2=28.12$, $p<0.001$). Women attending health education at the health facilities were more likely to take folic acid compared to those who did not receive health education ($\chi^2=26.59$, $p<0.001$).

All health workers had ever heard of spina bifida. The majority of the health workers, 80% (28/35), believed folic acid intake and good antenatal care could prevent spina bifida, 9% (3/35) believed there is no prevention, while 11% (4/35) were not aware of any. Of those who believed folic acid intake could prevent spina bifida (28/35), 7% (2/35) said it should be taken before pregnancy and early weeks of pregnancy, 25% (7/35) said preferably before birth or as soon as the woman realizes she is pregnant for 90 days, 39% (11/35) recommended folic acid use (5mg tablets once a day) throughout pregnancy, and 29% (8/35) health workers recommended folic acid use in only second and third trimester. All health workers felt that intake of folic acid was important for the babies general development, and mother's health. A fifth (7/35) of the health workers said folic acid is not always available at the health facility they work at. None of the health workers had heard about food fortification.

Discussion

Our findings show that knowledge about the preventative effect of folic acid on spina bifida, and folate intake before and during the first weeks of pregnancy is very limited in northern Uganda. Awareness of spina bifida (33.5%) in women attending ANC was in line with earlier studies, e.g. 25.5% in Nigeria (Rabiu, Tihamiyu, & Awoyinka, 2012), and 53% in Congo (Claude, Juvenal, & Hawkes, 2012).

With 17.3% of the Ugandan women in our study taking folic acid supplements during the first trimester of pregnancy, the percentage is slightly lower than the worldwide estimate of only 20% of pregnant women being able to follow the recommendation of folic acid intake for prevention (Scott, 2011). Whilst peri-conceptional intake of folic acid supplements was 17.2% in Tanzania (Ogundipe et al., 2012), our study population did not report any early intake of supplements. Women did report to regularly consume foods rich in folic acid. One of the limitations of our study is that it could not define the amounts consumed, as no food samples or biomedical investigations were conducted.

The lack of folic acid intake in northern Uganda could partially be explained by the lack of knowledge on the importance of intake prior to and in the first 28 days of pregnancy amongst the health workers interviewed. Health workers in our study had knowledge about the importance of folic acid intake

during pregnancy but not about the crucial pre-conceptual and early conceptual period, they were also not aware of the recommended dosage.

Sensitization of health workers and women in reproductive age is key to encourage early ANC attendance and folic acid intake. Alongside sensitization at the antenatal clinic, women should receive sexual and reproductive health education from an early age. Following the conflict, educational attainment has been lacking in the northern region, and girls secondary school completion remains low (Government of Uganda, 2011). Counseling on folic acid use in the media and marriage counseling may be considered to inform young couples as many will not have received formal sexual and reproductive health education. In Congo patient education through video media increased awareness and knowledge of spina bifida and folic acid (Claude et al., 2012). In northern Uganda mobile video media could be explored. Radio messages have proven powerful in transferring health messages in the region and could also be used (Bannink-Mbazzi et al., 2013; Kitara, Ocer, Lanyero, & Ocom, 2013; Mertens & Bannink, 2012).

To improve access to folic acid supplements, there is need to strengthen the supply chain and address stock outs, so that folic acid supplements are provided at the first antenatal visit, or ideally to all women in child bearing age. Gulu district health facilities had a 57% basic drug stock out in 2012 (AVSI Foundation & Gulu District Health Office, 2012). Aside assuring folic acid stocks in health facilities, we recommend sensitization on the correct dosage. In a WHO project in Uganda a huge difference was found between and within districts in the dosage of folic acid provided to pregnant women (WHO, 2001). The differences were not only caused by lack of supplies but also by health worker's lack of knowledge on updated guidelines. During the war health structures in northern Uganda dilapidated and health workers in the area may not have received the necessary training, resulting in knowledge gaps. With targeted recovery plans of the Ugandan Government, supported by donors, and national guidelines from the Ministry of Health, the health system is recovering and targets are set to improve maternal and child health (Government of Uganda 2010; 2007). Whilst educating health workers on the correct dosage, supplies of 400µg rather than 5mg tablets should be availed by the National Medical Stores to enable health workers provide the correct preventative dosage, as 1mg and not 5mg is the upper tolerable daily limit (WHO, 2012).

Together with improving folic acid supplementation, we also argue for mandatory food fortification and implementation of this process at national level to increase levels of folic acid intake for all women in childbearing age. Participants in our study were not familiar with food fortification. Globally 25% of folic acid-preventable spina bifida is being prevented, and mandatory food fortification is recommended to achieve total prevention (Youngblood et al., 2013). Sensitization about food fortification and recognition of the label can increase use of fortified foods. In South Africa folic acid fortification of staple foods reduced the prevalence of spina bifida with 41.6%, benefits outweighed the costs of food fortification (Sayed, Bourne, Pattinson, Nixon, & Henderson,

2008). It should be noted that not all women in reproductive age will benefit from food fortification, as many are subsistent farmers and are more likely to feed from their own produce than goods sold on the markets.

We recommend further studies involving women prior to conception, during pregnancy and after birth with frequent measurements of folic acid intake to understand the actual levels of folic acid intake in the population, and evaluation studies to measure possible effects of sensitization campaigns and training of health workers in future.

Conclusion

Folic acid intake is limited in northern Uganda. This is attributed to limited education and understanding of women and health workers about the importance of early folic acid intake, late presentation of women at antenatal care, poor supply chain and dilapidated health services caused by war and poverty. A combination of food fortification, sensitization of health workers, women, and improving folic acid supply is recommended to reduce the number of children born with neural tube defects such as spina bifida.

Acknowledgements

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Tables

Table 1. Demographic characteristics respondents northern Uganda

Characteristics	Mothers of children with spina bifida (N=15)	Women attending antenatal clinics (N=394)	Health workers in antenatal clinics (N=35)
Age (mean)	31 (range 22 – 36)	29 years (range 18 – 40, SD 7.8)	28 years (range 22 – 42)
Marital status	6.7% (1) single, 60% (9) married, 13.3% (2) separated, 20% (3)	19% (75) single, 72.2% (284) married, 5% (20) separated, 3.8% (15)	11.4% (4) single, 88.6% (31) married

	widowed	widowed	
Number of children	4.2 mean (range 1 – 8)	3.8 children (range 0 - 9)	3.1 mean (0 – 6)
Education	26.6 % (4) never went to school, 60.0% completed primary (9), 6.7% (1) completed secondary, 6.7% (1) higher education	15.8% (63) never went to school, 55.6% (219) completed primary, 22.4% (88) completed secondary, 6.2% (24) higher education	54% (19) nursing - enrolled, 11% (4) registered nursing - registered, 35% (12) midwifery
Religion	86.7% (13) Christian 13.3% (2) Muslim	97% (382) Christian 3% (12) Muslim	98% (34) Christian 2% (1) Muslim
Occupation	73.2% (11) peasant farmers, 13.4% (2) petty traders, 13.4% (2) others	44.7% (176) peasant farmers, 39.9% (157) petty traders, 15.5% (61) others	54% (19) enrolled nurse, 11% (4) registered nurse, 35% (12) midwife

Table 2. Folic acid intake in mothers of children with spina bifida and women attending ANC in northern Uganda

Started taking FA	Mother of children with spina bifida	Women attending antenatal care
Before conception	0% (0)	0% (0)
During pregnancy (first trimester)	0% (0)	8.1% (32)
During pregnancy (2nd and 3rd trimester)	26.7% (4)	42.4% (167)
Did not take FA	(11)	49.5% (195)

Table 3. Factors associated with folic acid intake in women attending ANC in northern Uganda

Factors	Folic Acid Intake			N (total)	χ^2
	Yes	No			
Knowledge of spina bifida	Yes	91 (45.5)	41 (21.1)	132 (33.5)	26.24*
	No	109 (54.5)	153 (78.9)	262 (66.5)	
Availability of drugs	Yes	168 (87.0)	121 (63.7)	289 (75.5)	28.22*
	No	25 (13.0)	69 (36.3)	94 (24.5)	
Health education	Yes	52 (25.7)	13 (6.6)	65 (16.3)	26.59*
	No	150 (74.3)	183 (93.4)	333 (83.7)	

* $p < 0.001$

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3.2 Interdisciplinary care for children with spina bifida in East and Southern Africa³

Abstract

In this article the International Federation for Spina Bifida and Hydrocephalus (IF), reflects on its findings of 20 years of work in East and Southern Africa.

The prevalence of newborns spina bifida and hydrocephalus in Low and Middle Income Economies (LMIE) is higher than in High Income Economies (HIE). Resources to care for children with spina bifida and hydrocephalus are limited in LMIE. Medical and local materials have to be used creatively and have to be combined with pragmatic choices in surgery and medical imaging. Active parent involvement in care and interdisciplinary approach are crucial for a successful long life care program. The IF program shows how a costs-saving approach is feasible and does not need to imply a decrease in quality of care.

³ Based on: Mertens, P. and Bannink F. (2012) Interdisciplinary care for children with spina bifida in East and Southern Africa. *Journal of Medicine and the Person*, 10, 3: 129-135. DOI 10.1007s/12682-012-0126x

Introduction

This article describes the journey of IF to improve care for children with spina bifida in East and Southern Africa in collaboration with local hospitals and community based rehabilitation programs.

Spina bifida is a Neural Tube Defect (NTD), a congenital disability whereby early in pregnancy, the spinal cord is not closed properly in one or more places. This causes some degree of paralysis below the lesion and in 66% progressive hydrocephalus (Warf et al., 2009). Depending on the height of the lesion children with spina bifida are either able to walk with or without aids or they have to use a wheelchair. In 90% of children causes the paralysis urinary and faecal incontinence (Verpoorten & Buyse, 2008).

Hydrocephalus develops when the balance between production and absorption of cerebrospinal fluid (CSF) is disturbed. This can also occur apart from spina bifida, as a result of an obstruction caused by tumours, infections or a cerebral haemorrhage. When a child develops progressive hydrocephalus, surgery is often needed to prevent secondary impairments. If not, the brain may be damaged. The excessive CSF is drained to the abdomen by placing a subcutaneous silicone tube (drain or shunt). While shunting was the most common treatment over the past decades (Thompson, 2009), it is today possible to create a by-pass, without using a shunt, by endoscopic surgery like Third Ventriculostomy (O'Brien, Javadpour, Collins, Spennato, & Mallucci, 2005; Warf, 2005a). In infants with myelomeningocele the cele has to be closed surgically in order to prevent infections.

The prevalence at birth of spina bifida in Australia, Europe, Northern and Latin America has decreased due to the use of folic acid through fortification of staple food or the daily intake of supplements (Miles, 2006), and antenatal screening, resulting in termination of pregnancies (Frey & Hauser, 2003). Nevertheless the estimated incidence of spina bifida in HIE is still 0.5 – 1.000 per live births (Shaer et al., 2007), for infantile hydrocephalus 0.48, and for hydrocephalus associated with myelomeningocele 0.18 (Persson et al., 2007).

The incidence of spina bifida in low and middle income economies (LMIE) seems to be higher than in high income economies (HIE): a hospital based study shows 2.61 per 1000 live births in Tanzania (Kinasha & Manji, 2002), 5 per 1000 live birth in northern China (Zhu & Ling, 2008), and 7 per 1000 deliveries in the middle belt of Nigeria (Airede, 1992). In the IF projects of East Africa 67% of the children have hydrocephalus without spina bifida (IFSBH, 2011). Warf and Campbell (2008) attribute the high incidence of infantile hydrocephalus in Uganda to the high incidence of neonatal infection and poor treatment of postnatal meningitis (Warf & Campbell, 2008).

Children with spina bifida and / or hydrocephalus in LMIE often come too late for treatment because parents are faced with many barriers (Alatise, Adeolu, Komolafe, Adejuyigbe, & Sowande, 2006).

The last decades there has been tremendous progress in the care and treatment for children with spina bifida and hydrocephalus in HIC. The care for these children in LMIE has to translate this

knowledge into a realistic and affordable program that reaches the target group. The IF program builds on the solidarity of persons with spina bifida and hydrocephalus in High Income Economies (HIE) with their peers in LMIE, and aims to find low cost sustainable solutions.

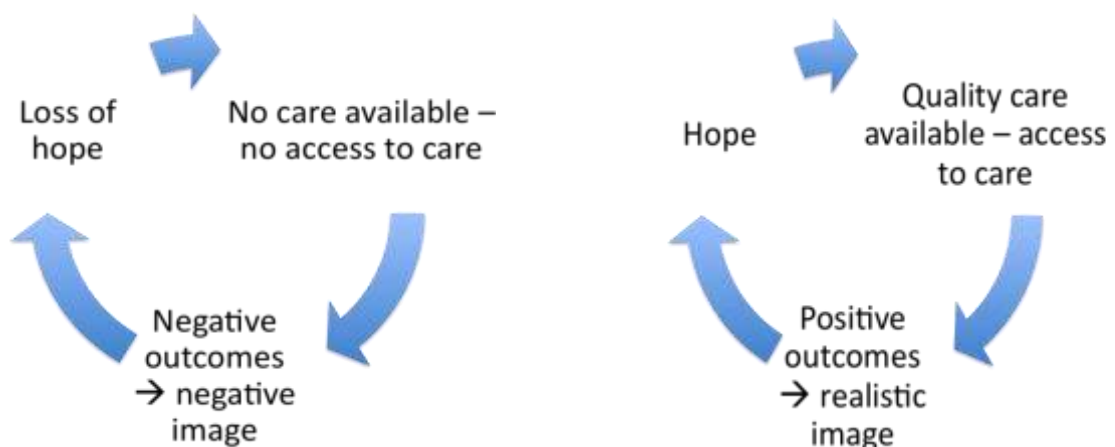
Materials and methods

IF started working in East Africa in 1992, in Kenya. The IF program then consisted of training co-workers of the Lilian Foundation, a Dutch Non Governmental Organization (NGO) working for children with disabilities in Africa.

Over the years the program expanded to 7 East African countries and 20 local and international partners. IF's program is ongoing, and currently consists of support to partner organisations' key staff, medical supplies, training of all involved in care (from the patient to neurosurgeons), primary prevention activities, and support to youth and parents support groups. IF and its partners developed training tools on continence management of the bladder and bowel and wound care which can be downloaded for their website.

The IF program in LMIE fights against the negative perception on these disabilities, by changing the negative vicious cycle in which the absence of care leads to a bad outcome and reinforces the negative stereotypical thinking on spina bifida and hydrocephalus. By investing in good care a virtuous cycle is created where good care leads to a positive outcome, a better quality of life and hope. Figure 1 illustrates the vicious and virtuous cycles.

Figure 1. The vicious and virtuous cycles of care, hope, and outcomes



IF works with persons with spina bifida and hydrocephalus: some of the first generation adults with spina bifida work as paid staff members in the IF projects. They are role models for children and create hope for parents and youth.

All IF's activities are implemented through government and private health and rehabilitation facilities in collaboration with the district local government, private non for profit health care providers (e.g. missionary hospitals and health centers), and Community Based Organisations.

A review of the various components of the program was carried out to describe successes and challenges of the past 20 years. IF progress reports and interview data from partner organizations were analyzed. As well literature published over the years on the provision of care for children with spina bifida in LMIE was reviewed, and analyzed.

Findings were grouped into the following thematic categories: self-help groups, creative use of medical and locally available materials, interdisciplinary care, and surgical choices.

Results

Beginning at the end of the chain – self help groups

In LMIE the healthcare system consists of a long chain of stakeholders, who all have their own priorities and specialties. They all are in place to serve the ultimate user, the so-called 'patient' who is too often at the end of this chain. Being at the end of the chain, the services available often do not reach or do not serve the needs of the patient. 'Beginning at the end of the chain' became the IF's motto for its projects in LMIE. The IF-network, being an impairment specific user organization, knows the barriers, the needs and possible solutions based on their own experience.

Parents in East and Southern Africa face many barriers before they find help for their children. Breaking down these barriers has to be part of the care program and therefore the expertise of the target group itself is crucial. Earlier publications have shown that involving families in the care for children and youth with chronic diseases is the key to success of the health services received. Kieckhefer & Thrans (2000) advocate for merging models of practice to provide a guidance approach that assists a child to develop into an independent, healthy functioning adult (Kieckhefer & Trahms, 2000). This is supported by Gall et al (2006) who advocate for a healthy dynamic relationship between health care providers, parents, and children, in which the roles of the various players change over time as the child grows and develops. The leadership for care is herein transitioned from the health care provider to the parents and in turn to the young person as developmentally appropriate (Gall, Kingsnorth, & Healy, 2006).

IF aims to actively involve patients and parent groups in its projects. The children, youth and adults are the experts who are directly involved in the design, implementation and evaluation of programs. They are partners in all IF's activities.

It is crucial to engage patients as partners in the development of understandable information for other patients. When health service users are involved in developing information materials, the

relevance, readability, and understandability of the materials increases (Nilsen, Myrhaug, Johansen, Oliver, & Oxman, 2006).

The need for correct and up-to-date information was the reason why active parents in HIE started self-help groups for spina bifida over 40 years ago. IF is the worldwide umbrella organization of all these self-help groups and encourages the foundation of new groups also in LMIE's. In all IF projects, IF facilitates this peer exchange because parent groups have shown to be beneficial in the creation of sustainable lifelong care.

Parents support groups in East and Southern Africa have evolved into active lobbying and capacity building associations, that identify, advise and support other parents, while also sensitizing communities, health workers, local and national leaders, teachers and others. The majority of members are mothers who are the primary caretakers of the children: 72.4% of these mothers are married or co-habiting, 17.2% were abandoned by their spouses after the birth of the child with spina bifida and/or hydrocephalus (IFSBH, 2011). From interviews we learned that most parents take part in the groups because they learn from the other parents and feel supported.

Less but better is also cost effective – creative use of medical and local materials

Although sometimes high-tech medical interventions are performed in LMIE's, the basic care and tools are often not available for the large majority of the population. *Where there is no doctor* is the title of David Werner's book on health care in developing countries (Werner, 1992). Werner explains clearly and with practical examples how a different environment requires a different approach. Specifically to improve care for children with disabilities he wrote *Disabled Village Children* explaining what can be done for children with disabilities in different social settings, illustrated by simple drawings, instructions and informative text (Werner, 1987).

IF builds on this basic information Werner developed and adds specific information for children with spina bifida and hydrocephalus, e.g. on continence management, as illustrated in figure 2.

Every extra step in care may be the barrier which prevents a child from eventually receiving the required treatment. For example: a surgeon trained in HIE may hesitate to perform surgery without all medical imaging he had at his disposal during his training. A surgeon who requires a costly CT scan before shunting may lose the child by this request, as many parents will not be able to afford this scan or will need time to collect sufficient funds for this. Clinical signs and measurement of the head circumference well recorded on a chart combined with ultrasound results have proven cheap alternatives for a computed-tomography (CT) or magnetic resonant imaging (MRI) scan (Munyi, Poenaru, Bransford & Albright, 2009). The use of simple measurements techniques is illustrated in figure 3a and 3b below.

Figure 2: Clean Intermittent Catheterization poster

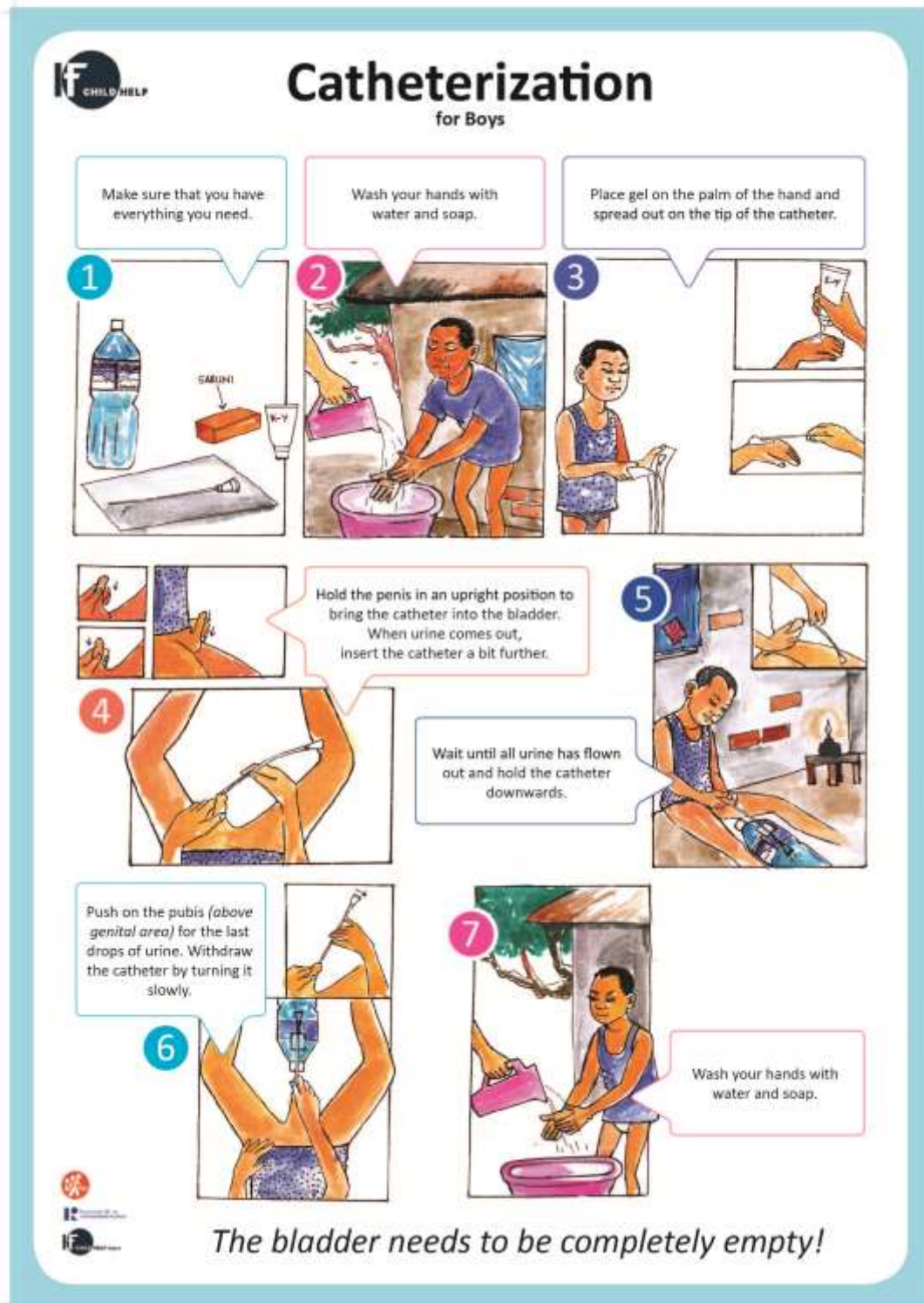
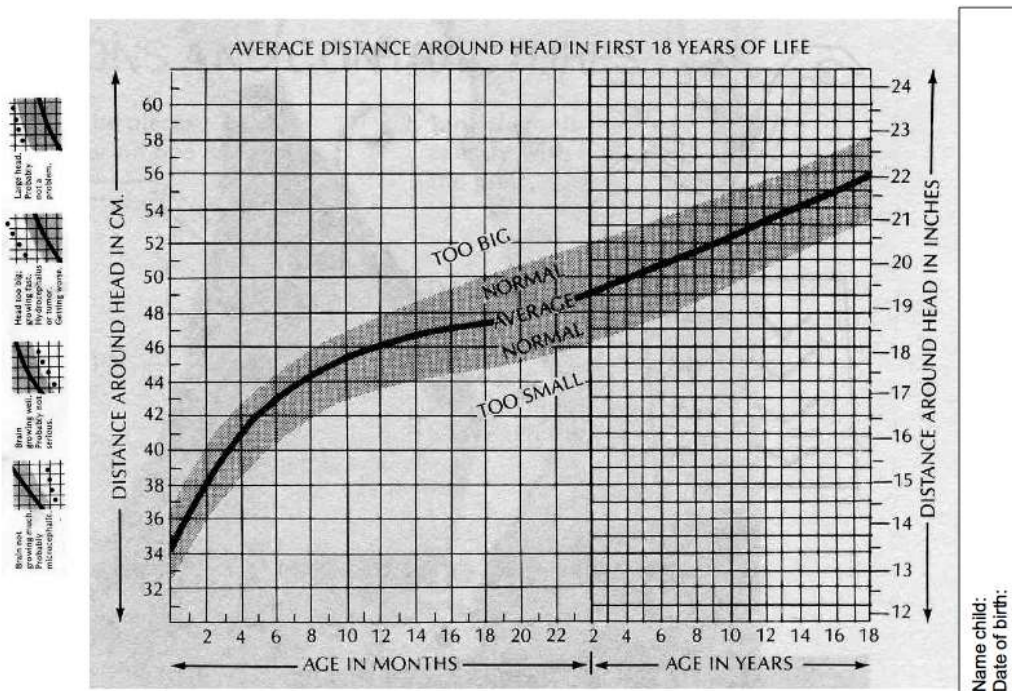


Figure 3a. Measuring head circumference with a measurement tape



Figure 3b. Chart used to register the head circumference over time



Shunts manufactured for High Income Economies are not an option in LMIE. In some countries, like Malawi, Zambia and Zimbabwe, creative surgeons developed shunts themselves (Malawi and Harare shunts) and in Sudan surgeons created a subcutaneous reservoir between the skin and the skull, in order to regularly drain the excessive fluid (Adeloye, 2012). Many children died from the complications of these self-made aids, but some benefited from it.

IF's work in LMIE got a real push after a congress in Japan, where an Indian professor informed IF about the Chhabra shunt system, a cheap, but effective Indian shunt by the producer Surgiwear, which IF could export to Africa. Initially many surgeons in Africa refused to use the Chhabra shunt. They stated that their patients deserved the same quality of shunts in high income countries. Therefore, IF asked Prof. Benjamin Warf to do a comparative study between the Codman Shunt and the Chhabra shunt in Uganda. Warf did not find any statistical relevant difference between both shunts (Warf, 2005). Only after publication of the results, IF was able to convince its partners in LMIE to use the inexpensive shunt. To date IF donated 23,000 shunts to its partners and projects.

For continence management early diagnosis and early institution of adequate medical treatment are crucial (Verpoorten & Buyse, 2008). Clean intermittent catheterization combined with anticholinergics (oral or intravesical) is the standard therapy (Bauer & Joseph, 1990). Verpoorten and Buyse (2008) reported this treatment as "effective in preserving renal function and providing safe urinary continence in more than 90% of patients with a neurogenic bladder". This treatment, if started early, can prevent both renal damage and secondary bladder-wall changes, thereby potentially improving long-term outcomes (Verpoorten & Buyse, 2008).

In East and Southern Africa, IF uses catheters of 15 euro-cents a piece. The patient with a neurogenic bladder must be catheterized 4 to 5 times a day. In the North, catheters are packed in a sterile blister pack, lubricated or not, and sold as a single-use product at 3 euros a piece. However Schlager et al (2001) showed that "using a new, sterile catheter does not decrease the high frequency of bacteria in patients with neurogenic bladder on intermittent catheterization" (Schlager, Clark, & Anderson, 2001). Emptying the bladder entirely is more important than using a new and sterile catheter every time.

Therefore in East and Southern Africa, IF uses the same catheter 5 times a day for three months, at a total cost of 15 euro-cents. Mothers are trained how to keep the catheter clean. Training is started early to preserve kidney function (Dik, Klijn, van Gool, de Jong- de Vos, van Steenwijk & de Jong, 2006). In Europe the equivalent cost is 1,350 euro per child per month.

IF's physicians developed a technique that enables the determination of the type of bladder, the pressure in the bladder and the plan of treatment, without expensive urodynamic investigations. A well-trained nurse under supervision of a physician, a simple ultrasound machine, a motivated mother, a feeding tube, a measuring-tape and a registration form are the only requirements for the investigation.

For the continence management of the bowel, IF is training parents, children and adults to do bowel wash outs with locally produced equipment and simple local toilets, as depicted in figures 4a and b.

Figure 4a. A self-made wash-out with a plastic bottle for bowel management.



Figure 4b. Self-made toilet for bowel washouts



IF advocates for basic cost effective care for all children with spina bifida and hydrocephalus. In countries where this basic care is not available to all, IF is questioning the promotion of expensive high tech medicine like nerve transplants, fetal surgery and the use of expensive shunts. IF and its partners developed tools and protocols for LMIE ensuring the needed standards of care.

Interdisciplinary approach or SHIP

Surgery only opens the door towards a decent life with spina bifida and hydrocephalus. Opening the door to life implicates starting medical, nursing, educational and other interventions. After surgery, children are going back to their homes, often located far from the hospital. Follow-up care is needed at a reachable place for the parents. In LMIE, a network of community based rehabilitation (CBR) programs have been developed. Mostly CBR consists of visiting the families at home and inform and teach the parents how to cope with day to day challenges their child faces. The facilities offering CBR are hosting outreach clinics organized by the referral hospitals.

The Cure Children Hospital of Uganda (CCHU) Hospital in Uganda, one of IF's most important partners, looked at the factors affecting survival of infants with spina bifida (Warf, Wright, et al. 2011). They found that the mortality was lower, even approaching that of their unaffected peers in districts with CBR programs.

In HIE countries, many university hospitals created a multidisciplinary team for spina bifida and hydrocephalus (Hoeman, 1997), including the related physicians, nurses, psychologists and other professionals. These teams reduced the number of medical interventions and complications through training parents and children in preventive measures, avoiding secondary complications like infections, pressure wounds, kidney damage and indirectly social exclusion.

This concept is translated in the IF program under the name SHIP. SHIP stands for S(spina bifida) H(hydrocephalus) I(Interdisciplinary)P(Program). Like on a ship, all stakeholders have to cooperate adequately to bring the passenger to his destination. In LMIE, the different stakeholders in care are spread over the country and distances create barriers. That's why coordinating meetings are needed and communication tools like a SHIP-passport where care activities are recorded are developed. This passport is owned by the patient.

IF's network is training all those involved: the children and their parents, the nurses, pediatricians and neurosurgeons. IF also provides the necessary (medical) material with the trainings.

Nowadays children with spina bifida and hydrocephalus have a normal life-expectancy. The more opportunities they get, the more self-confidence they develop, enabling to deal with most of their problems and leading a meaningful life. In Kenya, Uganda, and Zambia the first generation of these children are attending university. Nevertheless in LMIE, their situation is still challenging and inclusion in education is limited.

Surgical interventions

Although new-borns with spina bifida usually need surgery after birth, some children in LMIE are doing well even without surgery. Warf (2011) showed that 41% of children with hydrocephalus and spina bifida in Uganda do not need surgery for hydrocephalus (Warf, 2011). IF sometimes meets adults with spina bifida who survived without any medical intervention. The back closed spontaneously and the hydrocephalus did not develop progressively. Most of them have a larger head circumference but can cope well and have good cognitive development.

For the ones who do require surgery, IF's program in East Africa demonstrated that it is possible to provide high quality of surgical care for children within LMIE, but shunt complications in this environment are still life threatening. IF asked Dr Warf to investigate the treatment of hydrocephalus with Endoscopic Third Ventriculostomy (ETV), and equipped him with the required material in Uganda. While Warf showed that ETV was the preferred surgical procedure in older children and adults with aqueduct obstruction, he initially questioned the value of ETV in infants. ETV successfully treated 77% of those with post-infectious hydrocephalus and 89% of those with hydrocephalus of other etiologies for children over 12 months of age. For infants under one year of age the success ranged from 30 to 50% and 70% in the post-infection aqueduct obstruction (Warf, 2005). However when Warf combined the ETV technique with Endoscopic Choroid Plexus Cauterization or CPC, the results improved enormously and 3 in 4 children were helped with this combined technique (Warf, 2008). CPC is cauterizing part of the tissue that produces the cerebro spinal Fluid (or CSF).

In IF's projects in Uganda 60% of the children with hydrocephalus now receive ETV/CPC rather than a shunt placement (Warf, Stagno, Mugamba, 2011). This operating technique can, in absence of CT scan or MRI, serve as a tool for diagnosis at the same time. If a shunt appears necessary, it can sometimes be placed during that very same operation.

ETV/CPC and shunting requires skilled and trained neurosurgeons. Cure International and IF organize ETV/CPC training for neurosurgeons of LMIE in Uganda. Already 16 neurosurgeons from 13 different countries were trained in ETV and CPC and provided with the required material to perform surgeries in their countries after completion of the training.

Neurosurgical intervention in LMIE has proven not only to generate a healthy, but also long-term economic benefit (Warf, Alkire, et al, 2011).

Discussion

Through low cost interventions and involvement of parents, children with spina bifida can access good care in LMIE. Building onto rather than replacing public health care services is key to create long term sustainable solutions. Currently most of the care for children with spina bifida and hydrocephalus is provided by non for profit and faith based hospitals and organizations.

IF and its partners like Cure International in Uganda and Zambia and Bethany kids Kenya have not the ambition to take over the healthcare system, but proved that it is possible to provide decent care for our target group in LMIE. To ensure sustainability and build capacity in the public health system Cure International and IF trained neurosurgeons from governmental hospitals e.g. Dar es Salaam (Tanzania).

Finding, but also questioning solutions is a permanent process. It is only possible with the active involvement of the target group. IF aims to personalize the treatment for children with spina bifida and hydrocephalus, avoiding unnecessary expensive and time consuming treatment. All this is only possible through participation. Parent groups are active lobbyists to their government and local authorities for good care and an inclusive society.

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Chapter 4 Social inclusion and belonging



4.1 'I like to play with my friends': children with spina bifida and belonging in Uganda⁴

Abstract

This paper describes experiences of living and belonging from the perspectives of Ugandan children with spina bifida and their siblings and parents.

We explored belonging at micro, meso and macro level taking into consideration African Childhood Disability Studies, central concepts of family, cultural conceptions of disability, poverty, and the notion of 'ubuntu', and using child-friendly culturally adjusted interview methods including play.

Whilst children with spina bifida had a strong sense of belonging at household level, they experienced more difficulties engaging in larger social networks, including school. Poverty and stigma were important barriers to inclusion.

We propose strengthening the network at family level, where the environment is more enabling for the children to find a place of belonging and support, and expanding investment and awareness at community and national level.

⁴ Based on: Bannink, F., Idro, R., and van Hove, G. (2016) 'I like to play with my friends: children with spina bifida and belonging in Uganda. *Social Inclusion*, 4 (1). DOI: 10.17645/si.v4i1.630.

Background

African Childhood Disability Studies and Conceptions of Disability

Disability studies in sub-Saharan African countries have largely focused on adults with disabilities and caregivers of children with disabilities, and have primarily been conducted in South Africa. Children's narratives are absent in most disability literature (Curran & Runswick-Cole, 2014), and more so in African studies. Disability studies have argued for greater awareness and appreciation of diverse understandings of disability in low resource settings and highlighted the need for a different discourse (Grech, 2009; Meekosha, 2011; Seligman & Darling, 2009; Whyte, 1995).

Schalock (1997) argues that in cultures with an interdependent focus, acceptance in the group is a more significant contributory factor to quality of life than in independence or individualistically oriented cultures. Chataika and McKenzie (2013) build further on this, and explain that care and belonging may have a more prominent place than formal education and independence in the lives of African children with disabilities (Chataika & McKenzie, 2013). They suggest it is important that African Childhood Disability Studies explore family and cultural conceptions of disability, poverty, and the notion of 'ubuntu' ('I am because we are' or 'humanity to others').

Perceptions of impairment and disability are social phenomena subjected to 'substantive temporal, cultural and situational variation' (Ingstad & Whyte, 1995). Social-anthropological and historical studies have described cultural and belief systems of disability in Africa (Braathen & Ingstad, 2006; Devlieger, 1998; Ingstad, 1999; Miles, 2002, 2004). Findings show a complex range of cultural concepts including the child being 'cursed', 'bringing misfortune', being a 'gift' or presenting a challenge to the family (Devlieger, 1998; Franzen, 1990; Wright, 1960). Chataika and McKenzie (2013) describe the complexity of disability concepts in Southern Africa, and point out how they are strongly associated with spiritual understandings of the nature of disability (Chataika & McKenzie, 2013).

In Uganda, the focus of this study, most dialects lack a single word that translates into the English word "disabled"; however all descriptions combine the notion of physical limitation and powerlessness (Lwanga-Ntale, 2003). The definition of disability in Ugandan laws and policy documents is not harmonized. The Persons with Disabilities (PWD) Act 2006 defines disability as 'a substantial functional limitation of daily life activities caused by physical, mental or sensory impairment and environmental barriers resulting in limited participation'. By recognizing that disability is the result of the interaction between impairment and external barriers, the PWD Act aligns the legal definition of disability in the Ugandan law to that enshrined in the Convention on the Rights of Persons with Disabilities (CRPD), which Uganda ratified in 2008 (Enable, 2015). However unlike the CRPD, the PWD Act requires that disability be substantive, which has implications for disability rights at a practical level (Ojok & Wormnæs, 2013). The Uganda Bureau of Statistics (UBOS) estimates 12.5% of the population in Uganda lives with a disability (UBOS, 2014). Discriminatory attitudes and behaviours, a gap in implementation of the regulatory framework, lack of coordination

between government and civil society, and a fragmented programmatic approach all challenge the implementation of the CRPD in Uganda (UNICEF, 2014).

Children with Spina Bifida in Uganda

Spina bifida is a neural tube defect, a congenital abnormality causing disability, whereby the spinal cord and vertebrae do not form completely and the neural tube fails to develop normally. Worldwide incidence of spina bifida varies between 0.17 and 6.39 per 1000 live births (Bowman, Boshnjaku, & McLone, 2009; Kinasha & Manji, 2002; Msamati, Igbigbi, & Chisi, 2000; Shaer, Chescheir, & Schulkin, 2007). Incidence and prevalence rates in Uganda may be higher due to inadequate folate consumption by pregnant women (Bannink, Larok, Bauwens, Kirabira, & van Hove, 2015; Whyte, 1995) lack of pre-natal care (Miles, 2002), absence of secondary prevention services (Frey & Hauser, 2003), and higher exposure to environmental risk factors such as dioxins (Safi, Joyeux, & Chalouhi, 2012) and fumonisins intake (Hendricks, 1999; Marasas et al., 2004; Wild & Gong, 2010). Although Warf et al. estimate that 1,400 children are born with spina bifida in Uganda annually (Warf, Wright, & Kulkarni, 2011), no national data are available.

Most children with spina bifida have some degree of paralysis, which affects their mobility as well as bowel and bladder control (Northrup & Volcik, 2000; Verpoorten & Buyse, 2008). Sixty-six per cent of children with spina bifida in low-income countries develop hydrocephalus (Warf & Campbell, 2008). In Uganda, concepts describing children with spina bifida vary by region. Descriptions often refer to the physical appearance of the child, e.g. 'swelling on the back' (*ekizimba mu mugongo* in the Central Region), 'the one with a split spine' (*owacwekire orukizi* in the Western Region) or to the secondary impairments a child may have, e.g. 'the one smelling of urine' (*langwece* in the Northern Region) for children with non-managed incontinence problems, or 'the one with the big head' (*baana be gimitwe migali* in the Eastern Region) for those with hydrocephalus (Bannink, Stroeken, Idro, & Van Hove, 2015).

Health and Rehabilitative Care

Most children born with spina bifida need surgery to close their back in order to prevent infections. Children with spina bifida and progressive hydrocephalus also often need surgery to prevent secondary impairments (IFSBH, 2014). Children with spina bifida experience mobility challenges and incontinence which affect their participation in daily activities, and require the use of assistive devices and continence management (Abresch, McDonald, Widman, McGinnis, & Hickey, 2007; Andren & Grimby, 2000; Danielsson et al., 2008; Jansen, Blokland, de Jong, Greving, & Poenaru, 2009).

Surgery and rehabilitative care is expensive and inaccessible for many children born with a disability in Africa. Their families are often their main source of care and protection (Guyer, 1981). Families living in poverty often struggle to find resources to provide this care and protection (Miles, 2002) as

external and government support is limited (van der Mark & Verrest, 2014). In 1990, the Government of Uganda adopted Community Based Rehabilitation as a health service strategy to reach more persons with disabilities (NUDIPU, 2007). This strategy is still in place, but the efforts that are being made to provide basic services at community level for children with spina bifida are mainly largely remain funded by international donors and charities (Mertens & Bannink, 2012).

At the time of this study, the initial surgery (closure of the spine) in Uganda was only available in two public government funded hospitals (Mulago National Referral Hospital in Kampala and Mbarara Regional Referral Hospital), and one private specialized neuro-paediatric hospital (CURE Children's Hospital) in Mbale, eastern Uganda. In north, west, and central Uganda three rehabilitation centers, funded by international donors and charities offered occupational therapy, physiotherapy, continence management, and social support services for children with spina bifida and their families.

Education

A special needs education department was created in the Ministry of Education in Uganda in 1973 (USDC, 2003). To date, government policies continue to promote special needs education and the establishment of special schools rather than inclusive education for children with intellectual disabilities (UNICEF, 2014). In a study on parental stress of 139 parents of children with spina bifida in Uganda, we found that more than half of the children with spina bifida were going to mainstream schools, none were going to special schools. Inclusion in these schools remained limited, with parents reporting discrimination, exclusion, and lack of services to manage incontinence at school for their children (Bannink, Idro, & Van Hove, 2016). Challenges with inclusion have been reported earlier in South Africa (Chataika, McKenzie, Swart, & Lyner-Cleophas, 2012). Disability grants are available for families with a disabled family member in South Africa. While these grants helped financially, they were not found to improve the education or employment outcomes of persons with disabilities (Loeb, Eide, Jelsma, Toni, & Maart, 2008).

Poverty

Persons with disabilities typically live in poorer than average households (Emmett, 2006; Filmer, 2008; Lwanga-Ntale, 2003). Palmer further explains that most studies examine the income poverty rate of persons with disabilities, without taking into account the additional expenditure they incur which are attributable to their disability (Palmer, 2011). Looking after a child with spina bifida increases financial costs for families in the form of medical treatment, rehabilitation, and transport. It also has an indirect cost, by reducing the amount of time a parent is able to devote to income-generating activities, as many are directly involved in rehabilitation activities such as continence management. The monthly income of families in our study ranged from \$28 to \$689 with a median of \$82 (income derived from all sources, including wages, market sales, cattle, land and other assets). This is much lower than the total national average of \$156 (converted from Ugandan

Shillings), though closer to the average rural income of \$112 and related regional variations (UBOS, 2014). We found that looking after a child with spina bifida increased financial costs for families in terms of medical treatment, rehabilitation, and transport. Living in poverty may also have increased the risk of having a child with spina bifida in the first place, as the mother was unlikely to attend antenatal care, eat foods rich in folic acid or take supplements before and during her pregnancy, all of which could have prevented spina bifida.

In this paper we describe experiences of belonging and being of children with spina bifida and their families living in Uganda. Themes of belonging at micro, meso, and macro level were explored, taking family, friends, health care, school, and socio-economic factors into consideration.

Methods

Ethical approval and research clearance were obtained from Ghent University, Belgium, the Uganda Virus Research Institute, and the Uganda National Council for Science and Technology. Informed consent was obtained from all parents, and assent from children and siblings of 8 years-old and above where possible. Consent forms were translated into the local languages and discussed and agreed with the participants, with assistance of a translator if the participant or child did not speak English.

In total, 139 families were recruited and 139 parents, 97 children with spina bifida, and 35 siblings between 4 and 14 years of age were interviewed. Due to lack of contact details, children could not be traced from medical files and databases of CURE and Mulago hospitals as initially planned. Therefore, purposeful sampling was used in Mbarara, Kampala, and Mbale where CURE holds bi-monthly clinics. CURE hospital and the partnering rehabilitation centers in Kampala and Mbarara were requested to list the children registered with spina bifida in their follow up programs, and inform them of the study during home visits and during reminder clinic attendance telephone calls. In Gulu and Lira, where no follow up system or registry of the children was in place at the time, radio announcements were aired to inform parents of the forthcoming review clinic in the area. The announcements specifically invited parents of children with spina bifida and hydrocephalus between the age of 4 and 14 years to attend.

Qualitative semi structured interviews, observations, and quantitative functioning scales measurements were combined and administered to 100 families at the clinic and 39 families at home between June 2011 and December 2014. The siblings interviewed were recruited during home visits, purposefully selecting the sibling closest in age to the child with spina bifida in the household. Observations were carried out during home visits and during clinic days. The clinic observations were held at least twice in each of the five sites during review days held specifically for children with spina bifida and hydrocephalus.

The semi-structured interviews contained questions about the child's family, home setting, their typical day, what they like and dislike, friendships, school, and health care. Ugandan-made dolls, drawing paper and colours for children were used during the interviews to help children open up and

narrate their stories. E.g. when asking children about their family, children were encouraged to select a doll for each family member or draw their family and talk about them. The same approach was used to help children narrate stories about what other people would say to them e.g. at school, and to act out or draw situations which they enjoyed or disliked. The children enjoyed the interaction and were keen on visually demonstrating their family members and school setting.

In addition to the semi structured interviews, the Vineland Adaptive Behaviour Scales (VABS) Daily Functioning and Social Skills Sub Scales were administered by interviewing the parents and observing the children's abilities.

The interviews with the children, siblings, and parents were held in the local language of the area, and a translator was hired and trained to assist in conducting the interviews, and observations. Some of the interviews were conducted in English, if parents or children were fluent and requested it.

The semi-structured interviews were not audio recorded, but answers were written out during the interviews and transcribed upon completion. Family set up and drawings were noted and kept. The interview data were analysed by thematic coding and analysis of the manuscripts using NVivo10. In this process, themes were identified representing experiences of children with spina bifida.

Handheld records for the questions of the VABS subscales were kept during interviews, and entered into a database after completion. The sub-total scores for each subscale were calculated to compare means of the scores of the children with spina bifida and their siblings using SPSS16. Observations on interactions and non-verbal communication were noted and checked with the text transcribed.

Results

Demographics

A total of 97 children, 35 siblings, and 139 parents out of 139 families with a child with spina bifida were interviewed. Table 1 describes the demographics of the study population. The mean age of the 82 male (59%) and 57 female (41%) children with spina bifida was 6.04, ranging from 4 to 14 years of age (SD=2.01). 55.9% of them were going to school: 36.8% (50) in nursery school, 16.2% (22) in primary school, and 2.9% (4) in secondary school.

Table 1. Demographic characteristics of the study population (N=139 children, N=35 siblings)

Variable	Child	%	Sibling	%
<u>Gender child</u>				
Male	82	59.0%	14	40.00%
Female	57	41.0%	21	60.00%
<u>Child is schooling in</u>				
Nursery school	50	36.8%	0	
Primary school	22	16.2%	9	25.70%

Secondary school	4	2.9%	26	74.30%
Not schooling	60	44.1%	0	
<u>Type of disability</u>				
Spina bifida	76	54.0%	0	
Spina bifida and hydrocephalus	64	46.0%	0	
<u>Location / region</u>				
Central	65	46.8%	35	100%
East	26	18.7%	0	
West	29	20.9%	0	
North	19	13.7%	0	
<u>Religion</u>				
Christian	101	72.6%	32	80.0%
Muslim	26	18.1%	6	17.1%
Other	13	9.3%	1	2.9%
<u>Relationship parent</u>				
Mother	105	75.5%	26	74.3%
Father	17	12.2%	6	17.1%
Grandmother	10	7.2%	2	5.7%
Other	7	5.1%	1	2.9%
<u>Education level parent</u>				
None	6	4.5%	3	8.6%
Primary	75	56.4%	22	62.9%
Secondary	29	21.8%	6	17.1%
Vocational	12	9.0%	1	2.9%
University	11	8.3%	3	8.6%
<u>Marital status parent</u>				
Single	12	8.8%	0	
Married	103	75.2%	26	74.3%
Separated	11	8.0%	6	17.1%
Widowed	11	8.0%	3	8.6%
<u>Monthly household income</u>				
< 30 US dollar	25	18.8%	9	25.7%
30 - 60 US dollar	31	23.3%	10	28.6%
61 - 90 US dollar	28	21.1%	7	20.0%
> 90 US dollar	49	23.3%	9	25.7%
<u>Occupation parent</u>				
Finance / administration	4	3.0%	2	5.7%
Small scale private business	30	22.6%	7	20.0%
Teacher / education	8	5.6%	3	8.6%

Medical / paramedical	4	2.6%		
Technical / transport	10	7.1%	1	2.9%
Civil service / government	3	2.3%		
Peasant farmer	62	46.6%	19	54.3%
No occupation	13	9.8%	3	8.6%

Parents' ages ranged from 25 to 49 years, with an average age of 35.7 (SD 5.5). Mothers constituted the majority of the parental respondents, followed by fathers and grandmothers. Almost half of the respondents were farmers.

Of the 35 siblings interviewed, 40% were male and 60% female. The median age was 7.8, ranging from 4 to 14 years of age (SD=2.53). All of them were schooling; 25.7% (9) in primary, and 74.3% (26) in secondary school.

Household size ranged from 2 to 13 with an average of 8 people per household. The average monthly household income was \$82.

The majority (96.4%) of the 139 children with spina bifida in the study had undergone surgery to close their spine (myelomeningocele closure) earlier in life. Of the 64 children who had both spina bifida and hydrocephalus, 23 (35.9%) had undergone endoscopic third ventriculostomy while 12 (18.6%) had ventriculo-peritoneal shunts placed. Only 5 (3.6%) of all children in the study never had surgery. Most of the children (127 or 91.4%) received rehabilitation services such as physio- and occupational therapy.

Of the 139 children studied, 136 (97.8%) could sit unaided and 64 (48.9%) could walk unaided. Of the 75 who could not walk unaided, 23 (30.6%) used a wheelchair, the same percentage used crutches, and 5 (0.7%) used other aids. In total 23 (16.5%) used no aids and crawled.

Over 85% of the children in our study were incontinent. Table 2 shows percentages of children using clean intermittent catheterization (CIC) and bowel management to manage their incontinence.

Table 2. Incontinence in children with spina bifida in Uganda (N=139)

Variable	Yes	No
Child is continent of urine	15 (10.9%)	122 (89.1%)
Child uses catheterization	100 (78.2%)*	27 (21.3%)
Child is continent of stool	18 (13.1%)	119 (86.9%)
Child uses bowel management	92 (76.7%)**	28 (23.3%)

* 22 (22%) practices CIC without assistance of another person

** 7 (7.6%) practices bowel management without assistance of another person

Table 3 shows parents' ratings on the Vineland Adaptive Behaviour subscales. Daily functioning tasks such as removing a jumper, drinking from a cup, face washing and hair brushing were achieved by most of the children, while other tasks which involve more movement were more challenging, e.g. fetching water and dressing independently (including trousers/skirts and shoes). Compared to siblings (N=35) in the same age group, the scores were significantly lower with a mean total subscale score of 13.9 (SD 5.1) for the children with spina bifida and 19.2 (SD 2.11) for their siblings ($p < 0.001$). Social skills subscale scores of children with spina bifida were slightly but not significantly lower (11.49, SD 1.8) than those of siblings (11.97, SD 0.2).

Table 3: Vineland Adaptive Behaviour Daily Functioning and Social Skills Subscale Outcomes for children with spina bifida and siblings in Uganda (means, SD, and t-test)

Variable	Children with SB	Siblings	Difference
Daily functioning (Vineland) N=131	13.91 (5.1)	19.23 (2.1)	$t=6.03, p < 0.000$
Social skills (Vineland) N=133	11.49 (1.8)	11.97 (0.2)	$t=1.59, p=0.112$

My Home

When asked to describe their home, most of the children explained where they lived and who they lived with. In describing their families, most started by mentioning their parent(s), followed by their siblings and aunts or grandparents. Househelps were often referred to as 'aunts'. The majority of children also talked about their neighbours and physical environment.

"There is a mango tree in our compound. When it is mango season, we eat a lot of mangoes. We sit in the shade and play games. I like playing meso [a board game] with my neighbor." (14-year-old boy with spina bifida, Central Uganda)

"It's just me and my mum. We live in a hut. The neighbours are far. I sit outside under the tree when my mum is digging. Sometimes I do the dishes." (8-year-old boy with spina bifida and hydrocephalus, Northern Uganda)

"This is my home. It is on a hill. I like to play there with my sister [points in the direction of the corner of the compound]. My grandmother looks after us. Most of the time I stay home." (10-year-old girl with spina bifida during home visit, Eastern Uganda)

The majority of the children participated in some of the common Ugandan daily household activities, such as doing the dishes, and washing clothes: "I like to wash plates. My sister gets water and then I wash." (7-year-old girl with spina bifida, Central Region). Other common household activities were

more difficult for most, e.g. fetching water and sweeping the compound. Children with both spina bifida and hydrocephalus experienced greater difficulty in participating in daily household activities, as they tended to have relatively less developed motor skills.

“His head is too heavy, he cannot balance well, he easily falls or knocks things, he cannot help in the house.” (Parent of a 9-year-old boy with spina bifida and hydrocephalus)

Interactions observed during home visits showed how children are often allowed to participate and interact with others, but are not actively encouraged. Only a few parents made arrangements to improve accessibility in their compound, or encouraged their child to join others in play or household activities. Siblings often helped the children with spina bifida by doing household activities such as washing together. In two observations, a sibling brought a jerrycan with water, and poured water in the basin for the child with spina bifida, so that they could then help in washing the clothes. Not being able to go and fetch water on their own was something children expressed sadness about:

“I would like to go with my sisters and collect water, but it is not possible, they go and I am left at home, I cannot meet others on the road and talk about secret things like they do because I am always home.” (12-year-old girl with spina bifida, Eastern Uganda)

In some homes, children were neglected and not encouraged to participate in daily household activities and games. The majority of the children were found sitting on a mat or outside on the bare ground when visited at home. This was more common for those with both spina bifida and hydrocephalus, who had more severe cognitive and motor skills difficulties.

The games children with spina bifida participated in were often board games or swinging. Occasionally a child would participate in a ball or tap game while crawling. Mostly children with spina bifida would be observers if siblings were playing games which required more movement. In some cases, they would not be allowed to play outside, mostly when they had pressure sores or infections.

“I like to play outside but when I have a wound [pressure sore] I am not allowed to crawl and play outside, then I watch them from the house.” (7-year-old boy with spina bifida)

Family events such as family meetings and prayers, weddings, funerals and memorials are often referred to as ‘functions’ in Uganda are an important part of family life. When asked about these ‘functions’, children and siblings explained that they attended church functions, and occasionally weddings or funerals, depending on the location and distance they would have to travel. Single parents said they found it challenging to attend events together with their child with spina bifida as some felt stigmatized by relatives and others at the events.

“I went to the kwanjula [traditional wedding] of my aunt, it was here in the village, we enjoyed it.” (Child with spina bifida, Central Region)

“Sometimes our parents take us with them for functions, but if it is far we don’t go.” (Sibling of a child with spina bifida, Central Region)

“Our family buries near our home, when there is a funeral, we are around. If it is far we don’t go, I can’t carry her and people will talk.” (Mother of a child with spina bifida, Western Region)

My Friends

The majority of the children said they have at least one good friend, most of them said they have a number of friends, and play with neighbours and other children from their communities: “My best friend is Daniel [his neighbour, same age]. When he comes back from school we play together.” (5-year-old boy with spina bifida, Central Uganda, not schooling), or “My friends are Sheila [sister] and Mariam [neighbour], we make homework together” (12-year-old girl with spina bifida, Central Uganda).

About a fifth of the children interviewed say other children sometimes verbally abuse them: “Children say that my head is big like a pumpkin” (12-year-old boy with spina bifida and hydrocephalus, Western Region), or “In my community the children abuse me, they call me mulema [lame]” (10-year-old girl with spina bifida from the Central Region)

Most of the children were shy in speaking to others or initiating play when observed in their home, school and clinic setting. During interviews, parents expressed their concern that while their children had friends close by home, e.g. neighbours and siblings, they had difficulties socializing in school and the wider community: “My child doesn’t want to go far from home, they bully him about his big head.” Siblings commented that their brothers or sisters have friends, but that sometimes other children call them names because of their disability: “Children who don’t know her abuse her that she is cripple”, and “Our friends don’t allow my brother to play football with us because he uses crutches.”

Health Care

When asked about health care, children generally expressed excitement and happiness at attending rehabilitation services. Some differentiated between ‘going to the local clinic’ for general health issues, and going to their rehabilitation centres to see their occupational or physiotherapist. Most children knew the rehabilitation workers by name, and felt close to them. They described how the rehabilitation workers would visit them at home when they did not come to the clinic, and said they were happy to see them. A few children mentioned they did not like going to the general health

facility, as the nurses often told them to go back to the rehabilitation center: “We cannot manage children with that condition here.” Parents confirmed this, and explained that health workers appear anxious when they came to the clinic with their child for a malaria test or treatment of diarrhea or cough, and sometimes sent them back. The rehabilitation centers encourage families to attend general health services, as they do not provide this type of health care, but are focused on the disability specific rehabilitation only.

When asking about impairment-related difficulties, the majority of children, siblings, and parents mentioned ‘getting around’ and practicing Clean Intermittent Catheterization (CIC) as the most stressful parts of their daily life.

Getting Around

For children with mobility challenges, all the children, siblings and parents interviewed mentioned the difficulty getting around. A third of the children who could not walk did not have access to an assistive device. Those who did possess aids said they often crawled at home, and in places where their wheelchair cannot access public spaces. Some children who lived in hilly geographical areas said there was no point in having a wheelchair.

“I don’t like using my wheelchair at home, there is no space in the house, and outside there are the gardens, we live on a hill, it’s easier to crawl, especially in the rain season. I leave the wheelchair at a shop down at the main road.” (8-year-old girl, Western Uganda)

Almost half of the wheelchair users could use the wheelchair on their own. However, they reported that they often needed help on the roads as these were full of potholes and humps, which made it very difficult for them to move without another person. Siblings often assisted their brother or sister to go to school or other places.

“I would like to go to school by myself but it is not possible, I cannot cross the road near school, there is a deep ditch and many potholes, my wheelchair gets stuck, someone needs to push me.” (12-year-old boy with spina bifida, Central Uganda)

Most of the younger children were carried by their mothers when travelling. Parents explained that this made it hard to move long distances, as the children can be quite heavy, and are not easy to carry. Nevertheless, many parents continued to travel in this way, as there was no alternative and they wanted their child to go to school or the rehabilitation center.

“I have no choice, the wheelchair my child has cannot pass the road we take home, it is big, it is for adults, someone gave it to us. We leave the wheelchair at school, and I carry her on my back. I walk for almost one hour. I cannot change the school because here they

understand her, and help her. Other schools have refused her to enroll. I cannot afford to use public transport, I would have to pay for both of us and the wheelchair. Passengers may complain about having a disabled on board and having to wait for the wheelchair to be loaded into the vehicle too.” (Mother of a 7-year-old girl with spina bifida and hydrocephalus, Central Uganda)

Siblings indicated they felt responsible for their brother or sister since they were using a wheelchair and often needed their help to get around. Whilst most of them referred positively to helping out, some said they preferred to go somewhere alone, so they did not have to watch their sibling and make sure they are ok. Others indicated that the wheelchair or tricycle was helpful because they could carry things on it.

“I always push my sister to school. It’s heavy and the dust makes it hard to push her [in the dry season]. But the rainy season is worse, sometimes she has to stay home, because the wheelchair cannot pass the road.” (12-year-old sibling of a 9-year-old girl with spina bifida)

“My mother always tells me to take my brother with me but if I go around with him people look at us and call him names because he has a big head. I don’t like that. I want to just meet with my friends alone.” (14-year-old sibling of an 8-year-old boy with spina bifida and hydrocephalus)

“I have to help my sister to get to the main road because there is a ditch she can’t pass on her own. Her tricycle is good for fetching water, we can carry the jerrycans on it.” (9-year-old sister to a 14-year-old girl with spina bifida)

Incontinence

All children and parents said incontinence is a big challenge to them. The siblings interviewed were not engaged in continence management, but did say it was sometimes difficult to find a space for their brother or sister to practice CIC. Whilst most of the children explained they got used to practicing CIC and bowel management, it still interfered with their daily functioning. Most explained it was easier when at home. When travelling or at school or other places, it could be very difficult to find an appropriate place and water to practice CIC or bowel management.

“My child cannot use a normal latrine, they are often dirty, there is no space to do CIC or water to wash. Sometimes we just have to do it outside behind the latrine.” (Mother to a 4-year-old girl with spina bifida, Eastern Uganda)

Some of the older children who practice by themselves say it interrupts their class, as the time they need to practice CIC does not always match with break times. They said it was hard explaining the need to go at certain times to their teachers and classmates. However, if children had enrolled and stayed in the same school over a longer period of time, they felt teachers and students started understanding their needs better.

“I have to get out of class to do CIC, it is not easy, I miss part of the lesson, and it took long for my friends to understand that I had to go out, they thought I was just dodging.” (13-year-old girl with spina bifida in Western Uganda)

My School

Just over half of the children are in school, yet all are in the school going age. All 35 siblings in the similar age range were in school. Some of the children with spina bifida dropped out of school because of bullying: “I dropped out of school because my hands shake, I cannot write, they said I was stupid.” (8-year-old boy with spina bifida and hydrocephalus, Northern Region).

Others did not get a school place, as no school in their area would accept them: “I went to five primary schools. No one wanted my child, they said they cannot manage disabled children.” (Mother of a child with spina bifida, Eastern Region). Other parents said they did not have the finances to send all their children to school, and therefore preferred to select the ones without a disability as they would be more likely to complete their school successfully and find employment later:

“I have 7 children and I can afford to send 4 to school. I cannot send her [the child with spina bifida], I have to add transport for her too as she cannot walk to school, and it is unlikely someone will give her job when she finishes.” (Parent of a 5-year-old girl with spina bifida, Eastern Uganda)

Many parents also felt their child with spina bifida would be dependent on them the rest of their lives, yet children themselves had dreams about what they would be in future: “I want to be a teacher” (7-year-old girl with spina bifida, Central Uganda). “I want to be a doctor so I can make people better” (10-year-old boy with spina bifida and hydrocephalus, Northern Uganda), and “I will become a lawyer so I can make sure all children with disabilities go to school.” (12-year-old girl with spina bifida, Western Uganda). Other parents said their child could not manage school because their brain had been affected by their illness and the schools could not accommodate them.

Most of the children who were in school, said they enjoyed it: “My teacher is nice, she helps me and she tells stories.” (6-year-old girl with spina bifida, Eastern Uganda). They have made friends, though are not able to participate in some subjects such as physical education (PE): “They carry me to play with them outside. Sometimes they say I cannot do something because I am disabled.” (8-year-old

girl with spina bifida, Central Region). Her sibling confirmed this: “She is doing well at school but they don’t let her to do PE because they think she cannot do sports”.

Children and parents rated their performance in school as average. Some complained of secondary disabilities which hindered their performance, such as poor eye sight and difficulties in writing. None of the children had ever been assessed for cognitive functioning.

What I Enjoy Most

When asked to tell a story about something which happened that the child enjoyed and made him or her feel happy, the majority narrated situations in which they were playing with their siblings or friends in the neighbourhood:

“When I was playing hide and seek with my friends. I found the best hiding place under a bush and they could not find me.” (5-year-old boy with spina bifida, Western Region)

Some mentioned going to church or special days such as Christmas when they got new clothes:

“I like going to church with my mum and my sister, they have Sunday school and we sing songs and the teacher tells us stories about Jesus. It makes me very happy.” (7-year-old girl with spina bifida, Eastern Region)

A few older ones mentioned situations at school in which they did well in exams or got a compliment from the teacher:

“One day I had a math test. I find mathematics very difficult and usually I fail, but I worked very hard, and this time I passed. I was very happy, and my teacher said I had done a good job.” (13-year-old boy with spina bifida, Central Region)

Siblings often mentioned games their brother or sister enjoyed playing, and mentioned things they would do together. Parents described similar activities and explained how their children are less clingy and more comfortable at home.

“At home we are at peace. Nobody looks at us, no one calls us names. We just get on with what we need to do. She is calm when she is home, I can put her down and she doesn’t cry.” (Mother to a 4-year-old girl with spina bifida and hydrocephalus, Northern Uganda)

What I Don't Like

When asked to tell a story about something which happened that the child did not like and made him or her feel sad or angry, the majority narrated situations in which they were called names, had a fight with their sibling or friend, or were not allowed to play with others.

Parents explained that their children were often anxious in social situations and public places, and were clingier compared to their siblings, e.g.: "I cannot leave her with anyone, she needs me, I always have to be around, she fears people". A few siblings made similar statements such as "She doesn't want to be alone when we go somewhere, I have to stay close to her".

Similar observations were made during the interviews, in which many children were initially nervous and clingy and stayed very close to their parent(s). A few of them were able to express their fear verbally, as with this 9-year-old boy with spina bifida from the Northern Region: "I don't want to go somewhere without my mum, people say bad things".

Although not expressed directly by all children, most of them did seem to have a (negative) awareness of their impairment following comments made by other people. This was illustrated by statements about preferring to be at home, and avoiding or being anxious in larger public settings, and was observed during home and clinic visits. A few older children mentioned they would like to be treated the same way that children who do not have spina bifida are treated.

Discussion

Children with spina bifida in this study had a strong sense of belonging at household level, but experienced more difficulties in engaging in wider social networks including school. A discussion of belonging divided in thematic areas of family (micro), community (meso), and poverty, services and policy (macro) level is outlined below.

Micro—Family Level

Belonging to a family is a central and key concept in African societies (Bigombe & Khadiagala, 2003; Chataika & McKenzie, 2013; Guyer, 1981; Malinowski, 1929). In our study, children described their households as extended family units, often including not only direct relatives such as parents, siblings, aunts, uncles, and grandparents, but also house helps and neighbours. Whilst some of the children stayed alone with one parent, the majority lived in larger households of about 8 members. Children, siblings and parents described daily activities, play and interactions in and around their home compounds in which they were all involved. Participation in household activities is a key part of daily life and belonging in the Ugandan home setting. Children are often expected to assist in chores such as fetching water, washing clothes, dishes, and sweeping the compound. Children who participated in doing the dishes and washing clothes expressed a sense of accomplishment in this. The daily household activities provided social interaction between siblings and neighbouring

children. Children in our study expressed sadness about not being able to participate in fetching water.

Interviews with siblings and parents confirmed the same. Parents' ratings on the VABS daily subscale showed that children with spina bifida had more difficulties in daily functioning than their siblings. The lower scores could be explained by mobility challenges, lack of assistive devices and poor infrastructure. This is in line with findings from high income countries where physical wellbeing and functioning were significantly lower in children with spina bifida as the disease affects ambulation, functional mobility and self-care (Abresch et al., 2007; Danielsson et al., 2008; Roebroek, Jahnsen, Carona, Kent, & Chamberlain, 2009).

In cases of more severe disabling impairments, we noted that children were less involved and had less interaction with household members. When not participating in an activity, most children sat outside under the shade of a tree or, if living in a larger urban environment, inside, watching what others were doing. A number of children with spina bifida appeared neglected, malnourished, smelling of urine and in very dirty cloths. Warf, Wright et al earlier described finding situations of child neglect during home visits in the Eastern Region (Warf et al., 2011).

Children and siblings said that they participated in family events such as church functions, weddings, and funerals. Some parents explained that participation in these activities was sometimes challenging due to limited mobility and negative attitudes, which is in line with earlier findings of the authors (Bannink, Stroeken, et al., 2015).

Meso—Community Level

Most children describe a mix of experiences of acceptance and enjoyment of play with their friends, and being called names or bullied by others. Bullying and being stared at affected children's self-esteem and increased their anxiety, as we noted during interviews.

In the area of health and rehabilitative care, children said that they liked their rehabilitation worker, usually an occupational or physiotherapist, but described negative experiences with accessing general health care facilities. Children, siblings, and parents all cited mobility and continence management as challenges for participation in the community. for. These challenges have also been pointed out in other studies in high and low income countries (Kabzems & Chimedza, 2002; Martin, White, & Meltzer, 1989; Smith, Murray, Yousafzai, & Kasonka, 2004).

More than half of the children were going to school and enjoyed doing so. Inclusion physical education and accommodation of secondary disabilities which affect learning was challenging. Some parents were financially unable to send their child to school; the child with spina bifida was unlikely to go to school till all other siblings were in school, as parents expected the child with spina bifida would be less likely to complete school successfully and be employed later.

Most children in our study had at times experienced negative attitudes, verbal abuse, as had their siblings and parents. This left the children feeling unable to participate, and made some of them avoid or feel anxious about going to school and public places. Other children were able to develop a

sense of belonging at school. Our study did not look at bullying over time. It is expected that as a child participates in school for a longer period of time and can 'prove' he or she is able to do the same or similar activities as other children, attitudes and behavior of classmates and parents change. We noticed this when studying community attitudes and behavior over time as described by parents (Bannink, Stroeken, et al., 2015). In wider public settings, this may need more awareness raising and behavior change.

In high income countries, children with spina bifida faced more social skills and inclusion challenges than their peers (Wyszynski, 2006), and had lower social integration regardless of whether they could ambulate or use wheelchairs (Dicianno et al., 2009). Our study also found that access to assistive devices did not necessarily make ambulation or social inclusion easier.

Macro—Poverty and Services

The monthly income of families in our study ranged from 28 to 689 US dollars with a median of \$82 (income derived from all sources includes wages, market sales, cattle, land and other assets), which is much lower than the total national average of \$156 (converted from Ugandan shillings), though closer to the average rural income of \$112 and related regional variations (UBOS, 2014).

Whilst poverty was not mentioned directly by participants, the reality of living in a low resource community affected participation in our study population. For example, feeling sad about not being able to fetch water would not have been an issue if running water were available in the home, or the water point could be easily accessed by a wheelchair. Educational access for children with spina bifida is affected when parents can only afford to send 3 out of 5 children to school: the child with spina bifida is unlikely to be selected, as they may have secondary disabilities which make learning more difficult, while it may cost more to transport them to school than would be the case for children who can walk or bike to school on their own.

Witter and Bukokhe (2004) found earlier that "They [Ugandan children] have a positive view of their own potential role in mitigating poverty, and are highly critical of the current performance of local government". The children in our study did not speak about the role of the government. However, they spoke about wanting to enter professions in which they could help other children with disabilities access education and health care.

None of the families spoke about the CRPD which Uganda ratified (UN, 2006). Parents, neighbours, and teachers were mentioned as making the biggest difference and helping out in the inclusion of children in their communities. Reference was made to kindness of others and humanity (e.g. 'ubuntu bulamu' in the Central Region), in line with the key concept of 'ubuntu' in the African setting. Access to formal systems such as public rehabilitative and social services were not mentioned in our study.

Lack of an enabling environment hindered children's participation. The children experienced their impairments as being restrictive. Their experience of the world with their impaired body (Campbell, 2009; Hughes, 2007) and perception of their body prevented them from engaging in daily activities

such as fetching water, going to school, or play, which they saw as a missed opportunity to socialize. Some pointed out that assistive technology could be a burden when living in a hilly place. It is hard to imagine a parent or child living in a rural hilly area without roads arguing for a rights-based approach and the right to assistive devices for their child given that the entire population in the area lives in poverty, with limited access to health care, education, and has no running water.

In a study in South Africa, physical access, transport and medical information, and training and supporting teachers to respond to particular learning disabilities were identified as key areas that need to be addressed to allow children with disabilities to participate in the existing education system (Vosloo, 2009). A study in Uganda showed that children with disabilities are admitted without proper assessment of their educational needs, and resources are not available to provide them with an appropriate range of experiences (Kristensen, Omagor-Loican, Onen, & Okot, 2006). This is something that parents in our study referred to as well. Implementation of inclusive education is negatively affected by non-supportive attitudes of parents and community members, distance to school, unconducive school environment (access, materials, equipment), and lack of trained special need teachers (Abosi & Koay, 2008).

Limitations

Our study was limited to involving children who were receiving or attending follow up and rehabilitation care, and were able to speak. Comparing regional differences was limited by the low numbers of children with spina bifida found in Northern Uganda. Here a 22-year long conflict between the Government of Uganda and the Lord Resistance Army displaced over 90% of the population. Persons with disabilities often faced severe mobility and sanitation challenges in the camp (Muyinda & Whyte, 2011). Whilst some persons received assistance (Mbazzi, Lorschiedter, Hollyne & Opok, 2009) those with congenital disabilities such as spina bifida were less likely to receive services living in a conflict zone compared to those living in poverty in other areas of the country. This may explain the low number of children found in the northern region, as few were able to survive without the initial surgery.

The presentation of a 'voice' is time and space bound (Mazzei, 2016). We were unable to present an ontological voice in which past, present and future were united. However, we did attempt to present voices of the children, their siblings and parents their parents, partly representing an enactment of forces. We took into account observations, and used nonverbal interview methods such as play and drawing. We were limited by translating these into verbal language (Tisdall, 2012). Children could not be included as researchers in this study, resulting in the expert-child bias in interpretation of the text and nonverbal communication (Tisdall, 2012). However, we worked with support groups of parents of children with spina bifida to contextualize findings and checked interpretations with expert parents and rehabilitation workers.

Recommendations

In line with Chataika and McKenzie (2013), our findings support African Childhood Disability Studies in which family, cultural conceptions of disability, poverty, and the notion of 'ubuntu' are central concepts. We advocate working with children in their home setting, where belonging is felt. Family members and peers also play an important role in advocacy and agency in order for belonging to be experienced in the community and school settings.

Rather than employing a child rights based approach, we argue for a family based approach, in which interventions include relatives, start from home, and focus on supporting the family network. Instead of autonomy, interdependency is key in the lives of children with spina bifida. By having expert parents, siblings, and children, families can argue for better services and discuss their needs with health care workers and schools. Through self-advocacy, in which the self is defined in relation to others and humanity to others, rather than in relation to an individual, attitudes and practices of community members can change over time.

Key interventions requiring implementation at the national level include poverty alleviation strategies, improvement of general health care, community based rehabilitation services and better access to education. . Inclusion does not and cannot exist in a dysfunctional system of services based on right based policies which are not implemented. Critical analysis is needed of how to implement ratified conventions and policies in the Ugandan context, to make them applicable and useful.

Conclusion

This study contributes to African Childhood Disability Studies by describing how children with spina bifida and their families experience their daily life and create a sense of belonging in Uganda.

Provision of neurosurgery, rehabilitation, and assistive devices are key services which should be provided and certainly remove certain barriers. However, attitudes and the strong perception of having to 'fit in' the able bodied society (Campbell, 2009) still override the possibilities of children being included in their wider communities. Children mainly belong in their families; 'ubuntu' or 'ubuntu' (Chataika & McKenzie, 2013) is key and is felt at micro-level. We argue for building onto a network at micro level where the environment is more enabling for the children to find a place of belonging and support, and expand this at meso and macro level in support interventions for children in low-resource settings. A bottom up approach is needed to shift the paradigm at meso and macro level from an approach of excluding children with disabilities or making them 'fit in' the norm, to valuing them as unique persons with a sense of belonging and ability to create a society in which they are considered as participants and actors of change.

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4.2 Cognitive abilities of pre-primary and primary school children with spina bifida in Uganda⁵

Abstract

This study investigates cognitive abilities of pre/primary school children without and with spina bifida in Uganda.

Qualitative semi structured interviews and quantitative functioning scales measurements were combined and conducted with 133 parents, 133 children with spina bifida, and 35 siblings. ANCOVA was used to test for differences in cognitive scores between children and siblings. Logistic regression analyses were used to study predictive demographic, impairment specific, and environmental factors of cognitive functioning.

Children with spina bifida in Uganda had lower cognitive outcomes compared to their siblings. Cognitive outcomes were predicted by age, household income, motor functioning, and schooling. Better motor functioning was predicted by age, the presence of hydrocephalus, and parental support. Continence management was predicted by parental support and household income. Schooling was predicted by age, household income, and motor functioning. Limited access to neurosurgery and rehabilitative care, and schooling had a negative effect on cognitive functioning. Children of parents who have support had better motor functioning, and continence management. A holistic approach for children with spina bifida and their families, including community based rehabilitation; ensuring social support and livelihoods for parents; and access to health and education services can contribute to better cognitive outcomes.

Keywords: cognitive abilities, motor function, schooling, spina bifida, Africa

⁵ Based on: Bannink, F., Fontaine, J.R.J., Idro, R., and van Hove, G. (in press) Cognitive abilities of pre- and primary school children with spina bifida in Uganda. *International Journal of Educational Psychology*.

Introduction

Very little is known about the impact of spina bifida on cognitive development among pre- and primary school children in low income countries. In the current study we have investigated differences in cognitive abilities between pre/primary school children without and with spina bifida as well as the factors that best predict cognitive development among children with spina bifida in the Ugandan context.

Spina bifida

Spina bifida is a congenital disability and neural tube defect in which the spinal cord and vertebrae do not form completely and the neural tube fails to develop normally (Northrup & Volcik, 2000). In Uganda an estimated 1,400 children are born with spina bifida annually (Warf, Wright, & Kulkarni, 2011). The majority of these have some degree of paralysis, which affects mobility as well as bowel and bladder control (Abresch, McDonald, Widman, McGinnis, & Hickey, 2007; Andren & Grimby, 2000; Danielsson et al., 2008; Northrup & Volcik, 2000; Verpoorten & Buyse, 2008), and 66% develops hydrocephalus (Warf & Campbell, 2008). In hydrocephalus the natural circulation of cerebrospinal fluid (CSF) in the brain is obstructed and fluid accumulates. The excess fluid presses on the brain causing damage to the surrounding tissue. In babies and infants where the skull is still soft, the head enlarges (CURE, 2008) (IFSBH, 2014). Most children with spina bifida need surgery to close the back to prevent infections; and those with hydrocephalus need endoscopic third ventriculostomy (ETV) or placement of a ventriculo-peritoneal (VP) shunt to drain cerebral spinal fluid and prevent secondary impairments (IFSBH, 2014) (Thompson, 2009) (O'Brien et al, 2005) (Warf, 2005).

Cognitive functioning of children with spina bifida

Research in high income countries suggests that a broad range of cognitive abilities is affected in children with spina bifida (Dennis & Barnes, 2010). They are at risk for difficulties in attention (not sustained attention or ADHD) (Burmeister et al., 2005; Fletcher et al 2005; Rose & Holmbeck, 2007; Swartwout et al, 2008), short-term memory (Vacha & Adams, 2003), prospective memory, immediate and delayed episodic memory (Dennis et al, 2007), nonverbal learning disabilities (Rist et al, 2007), language comprehension and discourse (Huber et al, 2005; Barnes et al, 2007; Pike, Swank, Taylor, Landry, & Barnes, 2013), and approximate and standardized arithmetic measures (Raghubar et al., 2015). Children with spina bifida and hydrocephalus have lower cognitive function scores compared to children with spina bifida alone (Iddon et al, 2004).

To our knowledge no studies on cognitive functioning in pre-primary and primary school aged children with spina bifida have taken place in sub-Saharan Africa. Warf et al (2009) studied neurocognitive outcomes in infants with spina bifida aged 5 to 52 months in Uganda, and found infants not requiring treatment for hydrocephalus had significantly better neurocognitive outcomes compared to those who did require treatment (Warf et al., 2009). The present study focuses on the cognitive impact of spina bifida among pre-school and primary school aged children in Uganda. We compare children with spina bifida with their siblings and we investigate those factors that predict

differences in cognitive abilities within the group of children with spina bifida. We investigate the predictive value of demographic factors that have been found to relate to cognitive abilities in general, of characteristics that are typical for children with spina bifida, and of characteristics that are typical to the Ugandan context.

Demographic factors contributing to cognitive functioning

Demographic variables included in our study were age, gender, socio-economic status, and geographic location. We expected that cognitive outcomes increase with age and socio-economical status. We did not expect to find gender differences. One of the most basic findings in intelligence research is that cognitive abilities increase with age until late adolescence (Hunt, 2010). Whilst there is no evidence for general differences in cognitive ability for boys and girls, gender differences have been found on specific cognitive outcomes depending on the type of cognitive testing administered. For instance, girls performed better verbally, whilst boys performed better on quantitative reasoning in earlier studies (Halpern, 2013; Strand, Deary, & Smith, 2006).

Cognitive abilities correlate not very highly, but systematically (about .30) with socio-economic status (SES) (Hunt, 2010). Whilst income on its own does not predict cognitive abilities, parents with higher SES do spend more time with their children in ways that stimulate cognitive functioning compared to parents with lower SES (Hunt, 2010). Poverty is a factor contributing to cognitive functioning in children with spina bifida in high income countries (Dennis, Landry, Barnes, & Fletcher, 2006; Fletcher, Barnes, & Dennis, 2002). Dennis et al (2006) found socioeconomic status plays an important role in the provision of appropriate remediation for specific cognitive and academic skills. They argue for more studies on how environmental factors such as poverty affect cognitive outcomes which provide approaches to enhance outcomes aside scientific understanding (Dennis et al., 2006). In our study we expect that children from parents with higher SES will have better cognitive outcomes as parents are more likely to provide their children with basic needs and access services.

Differences in cognitive outcomes between urban and rural areas are linked to poverty, schooling, and other environmental factors (Dennis et al, 2006). Given the differences in income and social setting, we will investigate if there is a difference in cognitive outcomes between children living in urban and rural areas in Uganda.

Impairment specific factors contributing to cognitive functioning

In the analysis we include spina bifida specific variables such as the presence of hydrocephalus, neurosurgical treatment, motor function, and incontinence management.

Hydrocephalus

The neuropsychological profile of persons with hydrocephalus is one of relative impairment, whether or not spina bifida is present. Shunt infections, revisions, and a history of seizures predict poorer memory (Dennis et al, 2007), meta-cognitive abilities (Brown et al, 2008), executive functioning

(Tarazi, Zabel, & Mahone, 2008), and cognitive health (Kulkarni et al, 2004). In a comparative study Hampton et al. (2011) found higher neurocognitive scores in children with spina bifida without hydrocephalus compared to children with SB and hydrocephalus. A stepwise pattern in terms of number of affected domains from the shunt-treated group to the arrested-hydrocephalus group to the no-hydrocephalus group was identified (Hampton et al., 2011). We expect to find the same stepwise patterns, with possibly larger effect sizes due to the limitations in access to early neurosurgery and regular physio- and occupational therapy.

Neurosurgery

The majority of children with spina bifida and progressive hydrocephalus need neurosurgery. At the time of this study the initial surgery (closure of the spine) and ETV and VP-shunting for children with progressive hydrocephalus were only available in two public government funded hospitals (Mulago National Referral Hospital in Kampala and Mbarara Regional Referral Hospital), and one private specialized neuro-paediatric hospital (CURE Children's Hospital) in Mbale, eastern Uganda. Delay in neurosurgery has been associated with more neurodevelopmental delay (Warf et al., 2009). Shunts are sensitive to infection and malfunctioning (Hoppe-Hirsch et al, 1998; Hunt et al, 1999), and are related to epilepsy (Bourgeois et al, 1999; Kulkarni et al, 2004). ETV is an alternative primary treatment for hydrocephalus and alternative for malfunctioning and infected shunts (Warf, 2005). Earlier Warf et al. found no differences between cognitive outcomes of infants aged 5 to 52 months treated with ETV and VP shunt treatment (Warf et al., 2009). We will evaluate the effect of type of neurosurgery on cognitive functioning in pre-primary and primary school aged children.

Gross and fine motor functioning

Gross and fine motor functioning are affected in children with spina bifida. Gross motor skills such as movement of the upper and lower limbs, and eyes are core deficits in children with spina bifida (Dennis et al., 2006). Children with spina bifida with extensive paralysis will often require a wheelchair, while others may be able to use crutches, braces, or walking frames. They benefit from physio- and occupational therapy (Abresch et al., 2007; Andren & Grimby, 2000; Danielsson et al., 2008; Jansen, Blokland, de Jong, Greving, & Poenaru, 2009). Children with spina bifida are prone to pressure sores and need regular skin checks to prevent these (Lindsay, 2014).

The majority of children with spina bifida have impaired fine motor function and visual-motor integration. Handwriting and drawing are the most affected skills (Feder & Majnemer, 2007; Hetherington, Dennis, Barnes, Drake, & Gentili, 2006; Lindquist, Persson, Uvebrant, & Carlsson, 2008; Vinck et al., 2010).

We expect that children with better motor function have better participation in daily activities, including schooling, which will in turn improve cognitive functioning.

Incontinence

Most have bowel and bladder problems. Clean Intermittent Catheterization (CIC) and bowel management techniques are used to keep the child dry and clean (IFSBH, 2014; Verpoorten & Buyse, 2008). CIC and bowel management training in Uganda is offered by the four organizations who provide physical rehabilitative therapy. Through a CBR approach funded by private donors, children are followed up at home and are provided with low costs – if possible locally made - continence materials (Mertens & Bannink, 2012).

We study continence as a possible important factor for inclusion, as we expect that good incontinence management results into increased participation in daily activities, including schooling. A child with not managed incontinence will have accidents and smell of urine, and is more likely to be socially excluded.

Environmental factors contributing to cognitive functioning

Schooling

Although inclusion of children with spina bifida in normal schools is common in high and middle income countries, this remains a challenge in low income countries (Chataika, McKenzie, Swart, & Lyner-Cleophas, 2012). Implementation of inclusive education in Sub Saharan Africa is affected negatively by non-supportive attitudes of parents, teachers, and community members, distance to school, not conducive school environment (access, materials, equipment), and lack of trained teachers (Abosi & Koay, 2008; Vosloo, 2009). The Ministry of Education and Sports in Uganda has an Universal Primary Education (UPE) and inclusive education policy for access and inclusion of children with special needs in primary schools (Nyende, 2012), however implementation of inclusive education remains limited at grass root level (Ejuu, 2016). In our study we expect a large number of children to be out of school. In earlier findings parents of children with spina bifida indicated UPE schools and several private schools would not admit their child (Bannink, Stroeken, Idro, & Van Hove, 2015). Of those in school, children reported being bullied about their physical appearance and learning speed (Bannink, Idro, & Van Hove, 2016a). Children who are not schooling are likely to have lower cognitive outcomes and become socially more isolated as schooling itself has an effect on general cognitive development (Hunt, 2010; Sylva, 1994).

Social support

In high income countries family support has been identified as a factor contributing to cognitive functioning in children with spina bifida (Dennis et al., 2006; Fletcher et al., 2002). Having support increases the chances of access to remediation for specific cognitive and academic skills. Caregivers of children with disabilities in sub Saharan Africa often felt that they do not have sufficient time to cope with household tasks and feel isolated (Gona, Mung'ala-Odera, Newton, & Hartley, 2011; Hartley, Ojwang, Baguwema, Ddamulira, & Chavuta, 2004). Families of children with neuro-disabilities including spina bifida in Kenya (van't Veer et al., 2008), Malawi (Paget, Mallewa, Chinguo,

Mahebere-Chirambo, & Gladstone, 2015), and South Africa (Coomer, 2013) struggle with the social barriers towards care and support for their child. In absence of health care and social services for children with disabilities in sub Saharan Africa, the children's families are often their main source of care and protection (Guyer, 1981; Miles, 2002).

We expect that cognitive outcomes of children with spina bifida are related to whether parents have support from another adult or support group or not. We believe that parents who feel supported, are more likely to stimulate their children at home, resulting into better cognitive outcomes of their children.

In this study we will compare cognitive outcomes of children with spina bifida and their siblings. We will analyse which demographic (sex, gender, SES), spina bifida specific (hydrocephalus, neurosurgery, motor function, incontinence), and environmental factors (schooling and social support) predict cognitive functioning, and make recommendations for further studies and interventions in low resource settings.

Methods

Study design

Purposeful sampling was used in Mbarara, Kampala, and Mbale where CURE holds bi-monthly clinics. CURE hospital and the partnering rehabilitation centers in Kampala and Mbarara were requested to list the children registered in their follow up programs, and inform them during home visits and through telephone calls to attend the clinic. In Gulu and Lira where no follow up system or registry of the children was in place at the time, radio announcements were aired to inform parents about the upcoming review clinic in the area, and specifically invited parents with children with spina bifida and hydrocephalus between the age of 4 and 14 years to attend.

Qualitative semi structured interviews and quantitative functioning scales measurements were combined and conducted between June 2011 and December 2014 with 133 parents, 133 children with spina bifida, and 35 siblings. The siblings were randomly selected from the 63 families living in the central region.

The assessments were held in the local language of the area, and a translator was hired and trained for each area to assist in conducting the assessment. Some of the interviews were conducted in English, if parents, children or teachers were fluent and requested for this.

Ethical considerations

Ethical approval and research clearance were obtained from Ghent University, Belgium, the Uganda Virus Research Institute, and the Uganda National Council for Science and Technology. Informed consent was obtained from all parents and teachers, and assent from children and siblings of 8 years and above where possible. Consent forms were translated into the local languages and discussed and

agreed with the participant with assistance of a translator if the participant or child did not speak English.

Selected measures

Demographic and impairment specific variables such as the presence of hydrocephalus, neurosurgical treatment and rehabilitation services received, and incontinence management was collected from the parents for each child. Data on school going was collected and entered into a database.

Gross motor skills were assessed using basic measures on whether a child was able to sit or walk independently, and was using assistive devices. Fine motor skills were measured with the Vineland Adaptive Behaviour Scales (VABS) Daily Functioning Sub Scale and reflect both fine motor skills and daily functioning skills such as the ability to dress oneself, bath, and eat or drink independently. The reliability of the fine motor skill outcomes measure of the VABS was good with a Cronbach's Alpha of .82. The gross and fine motor skills measures were assessed by asking the parents about the child's functioning and observing the children's abilities.

Table 1. Psychometric assessment tools screened, piloted, and administered

Tools screened	Selected for pilot	Administered
Neurocognitive functioning		
Picture Vocabulary Scale - adapted from the Kilifi Picture Vocabulary Test, (Nampijja et al., 2010)	Picture Vocabulary Scale	Picture Vocabulary Scale
Kaufman Assessment Battery for Children II - validated for Ugandan population (Bangirana et al., 2009)	Subtests hand movement, block counting, number recall, triangles	Subtests hand movement, block counting, number recall (<i>triangles not understood</i>)
Wechsler Intelligence Scale for Children IV (Wechsler, 2003)	Block Design, Picture Concepts, Matrix Reasoning, Digit Span, Letter-Number-Sequencing, Coding Symbol Search	<i>Items not understood, digit span was understood but same measure as number recall of K-ABC</i>
Block Design - adapted from the British Ability Scales (Nampijja et al., 2010)	Block design	Block design
Halstead-Reitan Neuropsychological Battery - progressive figures and trail-making (Reitan, 1985)	Trail-making and Progressive Figures	<i>Fine motor skills difficulties made it hard for most to complete this test</i>
Visual Motor Integration Test (VMI)–visual perception / motor integration (Beery, Buktenica, & Beery, 1989)	Visual Motor Integration Test (visual perception, and motor integration)	Visual perception and motor integration

To measure cognitive functioning, a set of cognitive subtests from existing tools was screened, selected for piloting, and administered (Table 1). Cross-cultural application of neurocognitive assessments has been studied, and assessment batteries have been adapted for sub-Saharan African settings including Uganda, the majority in HIV and malaria studies (Abubakar et al., 2008; Alcock, Holding, Mung'ala-Odera, & Newton, 2008; Bangirana et al., 2009; Bangirana, Sikorskii, Giordani, Nakasujja, & Boivin, 2015; Boivin, 2002; Holding et al., 2004; Nampijja et al., 2010). The pilot of psychometric assessment tools took place in a group of 12 children in Kampala. Most assessment batteries were difficult to complete, and its cultural adequacy could be questioned. Based on the pilot, we selected subscales which were understood and were culturally relevant. The Picture Vocabulary scale works with pictures relevant to the Kenyan and Ugandan context, and the block design test was simplified using wooden blocks by Nampijja et al (Nampijja et al., 2010). The other tests were not adapted, but deemed fit for use in the Ugandan context. The subscales were combined and administered to all children. On average the cognitive assessments took 1.5 hours. Assessments took place at the rehabilitation sites or the child's home.

Data management

Basic demographic, impairment, schooling data, and records of the cognitive, gross and fine motor scores were written out during assessments, and entered into a SPSS database after completion. Demographic data included age, gender, religion, location, socio-economic status, parental education, occupation, and marital status. Impairment specific variables included were the presence of hydrocephalus, neurosurgical treatment and rehabilitation services received, incontinence status, and incontinence management. Information on schooling, class, and performance was collected for each child. Motor and cognitive outcomes were entered in handheld assessment scales, and entered into a password protected database. Study files were filed in a lockable cupboard.

Data Analysis

The sub-total scores for the VABS subscale were calculated to compare means of the scores between the children with spina bifida and their siblings using SPSS16.

Confirmatory factor analysis was carried out to investigate whether all cognitive subtests could be represented by a single underlying cognitive ability factor. Not all children were able to carry out all the sub-tests. Within the confirmatory factor model global cognitive functioning was estimated on the basis of those tests that were completed by each child.

ANCOVA was used to test for significant differences in cognitive scores between children and siblings using age as a covariate. The overall ability scores estimated on the basis of the confirmatory factor analysis was used as dependent variable.

Factors predicting inter-individual differences in cognitive functioning within the spina bifida group were investigated by regression analysis with a forward selection procedure. Because of its importance in predicting cognitive functioning, it was further investigated which factors predicted schooling and motor functioning using logistic regression and multivariate regression respectively.

Sample description

The study population consisted of 133 children with spina bifida (myelomeningocele type) (59.4% male, 40.6% female) of which 60 with spina bifida and hydrocephalus; 133 parents; 35 siblings (21 female, 14 male); and 30 teachers (22 female, 8 male) of children with spina bifida. Table 2 describes the demographics of the study population.

Table 2. Demographic characteristics of the study population (N=133 children with spina bifida, N=35 siblings)

Variable	Child	%	Sibling	%
<u>Gender child</u>				
Male	79	59.4%	14	40.00%
Female	54	40.6%	21	60.00%
<u>Child is schooling in</u>				
Nursery school	48	36.1%	0	
Primary school	22	16.5%	9	25.70%
Secondary school	4	3.0%	26	74.30%
Not schooling	59	44.4%	0	
<u>Type of disability</u>				
Spina bifida	73	54.9%	0	
Spina bifida and hydrocephalus	60	45.1%	0	
<u>Location / region</u>				
Central	63	47.4%	35	100%
East	26	17.3%	0	
West	29	21.1%	0	
North	19	14.3%	0	
<u>Religion</u>				
Christian	100	77.5%	32	80.0%
Muslim	26	20.2%	6	17.1%
Other	13	2.3%	1	2.9%
<u>Relationship parent</u>				
Mother	104	78.2%	26	74.3%
Father	14	10.5%	6	17.1%
Grandmother	9	6.8%	2	5.7%
Other	6	4.5%	1	2.9%
<u>Education level parent</u>				
None	6	4.7%	3	8.6%
Primary	72	56.7%	22	62.9%
Secondary	28	22.0%	6	17.1%
Vocational	11	8.7%	1	2.9%

University	10	7.9%	3	8.6%
<u>Marital status parent</u>				
Single	12	9.2%	0	
Married	101	75.6%	26	74.3%
Separated	9	6.9%	6	17.1%
Widowed	11	8.4%	3	8.6%
<u>Monthly household income</u>				
< 30 euro	25	19.7%	11	31.4%
30 - 60 euro	28	22.0%	7	20.0%
61 - 90 euro	26	20.5%	8	22.9%
> 90 euro	48	37.8%	9	25.7%
<u>Occupation parent</u>				
Finance / administration	5	3.8%	2	5.9%
Small scale private business	30	22.7%	7	11.8%
Teacher / education	9	6.8%	3	8.6%
Medical / paramedical	5	3.8%		
Civil service / government	2	1.5%		
Peasant farmer	65	49.2%	19	62.9%
No occupation	6	12.1%	3	8.6%

The average age of the children with spina bifida was 6.1 (SD=2.04), and 7.8 years (SD=2.53) for siblings. The majority of the children with spina bifida (122, 91.7%) had undergone surgery to close their spine (myelomeningocele closure) earlier in life. Whilst we looked for children in communities using radio announcements and searches through community leaders, we did not find more children who had not undergone surgery. We think that the majority who did not access neurosurgery died, as the initial surgery is key to survival in spina bifida. Of the 60 children who had both spina bifida and hydrocephalus, 25 (41.7%) had undergone endoscopic third ventriculostomy while 12 (20.0%) had ventriculo-peritoneal shunts placed. Here too, we could not find more children who had undergone VP shunt placement, which could be attributed to the high infection and mortality risks associated with this type of treatment in a low resource setting like Uganda. During our study 3 children died from the complications of shunt malfunctioning. The majority of parents took their children (123, 92.5%) for rehabilitation services such as physio- and occupational therapy. Parents' ages ranged from 24 to 46 years with an average age of 32.9 (SD 5.2) years. About a third, 31.8% (42) of the parents did not have support from another adult or parents support group. The household size ranged from 2 to 13 with an average of 6.5 persons per household (SD=2.49), with on average 4.3 children (SD 2.2) and 2.3 adults (SD 1.0) per household. The average monthly household income was 82 euro (range 12 to 604 euro). In total 63 families resided in the capital city and surrounding urban areas (47.4%), whilst 52.6% (70) lived in rural areas.

In total 54 children with spina bifida who were not in school, 74 children with spina bifida who were schooling, and 35 siblings who were schooling were included in the analysis of cognitive outcomes.

Gross motor skill outcomes of children with spina bifida were grouped into 4 groups: 58 children who can sit and walk (43.6%), 36 children who can sit, and use assistive devices to walk (27.1%), 37 children who can sit, not walk and do not use assistive devices to ambulate (27.8%), and 2 children who could not sit nor walk and are not using assistive devices (1.5%).

In total 106 children (79.7%) with spina bifida were incontinent; 97 of them used CIC. The continence factor was dichotomized by grouping children who were using CIC and bowel management together and comparing them to 25 children who do not use continence management.

Results

Cognitive test battery

Confirmatory factor analysis was carried out on the seven subtests. The one-factor model fitted the data well ($\chi^2 = 24.04$, $df = 14$, $p = .034$, $\chi^2/df = 1.72$, RMSEA = .07, CFI = .96, SRMR = .04. Table 3 shows the standardized factor loadings of each subtest. All subtests had high factors loadings (>.70).

Table 3. Confirmatory factor analysis of cognitive measures

Cognitive Sub Test	Fit factor loading
Picture Vocabulary Scale total score - measures verbal cognitive ability	.70
Block Design total score – measures spatial ability	.99
Block Count total score - measures visual processing	.89
Hand movement - measures short term memory / sequential processing	.98
Number recall – measures short term memory / sequential processing	.96
Visual Motor Integration – measures motor output skills coordination	.83
Visual Motor Integration – measures visual perception input skills	.79

Cognitive outcomes in children with spina bifida with their siblings

An Analysis of Covariance with overall cognitive functioning as dependent variable, group (siblings, schooled children with spina bifida, and non-schooled children with spina bifida) and sex as independent variables, and age and socio-economical status (income) as covariates showed a significant and strong group effect [$F(2,145) = 17.34$, $p < .001$, partial $\eta^2 = .193$]. There was no significant effect of sex [$F(1,145) = 0.56$, $p = .456$, partial $\eta^2 = .004$] and no significant interaction effect between sex and group [$F(2,145) = 0.291$, $p = .748$, partial $\eta^2 = .004$]. Moreover, both covariates were significant [$F(1,145) = 43.650$, $p < .001$, partial $\eta^2 = .231$ for age and $F(1,145) = 7.165$, $p = .008$, partial $\eta^2 = .047$]. Table 4 summarizes the mean scores and standard errors. Siblings of children with spina bifida had better cognitive outcomes compared to their schooling brothers and sisters with spina bifida, whilst children with spina bifida enrolled in school had better cognitive

outcomes compared to children with spina bifida not enrolled in school. No significant gender differences were found.

Table 4. Cognitive outcomes for schooling and not schooling boys and girls with spina bifida and their siblings controlled for age and household income.

		Mean	Std. Error	CI (min – max)
Child with spina bifida not schooling	Male	-1.104	.348	-1.793 to -.415
	Female	-1.081	.391	-1.854 to -.308
Child with spina bifida schooling	Male	.014	.268	-.515 to .543
	Female	.519	.334	-.140 to 1.179
Sibling schooling	Male	1.339	.482	.386 to 2.292
	Female	1.500	.411	.688 to 2.312

Note: covariates appearing in the model were evaluated at the following values: age child=6.5687, household income = 2.71.

Factors related to differences in cognitive functioning of children with spina bifida

To investigate the predictors of inter-individual differences in cognitive functioning within the group of children with spina bifida, a regression analysis was executed with forward selection procedure with age, sex, impairment (e.g. spina bifida alone or spina bifida and hydrocephalus), type of treatment received for those with hydrocephalus (ETV or VP shunts), continence, household income, geographical region, education level parents, and support received by parent as possible predictors. Only four predictors were selected for cognitive functioning on the basis of this forward selection regression procedure (fine motor skills, age, schooling, and income). These four predictors accounted for 50.4% of the total variance in cognitive functioning. Older children with better fine motor skills, who were schooling and came from households with higher household income had significantly better cognitive functioning (see Table 5).

Table 5. Selected predictors for cognitive functioning within the group of children with spina bifida

Predictors	<i>B</i>	<i>Beta</i>
Fine motor skills	.194	.390**
Age child	.294	.271**
Schooling	.942	.210**
Household income	.304	.153*

Note. ** $p < .001$, * $p < .05$

Because of the importance of schooling and motor skills, we have further explored which factors predict schooling and these motor skills.

To investigate the predictors of whether children with spina bifida were schooled or not, a logistic regression was executed with a forward selection procedure with age, sex, impairment (e.g. spina bifida alone or spina bifida and hydrocephalus), type of treatment received for those with hydrocephalus (ETV or VP shunts), continence, household income, geographical region, level parents, and support received by parent as possible predictors. Only three predictors were selected. The older the child ($\text{Exp}(B) = 1.595$, $\text{Wald} = 10.692$, $p < .001$], the higher the household income of the parents ($\text{Exp}(B) = 1.869$, $\text{Wald} = 9.087$, $p = .003$], and the better the fine motor functioning ($\text{Exp}(B) = 1.235$, $\text{Wald} = 15.338$, $p < .001$] the more likely the children with spina bifida was going to school.

To investigate the predictors of fine motor functioning among children with spina bifida a multivariate regression analysis was executed with a forward selection procedure with age, sex, impairment (e.g. spina bifida alone or spina bifida and hydrocephalus), type of treatment received for those with hydrocephalus (ETV or VP shunts), household income, geographical region, educational level parents, and support received by parent as possible predictors. Three significant predictors were identified: age, parental support and impairment. These three predictors accounted for 19.6% of the variance in fine motor functioning. The older the child ($\beta = .309$), parents having social support ($\beta = .246$), and children without hydrocephalus ($\beta = .214$) had significantly better fine motor skills [$F(3, 128) = 10.408$, $p < .001$, $R^2 .196$].

Moreover, fine motor skills were also significantly correlated with gross motor skills ($r = .616$, $p < .001$) and with continence ($r = .405$, $p < .001$). A multivariate and a logistic regression analysis with a forward selection procedure using the same predictors as for fine motor functioning identified similar predictors for gross motor skills and continence respectively. The older the child ($\beta = .216$), parents having social support ($\beta = .210$), and children without hydrocephalus ($\beta = .267$) had significantly better gross motor skills [$F(3, 129) = 7.805$, $p < .001$, $R^2 .154$]. When parents could rely on social support [$\text{Exp}(B) = 3.420$, $\text{Wald} = 3.383$, $p = .012$] and when parents had a higher income [$\text{Exp}(B) = 1.900$, $\text{Wald} = 8.036$, $p = .005$] children were more likely to be continent or manage their continence.

Discussion and conclusion

Cognitive testing

Environmental factors play a key role in the cognitive function of children with spina bifida in Uganda. Earlier Bangirana et al. and Nampijja et al. have provided evidence for successful adaptation of assessment batteries for Ugandan children taking cultural factors into consideration (Bangirana et al., 2009; Bangirana et al., 2015; Nampijja et al., 2010). Despite the cultural sensitivity of some of these instruments, most of these assessment tools were too challenging to complete for the children with spina bifida due to motor difficulties. To mitigate this, we did not apply tests which were too difficult from a motor perspective, and estimated cognitive functioning on the basis of less

challenging test. No speed tests were applied which enabled children with lower motor ability to take their time to answer the questions. Despite the adjustments made and testing the pilot in the central region, a number of children in the study group could still not perform the procedures. In some cases observations during the cognitive testing indicated that non-responding was caused by feeling shy rather than being unable to complete the tests. This mainly concerned children who were not schooling and were from rural areas where they lived in rather remote areas.

We do not have a comparable assessment tool to look at effect size between high income countries and our findings. However from the high number of children who could not complete the culturally and impairment specific testing set, we may conclude that the level of cognitive functioning was lower compared to high income countries.

Cognitive outcomes in children with SB and their siblings

Children with spina bifida in Uganda had lower cognitive outcomes compared to their schooling siblings. This is in line with earlier studies in which children with spina bifida have lower scores on visual perception, language comprehension, attention, processing and memory tasks compared to their peers (Dennis & Barnes, 2010; Dennis et al., 2007; Rose & Holmbeck, 2007). Hampton et al found differences of 13 to 20 points in IQ scores in children with spina bifida (Hampton et al., 2011). In our study we did not have Ugandan population norms which would allow us to precisely estimate the size of effects in terms of IQ points. Cognitive outcomes in our study population were predicted by age, household income, motor functioning, and schooling.

Demographic factors contributing to cognitive functioning

As expected age had an evident effect on cognitive functioning. No significant differences were found between boys and girls, which is in line with earlier studies of cognitive functioning in Ugandan children (Nampijja et al., 2010). Children from rural and urban areas had similar cognitive outcomes. Household income was a main factor in predicting cognitive outcomes. Interestingly parental education or social economical status did not predict cognitive outcomes, but household income did. Having sufficient income to send children to school and access rehabilitative care, is key in a low resource setting with limited public services. Earlier we found that parents would not prioritize sending their child with spina bifida to school if they did not have sufficient funds to send all children to school (Bannink, Idro, & Van Hove, 2016b).

Impairment specific factors contributing to cognitive functioning

Motor function

Children with better motor function had higher cognitive functioning scores. Although children with good gross and fine motor skills are expected to score higher on cognitive tests which require fine motor skills such as block designs, and the visual motor integration drawing tasks (Raghubar et al., 2015), we believe this result is not a direct effect. No timed tests were administered and if a child

could not complete a subtest, this was adjusted for, leaving out negative impacts on the total cognitive outcomes scores. Instead we believe children who have better motor function are more likely to participate in daily activities, and be stimulated through these, resulting in better cognitive development. Parental support and household income were important predictors for continence management. If parents feel supported, they may be more likely and able to carry out CIC several times a day. This in turn will help improve quality of life; earlier better motor function and continence management were related to better quality of life in Kenya (van't Veer et al., 2008). Higher household income would help with covering travel costs to rehabilitation centers for supplies of catheters.

Hydrocephalus

The type of impairment, neurosurgery or rehabilitation services the children received did not affect the cognitive outcomes in children in our study. In earlier findings Ugandan infants not requiring treatment for hydrocephalus had significantly better neurocognitive outcomes compared to those who did require treatment, but no differences were found between cognitive outcomes of infants treated with ETV and VP shunt treatment (Warf et al., 2009). We did not find significant differences between children with and without hydrocephalus controlled for fine motor development. However hydrocephalus did predict motor function, which significantly contributes to cognitive outcomes, indicating an indirect effect. Hampton et al (2011) found lower cognitive outcomes in children with spina bifida and hydrocephalus compared to children with spina bifida alone. It is possible that our sample was too small to find any direct effect, or that the effect is reduced as other factors in our setting are more important, e.g. having sufficient income, and better motor functioning and going to school. The effect of the presence of hydrocephalus and type of surgery may not weigh sufficiently in a setting where a child is not being schooled or has not access to rehabilitative care.

Environmental factors contributing to cognitive functioning

Schooling

Children with spina bifida enrolled in school had better cognitive outcomes compared to children with spina bifida not enrolled in school. Schooling was predicted by age, household income, and motor functioning. When controlling for motor function and income, schooling still had a significant effect on cognitive functioning. This pleads for increasing school enrolment and inclusion.

In total 58.7% of the children with spina bifida in our study were in school. This is low compared to the national net enrolment ratio for primary school of 94.5% (Government of Uganda, 2013), but high compared to the UNICEF study in which only 9% of children with disabilities were enrolled in school (UNICEF, 2014). It should be noted that this last study provided a percentage for children with all types of impairments. Whilst children with spina bifida have cognitive functioning deficits, they generally are able to enrol and participate in school and follow the national curriculum material. As well our study mainly included children who had received neurosurgery and may have been more

likely to survive, receive care, and enrol in school compared to a child with cerebral palsy or intellectual disability.

Parental support

Aside the positive effect of schooling on cognitive outcomes, we believe that having support and more income, directly affects the child. Most likely parents who receive social support and have a higher income, have more time to take their children for physical rehabilitation, and provide direct stimulation at home, which stimulates the motor and cognitive development, and CIC. They may equally have more funds to purchased mobility devices and physiotherapy compared to parents with lower incomes. Even in high income countries, physical impairment outcomes such as continence, and community ambulation are lower in groups without private insurance (Schechter, Liu, Swanson, Ward, & Thibadeau, 2015).

Limitations

Limitations of our study included purposeful selection of participants. Despite radio announcements and searches for children with spina bifida in the communities by local leaders, we did not find any children who had not received neurosurgery and rehabilitation. Most likely those who did not receive this died. The children who participated in our study may have therefore only represented those who did 'better' or did not have shunt infections, resulting in overly positive cognitive outcomes.

Another limitation was the lack of a validated and normed cognitive assessment tool for Ugandan children with spina bifida to define cognitive assets and deficits in detail and make recommendations on neurocognitive learning goals. The impact of medical factors such as Chiara II malformation could not be included in the study as information and imaging facilities were not available. The number of siblings included in the study was limited due to logistic constraints in follow up of children upcountry.

Implications for future studies and interventions

In future a cohort study following children from birth is recommended to understand mortality, survival, and motor and cognitive development in relation to their environment and rehabilitation care received. In future studies we recommend selecting families in one or two geographical areas and follow the child, siblings, parents, and other caregivers over time.

The feasibility and meaning of performing psychometric procedures in children with severe impairment has been questioned (Fletcher, 2014). We found difficulties in assessing children from rural areas who lived isolated lives and would not answer questions. We equally struggled to find appropriate tests which could be completed by children with spina bifida. Fletcher and Dennis have argued for the use of adaptive behaviour assessments as a supplement to cognitive neuropsychological assessments and more studies on how environmental factors affect cognitive functioning providing approaches to enhance outcomes (Dennis et al., 2006; Fletcher, 2014). Building

on this, and our findings which indicate the importance of household income, motor function, and parental support we argue for awareness raising on the cognitive profiles of children with spina bifida and develop low costs strategies to help them in learning, and providing parental support.

Livelihood support to increase household income will be a key strategy to increase access to services and improve cognitive functioning in the long run. Successful approaches reported by non governmental and governmental programs have included village savings and loan associations and small scaled capacity building programs, and could target more families of children with disabilities (Government of Uganda, 2014; NUDIPU, 2016; USAID & AVSI, 2016).

We argue for implementation of public health community based rehabilitative services. Despite policies in place, none of the families in our study access governmental CBR services. Better referral networks from public facilities where CBR is lacking to private facilities is recommended to ensure children receive rehabilitative therapy.

Lastly but most importantly we argue for more support for parents as parental support improves children's outcomes in the long run: from better motor skills to school enrolment and better cognitive outcomes. This support can consist of the presence of another adult in the home to help in daily tasks, but also of a peer support group to discuss care and challenges with other parents with spina bifida. The International Federation for Spina Bifida and Hydrocephalus (IF) had such support groups in place and encourages set up and support of new groups in low resource countries (Mertens & Bannink, 2012). Further use and support of such groups can enhance the future education outcomes of the children.

By ensuring a holistic approach for children with spina bifida and their families, involving the physio-, and occupational therapist in community based rehabilitation to develop fine and gross motor skills; making sure parents receive social support and improve their livelihoods so that children can go to school; cognitive outcomes can improve over time.

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4.3 Teachers' and parents' perspectives on inclusive education for children with spina bifida in Uganda⁶

Abstract

Despite all the policies in place implementation of inclusive education remains limited at grass root level in Uganda. This paper explores accessibility and inclusion of children with spina bifida in primary schools with parents and teachers.

Qualitative semi structured interviews and school observations were combined and conducted with 63 parents and 30 teachers in Uganda's central region. The Index of Inclusion was used as a guide in the interviews with parents, students, and teachers, and the Trip Chain concept and drawings of the Ugandan Accessibility Standards were used to measure accessibility.

Children with spina bifida with poor physical and cognitive functioning from families with a low household income were less likely to be in school compared to children with better functioning scores and a higher household income. Physical accessibility to schools for children with spina bifida is very limited. Classroom participation is affected by lack of space, materials, knowledge and experience of teachers to use diversified teaching methods. Education performance is rated lower by teachers than parents. Inclusive policies to include the children and prevent bullying are in place but lack implementation.

To achieve inclusive education for children with spina bifida, awareness raising to reduce discrimination, training and on job mentoring to support teachers and schools, and earmarking funds for inclusiveness in schools for children with disabilities is required.

⁶ Based on: Bannink, F., Idro, R., and van Hove, G. (2016) Teachers' and parents' perspectives on inclusive education for children with spina bifida in Uganda. *Journal of Childhood & Developmental Disorders*, 2, 2: 18. DOI:10.4172/2472-1786.100026.

Introduction

Children with spina bifida in Uganda

Spina bifida is a congenital disability and neural tube defect; the spinal cord and vertebrae do not form completely and the neural tube fails to develop normally. The worldwide incidence of spina bifida varies between 0.17 and 6.39 per 1000 live births (Bowman et al., 2009; Kinasha & Manji, 2002; Msamati et al., 2000; Shaer et al., 2007). Incidence and prevalence rates in Uganda may be higher due to inadequate folate consumption by pregnant women (Bannink, Larok, et al., 2015; Whyte, 1995), lack of pre-natal care (Miles, 2002), absence of secondary prevention services (Frey & Hauser, 2003), and higher exposure to environmental risk factors such as dioxins (Safi et al., 2012) and fumonisins intake (Hendricks, 1999; Marasas et al., 2004; Wild & Gong, 2010). No national data are available in Uganda. Warf et al estimate that 1,400 children are born with spina bifida in Uganda annually (Warf, Wright, et al., 2011), and 66% of children with spina bifida develop hydrocephalus (Warf & Campbell, 2008).

For children with spina bifida participating in daily activities is challenging as the majority have some degree of paralysis, which affects mobility as well as bowel and bladder control (Northrup & Volcik, 2000) (Abresh et al, 2007; Andren & Grimby, 2007; Bannink, Idro, & Van Hove, 2016a; Danielsson et al, 2008; Verpoorten & Buyse, 2008). Most children with spina bifida need surgery to close the back to prevent infections; children with progressive hydrocephalus need endoscopic third ventriculostomy (ETV) or placement of a ventriculo-peritoneal (VP) shunt to drain cerebral spinal fluid and prevent secondary impairments (IFSBH, 2014; O'Brien et al, 2005; Thompson, 2009; Warf, 2005).

Surgery and rehabilitative care is expensive and inaccessible for many children born with a physical disability in Africa. In East Africa efforts are made to provide basic services at community level for children with spina bifida, but largely remain funded by international donors and charities (Mertens & Bannink, 2012). In Uganda the initial surgery (closure of the spine) at the time of this study was only available in two public government funded hospitals (Mulago National Referral Hospital in Kampala and Mbarara Regional Referral Hospital), and one private specialized neuro-paediatric hospital (CURE Children's Hospital) in Mbale, eastern Uganda. In north, west, and central Uganda three rehabilitation centers, funded by international donors and charities offer occupational therapy, physiotherapy, continence management, and social support services for children with spina bifida and their families.

Children with spina bifida have intelligence in the normal range, tending to be in the low normal range. They have a specific cognitive phenotype with functional assets and deficits in timing, attention, movement, perception, language, literacy, and numeracy (Dennis & Barnes, 2010). The neuropsychological profile of children with spina bifida and hydrocephalus is often more impaired (Iddon et al, 2004). Shunting, shunt infections, revisions, and history of seizures predict poorer

memory (Dennis et al, 2007), meta-cognitive abilities (Brown et al, 2008), executive functioning (Tarazi et al, 2007) and cognitive health (Kulkarni et al, 2004). In Uganda infants not requiring treatment for hydrocephalus had significantly better neurocognitive outcomes than those who did require treatment (Warf et al., 2009). In Uganda children with spina with better motor function outcomes, are more likely to be in school, and have better cognitive outcomes (Bannink, Fontaine, Idro, & Van Hove, 2016).

Special needs and inclusive education in Uganda

Although inclusion of children with spina bifida in normal schools is common in high and middle income countries, this remains a challenge in low income countries (Chataika et al., 2012). Implementation of inclusive education is affected negatively by non supportive attitudes of parents and community members, distance to school, not conducive school environment (access, materials, equipment), and lack of trained special needs teachers (Abosi & Koay, 2008). In South Africa, physical access, transport, medical information, and training and support of teachers to respond to particular learning disabilities were identified as key areas that need to be addressed to allow children with disabilities to participate in the existing education system (Vosloo, 2009). A study in Uganda shows that children with disabilities are admitted without proper assessment of their educational needs and the resources are not available to provide them with an appropriate range of experiences (Kristensen et al, 2006).

In Uganda Persons with Disabilities (PWDs) are represented in the national and local governance structure. Through the National Council for Children and the National Action Plan for PWD, strategies have been developed to cater for children with disabilities. In 1955, special needs education began with the creation of a school for children with visual impairments by the British, followed by education for children with hearing impairments in 1958. In 1965 the International Labour Organisation (ILO) conducted the first Sub-Saharan National Census of the Disabled and urged government to be more vigilant and active in the provision of education for PWD. In 1973 a special education department was created in the Ministry of Education (USDC, 2003), now called the Special Needs and Inclusive Education department. Since 1991, primary school teachers and other professionals have been trained at the Uganda National Institute for Special Education (UNISE). In 1992, a nationwide network of centres at district level started to address and coordinate special needs education locally (Government-of-Uganda, 2015). In 1997 Uganda endorsed the Universal Primary Education policy, and in line with the Education for All initiative the target group of children with disabilities expanded to including orphans, traumatised children, HIV positive children and others with special needs. A new Basic Education Policy for Disadvantaged Children was endorsed in 2002, which increased the demand for training of staff at all levels (DANIDA, 2005). Uganda signed the UN Convention on the Rights of Persons with Disabilities in 2008. In 2010 the Uganda National Action on Physical Disability (UNAPD) and the Ministry of Gender Labour and Social Development (MoGLSD) launched the Accessibility Standards, which provide guidelines for a barrier-free

environment (UNAPD & Government-of-Uganda, 2010). The Ministry of Education has an inclusive education policy in draft for inclusion of children with special needs in primary schools (Nyende, 2012) Despite all the policies in place implementation of inclusive education remains limited at grass root level (Ejuu, 2016). According to a UNICEF report only 9% of children with disabilities in Uganda attended school in the period 2009 – 2011, and only 6% completed primary school and went to secondary school (UNICEF, 2014).

In this paper we describe the primary school setting for children with spina bifida in the central region of Uganda and explore accessibility and inclusion with parents and teachers.

Methods

Study design

The sample for this study was selected from a larger study population of 139 children with spina bifida in Uganda for which purposeful sampling was used in rehabilitation centres throughout the country to recruit families of children with spina bifida aged 4 to 14 years. For our study we selected all 63 children from the central region of Uganda from the database with 139 children. Of the 63 children, 41 children were schooling. Basic demographic data, motor functioning, continence management, and cognitive functioning data were earlier collected in the larger study of all 63 children and families. In addition we collected school specific information for the 63 children. The parents of the 41 children were approached for permission for a school visit, interview with the teacher, and classroom observation, and 36 accepted. In total 30 schools and teachers consented to participate. Qualitative semi structured interviews and school observations were combined and conducted with 30 parents and teachers between June 2011 and December 2014.

Ethical considerations

Ethical approval and research clearance were obtained from Ghent University, Belgium, the Uganda Virus Research Institute, and the Uganda National Council for Science and Technology. Informed consent was obtained from all parents and teachers, and assent from children and siblings of 8 years and above where possible.

Selected measures

The Index of Inclusion (Booth & Ainscow, 2002) was used as a guide in the interviews with parents, students, and teachers. No full questionnaires were administered or scale scores calculated, as some of the items were not applicable in the Ugandan setting. Instead the overall themes of inclusive cultures, policies, and practices were explored in semi-structured interviews appropriate for the school setting. Questions about relationships and respect between students and teachers were unmerged and rephrased to fit the cultural setting.

We used the Trip Chain concept and drawings of the Ugandan Accessibility Standards (UNAPD & Government-of-Uganda, 2010) to measure accessibility in schools. A Trip Chain is defined as “the sum of all parts of movement from one place to another which must be accessible in order to ensure a barrier-free environment” in the Accessibility Standards. In the case of the children in this study, this included the ability to independently: 1) walk or wheel from home to school on a sidewalk or pathway or use public transport 2) enter the school compound, offices, and classrooms (e.g. ramps available for wheelchair users) 3) manoeuvre within the school buildings and compound (e.g. corridors, inside classroom, sports field) 4) use the toilet facilities 5) use boarding facilities.

Data management and analysis

Basic demographic data were registered during assessments, and entered into a database after completion. The semi structured interviews were transcribed, translated, coded and analysed using thematic analysis in NVivo version 10. Descriptive statistics were calculated from observations and the index questions on school accessibility using SPSS16.

Results

Study population

The study population consisted of 63 parents of children with spina bifida; 30 teachers (22 female, 8 male) of children with spina bifida. The children’s average age was 5.97 (SD 2.20) years. In total, 41 children were schooling, and 22 were not. Of the schooling children, 26 were in nursery school, 11 in primary, and 4 in secondary school. All children with spina bifida had undergone surgery to close their spine (myelomeningocele closure) earlier in life. 22/27 children who had both spina bifida and hydrocephalus had surgery for hydrocephalus. The parent’s average age was 33.3 (SD 5.17) years. The majority (over 80%) of the families was Christian, 18% was Muslim. Table 1 describes the demographics and impairment characteristics of the study population divided into the families of children who were and were not schooling.

Table 1 Demographic and impairment characteristics of the study population (N=63 children with spina bifida)

Variable	Child in school (N=41)	%	Child not in school (N=22)	%
<u>Gender child</u>				
Male	26	63.4%	10	54.5%
Female	15	36.6%	12	55.5%
<u>Type of disability</u>				
Spina bifida	23	56.1%	13	59.1%
Spina bifida and hydrocephalus	18	43.9%	9	40.9%
<u>Child is continent / uses CIC</u>				
Child practices CIC	36	87.8%	17	77.3%
Child does not practice CIC	5	12.2%	5	22.7%
<u>Child gross motor skills</u>				
Cannot walk, has no assistive devices	8	19.5%	8	36.4%
Can walk with assistive devices	12	29.3%	6	27.3%
Can walk without using assistive devices	21	51.2%	8	36.4%
<u>Child fine motor skills</u>				
Average score	15.85	(SD=3.53)	11.7	(SD 4.88)
<u>Relationship parent/caregiver</u>				
Mother	36	87.8%	19	86.4%
Father	1	2.4%	2	9.1%
Other	4	9.8%	1	4.5%
<u>Education level parent</u>				
None	2	4.9%	3	13.6%
Primary	18	43.9%	9	40.9%
Secondary	12	29.3%	5	22.7%
Vocational	4	9.8%	3	13.6%
University	5	12.2%	2	9.1%
<u>Marital status parent</u>				
Single	5	12.2%	1	4.5%
Married	32	78.0%	18	81.8%
Separated	1	2.4%	2	9.1%
Widowed	1	2.4%	1	4.5%

Monthly household income

< 30 euro	0	0%	4	18.2%
30 - 60 euro	7	17.1%	5	22.7%
61 - 90 euro	10	24.4%	2	9.1%
> 90 euro	22	53.7%	9	40.9%

Parent has support from another adult on a daily basis

Yes	26	63.4%	11	50.0%
No	15	36.6%	11	50.0%

Occupation parent

Finance / administration	5	3.8%	2	5.9%
Small scale private business	30	22.7%	7	11.8%
Teacher / education	9	6.8%	3	8.6%
Medical / paramedical	5	3.8%		
Civil service / government	2	1.5%		
Peasant farmer	65	49.2%	19	62.9%
No occupation	6	12.1%	3	8.6%

In total 30 primary school teachers were interviewed at nursery and primary schools where children with spina bifida were enrolled. Children (18 male, 12 female) were observed in class, and parents/caregivers (27 mothers, 1 father, 1 foster parent, and 1 elder sister) were interviewed. In total 16/30 children in school could walk without assistive devices; 6 children used crutches, and 8 used a wheelchair to ambulate. Fifteen had spina bifida, and 15 spina bifida and hydrocephalus. 24 practiced catheterization, 1 used pampers, 5 were continent of urine. The teachers were the child's class teacher and interacted with the child on a daily basis. The class levels varied from lowest level nursery school ('baby class') to primary school level 5 ('P5'). The class size varied between 28 and 69 children.

School enrolment

The demographics and interview data showed that children with spina bifida with better gross and fine motor skills, continence management, cognitive outcomes, higher household incomes, and higher levels of parental support were more likely to be in school than those with lower outcomes on these variables. Parents described this as following: "I went to 5 different schools but none of them would accept him, they said they cannot manage with a wheelchair and his continence management needs"; "I have 6 children and cannot afford to send all to school. As she has some difficulties in understanding, I decided to send the other ones first". Parents whose children were not in school indicated this was due to insufficient income (12/22), the child not being able to go to school due to poor physical and cognitive functioning (8/22), and the child still being young (2/22). The latter was only mentioned for children of 4 years of age. The sample was too small to carry out reliable statistical testing. All children who were schooling were in private schools. Only 4 parents had tried

enrolling their child in a public school, but the children had not been welcomed and were bullied, after which parents tried to secure funds for a simple private school: “I had no money, I had to send him to a government school, but the other children abused him, and the teacher was not interested in helping him, so I took him out after 1 term”.

Physical accessibility of school facilities

In total 24/30 parents and 17/30 teachers felt the school was accessible for their children, 24 parents felt it was, compared to 17 teachers. In the school observations, we noted that parents and teachers rated the school accessible if their child could enter their classroom without help. We observed that only 7 out of the 30 schools had classrooms which were accessible for wheelchair users and children using crutches using the Accessibility Standards. In two (2/30) of the schools cemented ramps were constructed, three (3/30) had installed wooden ramps, and two had a marram ramp (which was not functional in the rainy season). Parents paid for the wooden ramps.

None of the classrooms had space to manoeuvre a wheelchair in. The other school offices, libraries, and sports fields were not accessible according to the Accessibility Standards.

In 5 out of the 30 schools children could practice continence management and had accessible toilets. Parents explained practicing CIC was a major challenge, as the schools only had normal latrines which were generally unclean and have no place or seat to put the CIC materials and practice CIC. However in 3 of the schools the children used the teacher’s toilets, in 2 schools actual bars and ramps were constructed to make the student latrines accessible.

None of the roads to school were accessible for children using assistive devices: 22 out of the 30 children were brought to school by their parent(s), a ‘*boda boda*’ (4, local motorbike transport), or their sibling pushing their wheelchair (2). The other 2 could walk to school by themselves. None of the children using assistive devices could ambulate to school on their own without assistance.

None of the children was in boarding school. The school with boarding sections had no accessible entrances to the dormitories, no accessible bathrooms, and insufficient space to manoeuvre in.

Classroom participation

All teachers were solely responsible for their class and had no teaching assistants. All schools were private schools and had desks, chairs, and a blackboard in the classrooms. Some of the more expensive private schools had a variety of toys and books available in the nursery section. In primary schools, textbooks were not available in class for children, 8 schools had a library where children could access the books. In the primary schools and most nursery schools, children copied notes from the blackboard or transcribed what the teacher was reading out. Teachers did not have tools or materials to offer diversified teaching activities. None of the children who were using assistive devices (14) participated in physical education / sports lessons.

The teachers felt they try their best to teach all children in class, and expressed to find it difficult to find enough time to help each child individually. This makes it harder to teach the child with spina bifida well they say, as they often need more attention and time.

Most teachers (24/30) felt they included the child by making sure the child sat in front of the classroom: "so I can see her well and check if (s)he is following", spending some extra time to make sure they understood the exercises and work given: "I check if he has copied the assignment well and understands it", and discussing their physical needs with the parents: "I told his mother she needs to buy a table desk for his wheelchair".

Teachers explained they often ask other students to help the child with spina bifida to go outside, or copy notes for them if writing is difficult or slow, or the child has missed class. They would not ask the child with spina bifida to help another child.

Of all the children who practiced CIC teachers mentioned the challenge in practicing CIC during the normal school day: "she needs to go to the bathroom during the lesson at certain hours and it takes long, then she misses a lot of what we are doing in class"; 'his mother comes to take him out to urinate, but when she cannot come, he smells of urine and the other children complain". When asked if teachers could assist five (5/30) teachers explained a teacher or school nurse is assisting the child, but most felt it was the child and parent's responsibility to organize CIC in school.

During the classroom observations, it was noted that children with spina bifida were called to answer questions and participate in class activities. It was observed that the children often sat in front of the class, closer to the blackboard. In most schools teachers explained that this was due to vision or ambulation problems, e.g. "he has difficulties seeing, so it is better to sit close to the blackboard" and "there is little space for the wheelchair in the back, here in front it is better for her".

Educational performance

We asked parents and teachers to rate the child's performance as poor, fair, or good. Whilst 11 parents felt their child performed well, only 7 teachers did; 11 parents and 10 teachers felt the child performed fair; and 8 parents and 13 teachers felt the child performed poorly.

The majority of teachers said the children are slow learners compared to the other children in class. When asked about performance in different subjects or tasks most class teachers responded "it is not so good" or "(s)he needs to improve". Some were dismissive in whether or not this child would be able to complete school and pass examinations, especially in those with hydrocephalus. One teacher felt it was not useful to send the child to school "it is a waste of money, he does not understand most of what we teach, I think he will stop after P2 [primary 2, second year of primary school]". Only 4 out of the 30 teachers felt the child would be able to complete secondary school, and only 2 said they felt the child could go to university: "it will be difficult to complete A-level [secondary examinations to qualify for university admission], but maybe she can try, she is bright".

Parents were concerned about their child's performance and felt there was no room to adjust or understand their child's difficulties in reading or processing information. None of the children had

received any cognitive assessments prior to this study nor had families been informed about possible cognitive deficits and assets related to spina bifida. Five (5/30) parents said to have meetings with the teachers regularly and felt the school informed them about their child's development and needs adequately; these were parents whose children were enrolled in more expensive schools which had more facilities and teachers available.

Inclusive school policies

The teachers interviewed explained their school follows the regulations and curriculum of the Ministry of Education and Sports. This includes giving children with disabilities a place to study. However they explained that despite these policies being in place it is often difficult for the children to participate as the number of children in class is very high, and the school have very limited resources in making their buildings accessible and teaching materials available.

Teachers said they expect the children with spina bifida to participate in class like any other child. They said they are not expected to participate in physical education if they cannot walk. Homework expectations are the same as for the other children. Homework instructions are copied from the blackboard or in a few occasions given on a piece of paper and are usually expected to be handed back the next day. No extra time is provided for examinations whether or not the child appears to have learning difficulties. Report cards are based on the national curriculum and marking system. There are no individualized score cards, education plans, or appraisal systems.

Teachers explained that whilst bullying is discouraged it often happens as children can be rude to each other. Half of the teachers felt it was part of their role to prevent bullying and take action if the child with spina bifida was bullied in class. The other teachers felt it was the school management and parents to make sure that they dealt with such behaviour. Some said it is the parents fault: "if you [parent] send your child here and he cannot walk and has no wheelchair they will make fun of him when he is crawling". Parents explained their child is sometimes bullied in school. Most of the bullying was centred on their physical appearance and slow learning. Some tried to address the bullying through talking to the school administration without much result. Almost a third (9/30) of the parents changed school at least once because of bullying. Parents did mention that if children are in school for a longer time, the bullying reduces as children get used to them.

Discussion

Schooling for children with spina bifida in Uganda is very challenging. Only 65% of the children in our study population were schooling. Nationally the net enrolment ratio in primary schools in the same period was 94.5% (Government of Uganda, 2013). The main reasons for not being in school for children with spina bifida were poor physical and cognitive functioning, and lack of income to pay for school fees. Although public schools exist, none of the parents felt their child could be included in these. In earlier publications we found most children enjoy going to school, but parents indicated

finding a school that will admit and include their child is challenging (Bannink, Stroeken, et al., 2015). Parents in our study had to pay tuition fee for their child in the private schools, and a child had to often do an entrance examination. Earlier we found parents household income affected schooling (Bannink, Fontaine, et al., 2016), and parents would not prioritize sending their child with spina bifida to a private school if they did not have sufficient funds to send all children to school (Bannink, Idro, et al., 2016a). As schooling in turn will affect cognitive outcomes positively (Sylva, 1994) (Bannink, Fontaine, et al., 2016), the chances to enrol into school later, e.g. when funds are available, may decrease even further as the gap between the child's development and those of their school going peers will enlarge, and the chance to pass the examination test reduces.

For those in school, physical accessibility of school facilities was very limited. Looking at the Accessibility standards, none of the children could walk or wheel from home to school on a sidewalk or pathway or use public transport; only a few could enter the school compound, offices, and classrooms (e.g. had ramps); none could manoeuvre within the school buildings and compound (e.g. inside classroom, sports field) and none could use boarding facilities. Toilet access and incontinence management was the main challenge for the children. Although some schools attempted to create accessible toilets to practice CIC, the majority did not have these facilities available. The physical impairment and physical access difficulties in education were emphasized by parents and teachers, more than cognitive difficulties or social inclusion.

Teachers did explain they were limited by the lack of materials, high numbers of students in class, and teaching methods. Nationally the pupil to classroom ratio is 57, and teacher to student ratio 45. Being inclusive in the classroom was mostly seen as ensuring the child could enter the classroom, offering him or her a position near the black board, and ensuring the child would be able to copy the notes. Teachers were not conversant with the possibility of children's participation in physical education / sports whilst using assistive devices. Further support and activities to increase participation in sports are recommended. Private non for profit organizations have started organizing sports days, and are promoting sports for children with disabilities (CoRSU, 2015; Mukisa-Foundation, 2015).

Knowledge of the effect of cognitive outcomes on inclusion was limited in the schools we visited. Although teachers and parents explained that the children are 'slow learners', none was aware that spina bifida comes with specific cognitive assets and deficits (Dennis & Barnes, 2010). Some of the teachers who defined children as 'slow learners' tried to spend more time with them or break down tasks for them, however no clear strategies were in place to address learning difficulties. Educational and neuropsychological testing is very limited in Uganda. Bangirana et al (2009) have created a Ugandan validated test battery which is used to assess neuropsychological functioning in the national referral hospital Mulago in Kampala, the capital city (Bangirana et al., 2009). Earlier we noted children were unable to understand or complete a number of subtests from this battery due to their physical impairment (Bannink et al, 2016). We argue for awareness raising on the cognitive

profiles of children with spina bifida and strategies to help them in learning in schools rather than individual testing of the child as a strategy to address their 'slow learning'. The International Federation for Spina Bifida has sensitization materials available on inclusion of children with spina bifida in schools (IFSBH, 2010). These could be adjusted and translated for use in low income countries.

Bullying in schools is not well controlled and it is unclear if the responsibility for this lies with the parents or teachers. Awareness raising on the impairment, inclusive education, and more active implementation of child protection policies in school may help prevent and stop bullying. The Ugandan Ministry of Education and Sports, Ministry of Gender, Labour and Social Development in collaboration with international and national partners have implemented various child protection programs in schools over the past decade (AVSI, TPO, CARE & FHI, 2015; Government of Uganda, 2012, 2014b). Further efforts are required to protect children with disabilities in schools and communities. E.g. training of teachers should be reoriented to give teachers the capacity to diversify teaching (Ejuu, 2016). The schools in our study followed the national curriculum and did not offer diversified teaching, nor readers, scribes or extra time in examinations. To diversify teaching methods, a revision of the national curriculum and method of teaching may be required, as use of visual and image activities, play, art, and use of assistive electronic devices in schools is currently not available. Having embraced the Education for All objectives, there is need to critically look at the way children are involved and taught in class to enable teachers and schools to achieve this.

Conclusion

To achieve an inclusive education community, we recommend awareness raising to reduce discrimination, training and on job mentoring to support teachers and schools, and earmarking funds for inclusiveness in schools for children with disabilities.

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Chapter 5 Family relationships and care



5.1 Parental stress and support of parents of children with spina bifida in Uganda⁷

Abstract

Children with disabilities in Sub-Saharan Africa depend for a large part of their functioning on their parent or caregiver. This study explored perceived stress and support of parents of children with spina bifida living in Uganda and the factors that influence it.

In total 134 parents were interviewed. Focus group discussions were held with 4 parents support groups in 4 different regions within the country. The Vineland Adaptive Behaviour Scales (VABS) Daily Functioning Sub Scales and Parental Stress Index Short Form (PSI/SF) were administered to measure the child's daily functioning and parental stress levels.

Parental stress was high in our study population with over half of the parents having a >90% percentile score on the PSI/SF. Stress outcomes were related to the ability to walk (Spearman's correlation co-efficient [ρ]=-.245), continence (ρ =-.182), use of clean intermittent catheterization (ρ =-.181) and bowel management (ρ =-.213), receiving rehabilitative care (ρ =-.211), household income (ρ =-.178), geographical region (ρ =-.203), and having support from another parent in taking care of the child (ρ =-.234). Linear regression showed parental stress was mostly explained by the child's inability to walk (β =-.248), practicing bowel management (β =-.468), and having another adult to provide support in caring for the child (β =-.228). Parents in northern Uganda had significantly higher scores compared to parents in other regions (Parental Distress $F=5.467^*$; Parent Child Dysfunctional Interaction $F=8.815^{**}$; Difficult Child score $F=10.489^{**}$).

Parents of children with spina bifida experience high levels of stress. To reduce this stress, rehabilitation services should focus on improving mobility. Advocacy to reduce stigmatization and peer support networks also need to be strengthened and developed.

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Introduction

Spina bifida in Uganda

Spina bifida is a neural tube defect, a congenital abnormality causing disability, whereby the spinal cord and vertebrae do not form completely and the neural tube fails to develop normally. The worldwide incidence of spina bifida varies between 0.17 and 6.39 per 1000 live births (Bowman et al., 2009; Kinasha & Manji, 2002; Msamati et al., 2000; Shaer et al., 2007). In Uganda incidence data is not available. Warf et al estimated a birth incidence of 1 in 1,000, translating into 1,400 children born with spina bifida in the country annually (Warf, Wright, et al., 2011).

Most children with spina bifida have some degree of paralysis, which affects mobility as well as bowel and bladder control (Northrup & Volcik, 2000) (Verpoorten & Buyse, 2008). This affects participation in daily activities (Danielsson et al., 2008) (Jansen et al., 2009). Sixty-six per cent of the children with spina bifida develop hydrocephalus (Warf & Campbell, 2008).

Surgery and rehabilitative care is expensive and inaccessible for many born with the disability in Africa. Families need to find resources for children with disabilities while already constrained. In Uganda, the initial surgery (closure of the spine) at the time of this study was only available in Mulago National Referral Hospital in Kampala, and CURE Children's Hospital in Mbale, eastern Uganda. Recently Mbarara Regional Referral Hospital started offering the same. In north, west, and central Uganda, three rehabilitation centres offer specific occupational therapy, physiotherapy, continence management, and social support services for children with spina bifida and their families in Uganda. Policies such as the United Nations Convention on the Rights of Persons with Disabilities and Disability Act amongst others are in place, and efforts are made by the Ugandan government and not for profit organizations and hospitals to provide basic services for children with spina bifida and other disabilities (Oyaro, 2014) (Mertens & Bannink, 2012). Despite these efforts, poverty continues to affect access to services for the majority of the population, and more so families of children with disabilities (Bannink, Stroeken, et al., 2015; Lwanga-Ntale, 2003; Miles, 2002).

Parental stress and disability

Parental functioning is of great importance for children with severe disabilities who depend for a large part of their functioning on the parent or caregiver. Research on family functioning and psychosocial adjustment of families of children with spina bifida in high income countries support a resilience–disruption view of family functioning, whereby the presence of a child with spina bifida disrupts normative family functioning at first, but after a period of time families adapt and exhibit considerable resilience (Holmbeck et al., 1997; Vermaes et al., 2007). The level of impairment has not been related to the general level of family functioning in Europe and the United States (Ulus et al., 2012; Wiegner & Donders, 2000). Nevertheless spina bifida has been found to have negative medium-large effects on parents' psychological adjustment and functioning (Holmbeck & Devine, 2010; Vermaes, Janssens, Bosman, & Gerris, 2005; Vermaes, Janssens, Mullaart, Vinck, & Gerris, 2008).

In Vermaes' review, child, parent, family, and environmental factors were found to be associated with variations in parents' psychological adjustment (Vermaes et al., 2005). Mothers are often at higher risk for parenting stress than fathers due to role differentiations in care and work (Vermaes et

al., 2008). Children with hydrocephalus (literally 'excess water in the brain') are often in need of neurosurgical treatment which may involve insertion of a shunt or creation of a bypass within the brain to allow the cerebrospinal fluid drain. Shunts are prone to infections which can be life threatening. For children with both spina bifida and hydrocephalus such shunt dependency has been associated with higher levels of parental anxiety and depression (Malm-Buatsi et al., 2015).

Cultural differences have been described in a study of Hispanic parents in the US, in which mothers of children with spina bifida were found to be at risk for lower feelings of satisfaction and competence as parents compared to non Hispanic parents (Devine, Holbein, Psihogios, Amaro, & Holmbeck, 2012). In Malaysia mothers of children with spina bifida had significantly higher stress scores compared to mothers with children without a disability (Ong et al., 2011). In the same country, clean intermittent catheterization was the only medical factor associated with such stress. This is a technique used to empty urine from the bladder using a catheter. Most children with myelomeningocele, the severe type of spina bifida in children involved in this study, are incontinent of urine and benefit from practicing intermittent bladder catheterization as this helps them to participate in daily activities without losing urine (IFSBH, 2014). Parental stress in Malaysia was also mediated by single parenthood, caregiver status and the child's adaptive skills (Kanaheeswari et al., 2011).

African studies on parental stress in parents of children with spina bifida are limited. In South Africa Greeff and Nolting showed that families of children with developmental disabilities adapt better when they are more accepting of the situation, have more investment in the family unit, and have positive patterns of communication and attitudes towards challenges (Greeff & Nolting, 2013). In Nigeria and Tunisia, parents of children with neurodevelopmental disabilities and learning disabilities had higher rates of depression and anxiety compared to parents of children without a disability (Abasiubong, Obembe, & Ekpo, 2006; Ben Thabet et al., 2013). In Malawi parents of children with neurodisability following brain injury experienced barriers to care and support (Paget et al., 2015). Similar findings were seen in South Africa in families of children with mental health disabilities (Coomer, 2013). In Kenya mothers of children with spina bifida struggled with the financial implications of the child's disability; most of them received some support from other parents and religious communities (van't Veer et al., 2008).

In this paper we aim to explore perceived stress and support of parents of children with spina bifida living in Uganda and examine the factors that influence it. By understanding the stress factors, we hope to inform health and community based rehabilitation services for children and parents with spina bifida in the country. We believe parental stress and support is highly dependent on the cultural and socio-economic context where parents live. Contextual factors such as the differences in rural and urban setting, and the 22 year conflict between the Lord's Resistance Army and the Government of Uganda which displaced over 2 million people in the north of the country (Muyinda & Whyte, 2011), will be taken into account.

Research methods

Ethical considerations

Ethical approval and research clearance for the study were obtained from Ghent University, Belgium, the Uganda Virus Research Institute, and the Uganda National Council for Science and Technology. Informed consent was obtained from all parents.

Recruitment of participants

Selection of participants was performed in Mbarara, Kampala, and Mbale where CURE holds bi-monthly clinics. CURE hospital and the partnering rehabilitation centers in Kampala and Mbarara were requested to list all children registered in their follow up programs, and inform them during home visits and through telephone calls to attend the clinic. In Gulu and Lira where no follow up system or registry of the children was in place at the time, radio announcements were aired to inform parents about the upcoming review clinics in the area, and specifically invited parents with children with spina bifida between the age of 4 and 14 to attend.

Data collection

Explorative mixed study methods were employed to explore factors affecting parental stress and support in the Ugandan setting (Creswell & Clark, 2007; Singh, 2007). Both quantitative and qualitative data was collected concurrently from 134 parents between June 2011 and December 2014.

Quantitative instruments were used to explore daily functioning and stress levels. A selection of 10 items of The Vineland Adaptive Behaviour Scales (VABS) Daily Living Skills subscale relevant to the Ugandan setting were used. Items included measures of daily functioning tasks such as removing a jumper, drinking from a cup, washing ones' face, fetching water and dressing independently. Items were scored 2 (behaviour is usually or habitually performed), 1 (sometimes or partly performed), or 0 (never performed) (Sparrow et al., 1984).

The Parental Stress Index Short Form (PSI/SF) consists of 36 items scored on a 5 point Likert scale. The items are divided over 3 subscales: Parental Distress (PD), Parent-Child Dysfunctional Interaction (P-CDI), and Difficult Child (DC). A total stress score is computed from the three subscales and indicates the overall level of parenting stress in the areas of personal parental distress, stresses derived from the parent's interaction with the child, and stresses that result from the child's behavioural characteristics. Parents who obtain a total stress score above 90 are experiencing clinically significant levels of stress (Abidin, 1995).

The presence of a (house) helper or other adult at home involved in the care of the child alongside the primary caregiver was registered as a measure of perceived support. Membership of a parent support group was documented to understand wider support networks.

Qualitative methods were used to explore the day to day reality of parents of children with spina bifida in the Ugandan setting. Semi structured interviews were held with all 134 parents. In addition, focus group discussions were held with 4 parents support groups, in Gulu, Kampala, Mbale, and Mbarara respectively. Questions for the interviews and focus group discussions were formulated to

develop a more in-depth understanding of perceived parental stress, coping strategies, and the role of support from others. Specific focus group discussions with parent support groups were held to further understand their activities and possible contributions in reducing parental stress and increasing perceived support. In total 5 of the 134 parents took part in the focus group discussions, the other members of the focus groups (26) were not interviewed individually.

The interviews with the parents were held in the local language of the area, and a translator was hired and trained for each area to assist in conducting the interviews and focus group discussions. Some of the interviews were conducted in English, if parents were fluent and requested for this.

Data analysis

The semi structured interviews and focus group discussions were transcribed, translated, coded and analysed using thematic analysis in NVivo version 10. Basic demographic data, details on the child's impairment, social support and scores for the VABS Daily Functioning Subscale and PSI/SF were entered and analysed using SPSS16. The sub-total scores for each subscale was calculated and compared with >90% percentile scores from the PSI manual which indicates high levels of stress. To understand relationships between demographic and impairment variables and parental stress levels Spearman Rho's correlations were computed. To compare regional differences a MANOVA between PSI scores between the various regions was carried out. Linear regression analysis was conducted to understand which factors mostly contributed to parental stress.

Results

Description of study participants

Table 1 describes the demographics of the study population. Parents' ages ranged from 24 to 46 years with an average age of 32.9 (SD 5.2) years. The majority of the parental respondents were the mother to the child, followed by fathers and grandmothers. Over 75% was married and Christian. Almost half of them were farmers.

The mean age of the 80 (59.7%) male and 54 (40.3%) female children with spina bifida (myelomeningocele type) was 6.1 (SD=2.0) years, ranging from 4.0 to 14.0 years of age. Only 76 (57.3%) of them were going to school: 49 (37.4%) were in nursery school, 22 (16.8%) in primary school and 4 (3.1%) in secondary school. The household size ranged from 2 to 13 with an average of 6.5 persons per household; an average 4.3 children (SD 2.2) and 2.3 adults (SD 1.0) per household. The average monthly household income is 82 euro (range 12 to 604 euro). The majority of the children (128, 95.5%) had undergone surgery to close their spine (myelomeningocele closure) earlier in life. Of the 61 children who had both spina bifida and hydrocephalus, 22 (35.9%) had undergone endoscopic third ventriculostomy while 1 (18.6%) had ventriculo-peritoneal shunts placed. Only 2 (3.6%) of the children of parents in the study never had surgery. Most parents took their children (118, 90.8%) for rehabilitation services such as physio- and occupational therapy.

Parents' Stress Index Outcomes

The parent stress index questionnaire was administered to all parents. Scores were normally distributed for all items including the total subscales. We excluded 3 cases from the analysis as scores showed defensive responding (score <10). Table 2 shows the subscales scores. More than half of the parents (52.7%) scored above the 90% percentile on total stress. Higher scores were seen on the Parent Child Dysfunctional Interaction domain with 48.1% (63) scoring above the 90% percentile, followed by the Difficult Child Domain with 32.8% (43) scoring above the 90% percentile, and the Parental Distress Score in which 14.5% (19) of the parents scored above the 90% percentile.

Spearman's Rho correlations were calculated for demographic variables, impairment related variables e.g. incontinence and mobility, daily functioning skills, and perceived support from another adult and parents support groups. Table 3 shows the variables which had significant Spearman rho correlations. Variables with significant correlations with PSI outcomes were the child's ability to walk, continence and use of clean intermittent catheterization and bowel management, receiving rehabilitative care, household income, location (region where the child and family lives), and having support from another parent in taking care of the child. Household income correlated positively with school going, region, parental education, the use of assistive devices, receiving rehabilitation services, using catheterization, and having support from another adult.

Based on the significant correlations found, linear regression analysis was performed to investigate factors that were determinants for high scores in the parental distress (PD), difficult child (DC) and parent-child dysfunctional interaction (P-CDI) subdomains of the PSI/SF.

All variables with significant correlations were entered in a stepwise regression model. The best predictive models are displayed in table 4. Results are expressed as beta coefficients, R square and F values in table 4. The ability to walk, having support of another adult, and the location where the child and family are residing were the significantly contributing predictors.

MANOVA results showed parents in northern Uganda had significantly higher >90 percentile scores on the PSI subscales compared to parents in other regions: parental distress $F=5.467^*$; parent-child dysfunctional interaction $F=8.815^{**}$; difficult child $F=10.489^{**}$. No significant differences were found between the other regions.

Children's mobility and incontinence

The inability to walk was the largest contributor to parental stress and perceived parental difficulty in managing and caring for the child. Parents of children with both spina bifida and hydrocephalus had higher scores on dysfunctional interaction between parent and child compared to those with only spina bifida only.

The majority of the children with spina bifida whose parents were interviewed were able to sit without assistance, and could speak. Half of them were able to walk without assistive devices. Of the ones who were in need of assistive devices, a third used crutches, another third a wheelchair, whilst the others had no access to assistive devices and crawled. In our study population 88.6% of the children were incontinent, and 81.1% of them used clean intermittent catheterization to manage the incontinence. Table 5 shows the various percentages for the impairment related variables.

In the interviews, parents indicated mobility was a major challenge and stressor. Many parents narrated how they carried their children on their back to the land they cultivate (most of them are subsistence farmers), to public transport and sometimes to school when the children were young. Some still continue to do so as assistive devices are not always available or applicable for use in their setting.

“She is getting heavy now. She was easy to carry, she is still a bit small, but her head is heavy. I have no alternative; there is no one to help at home, so I have to bring her with me to the garden when I go to dig. I put her under a tree in the shade.” (Parent of a 5 year old girl with spina bifida in northern Uganda)

“When it is rainy season our roads are too slippery for him. He can’t go to school then. On other days the wheelchair is good, we can push him, and he tries to use it himself.” (Parent of an 11 year old boy with spina bifida in western Uganda)

Although incontinence did not explain parental stress for a large part on the PSI outcomes, apart from practising bowel management on the parental distress subscale, in the focus group discussions and interviews the majority of parents said incontinence was the most complex factor in managing their child’s health, more than the (partial) paralysis. Whilst the majority did practice catheterization, parents complained practicing catheterization and bowel management outside home and the rehabilitation posed a big challenge. Managing their children’s catheterization meant that parents had to be close by and available to help the child practice catheterization e.g. at school every 6 hours. Accidents or not well managed catheterization also caused stress as the child would be wet and smell, which would result to others commenting or pitying the parents.

“We have used catheterization since my child was 2 years old. We are used to it, but when we go out or I am away, it is difficult. In most public places there is no place to practice catheterization. We have to do this behind the latrine in the open, which is not good.” (Parent of a 4 year old daughter with spina bifida and hydrocephalus in western Uganda)

Parents’ support systems

Almost half of the parents (63, 47.7%) mentioned they have support of another adult in the household to care for their child with spina bifida. Having another adult to support in providing care significantly contributed to having less dysfunctional parent child interaction on the PSI outcomes. This other adult would be a spouse, relative, or house help. In the focus group discussion and interviews parents who did not have support from another adult, explained they found it very stressful to be constantly responsible for their child and often felt isolated, as they could not attend family and community functions due to their care obligations. Whilst some could leave their other children with neighbours or friends, they said it was hard to find someone who would be ready to take care of their child with spina bifida. Parents in the north and east had less support from other adults compared to those in the central and western regions.

“I don’t have a house help, when I need to go somewhere I have to take her with me. I can’t leave her with the neighbours; they don’t know how to look after her.” (Parent of an 8 year old daughter with spina bifida in eastern Uganda)

In total 55 (41%) of the parents was a member of a parent support group. No interaction was found between PSI outcomes and membership of a parent support group. Table 6 provides details of the reasons of (not) participating in a parent support group. From the interview those who were members of a parent support group indicated it mostly helped them to learn more about taking care of their child (50.9%), followed by feeling encouraged by other parents (14.5%), enjoying sharing experiences and learning from each other (14.5%), and income generating opportunities (12.7%).

“Coming to the parent support group is helpful because they teach me how to do catheterization and they understand my problems.” (Parent of a 4 year old girl with spina bifida in the western region)

The majority of parents who were not members of a parent support group said they were not aware parent support groups existed (55.1%), some mentioned they had heard about parent support groups but lived too far away from the rehabilitation centre to attend meetings regularly.

“I did not know there is a support group [...] Also I live far away, I travel 3 hours to Gulu [where the PSG meets]. It is not easy to attend” (Parent of a 7 year old boy with spina bifida in the northern region)

To explore the support given by the PSG further, focus group discussions were held with 26 mothers and 6 fathers active in parents support groups in Gulu (9; 8 mothers, 1 father), Kampala (6, 5 mothers, 1 father), Mbale (8; 6 mothers, 2 fathers), and Mbarara (9; 7 mothers, 2 fathers).

Parents explained the groups started with help of the rehabilitation centres in 3 sites; in Gulu the group started based on a community based rehabilitation initiative which was started up during the conflict in the area. Their main objectives of the groups were to motivate and support each other in caring for their children, provide information and training on care and rehabilitation, and create awareness in their communities about the disability. Parents said they themselves enjoyed participating in the PSG as they would learn more about taking care about their child, and were able to advise and encourage others. The majority indicated that the feeling of ‘not being alone’ in this situation, and being able to share specific challenges with others with similar experiences, helped them to continue caring for and love their child more.

“When I attended the first time, I was so surprised and happy I was not the only one with a child like mine. There were children who could not even sit. The other parents were so encouraging; they gave me hope and a lot of information and ideas on how to help my child and myself.” (Parent of a 7 year old daughter with spina bifida and hydrocephalus in central Uganda)

Some parents mentioned the group had given them new friends, and support networks. E.g. in case of advocating for children, parents in Kampala and Mbarara gave examples on how other parents would join them in visiting schools trying to enrol their child. Many schools would refuse children with physical impairments, but if going as a group or pair, head teachers were said to take their call more seriously and were more likely to give them a space. Similar advocacy took place on inclusion and reducing stigma in communities, where parents would move around with their children explaining the impairment to community members, and need for their children and families to be included. In Mbale most members felt the group gave them spiritual support too.

“I had gone to 4 schools and every time the head teacher said they can’t manage my child in school. There was a parent in the group, she had the same problem but had managed to find a school. She was a very strong woman. She came with me and spoke with the head teacher of a school near our home. He accepted and now my child is in primary 1.” (Parent of an 8 year old daughter with spina bifida in western Uganda)

“The PSG helps you to remember to come for follow up and to work hard. It helps to talk with other parents and to know how to do catheterization. We pray for each other and thank God for our children and we tell others about them so they stop discriminating us.” (Parent of an 11 year daughter with spina bifida in eastern Uganda)

In northern Uganda most parents felt the group gave them an opportunity to start income generating activities together, they had started a rotating loan scheme which was helping members to set up small businesses. They especially felt stigma was high in their area, and support for them was difficult as some people believed the impairment was contagious. Similar activities were mentioned in Mbarara and Kampala. Stigma was mentioned in these areas too but less directly connected to physical contact as in the northern region.

“Here [in the north] people think our children’s disability is contagious, they fear us, they avoid us. In the PSG we can work together, we understand each other. We bought goats for the group, and when they produced we all got one. We started planting maize in the last rainy season together too.” (Parent of a 4 year son with spina bifida and hydrocephalus in northern Uganda)

Discussion

This study describes parental stress and support of parents of children with spina bifida in Uganda. PSI scores were high for our study population with more than half scoring above the PSI-SF cut off point for clinically significant levels of stress (Abidin, 1995). Stress outcomes were related to the inability to walk, continence and use of clean intermittent catheterization and bowel management, receiving rehabilitative care, household income, region, and having support from another parent in taking care of the child. Parental stress was mostly explained by the child’s inability to walk, practicing bowel management, and having another adult to provide support in caring for the child.

Our study does not support the view that the level of impairment is not related to the general level of functioning or severity of the disability as earlier explained in studies of families with a child with spina bifida in Europe and the United States (Ulus et al., 2012; Wiegner & Donders, 2000). Not being able to walk has great implications in an environment in which assistive devices are not easily accessible and where the environment is disabling. There are no social services or individual disability grants provided by the Government of Uganda to families with children with disabilities. As narrated by parents in the qualitative data, parents are responsible for carrying their children to school and feel that even if a child has a wheelchair, they often cannot use this on their own. Earlier we showed how children with spina bifida had more difficulties in daily functioning than their siblings, which played an important role in inclusion (Bannink, Idro, et al., 2016a).

Parents expressed stress around incontinence management. Due to high and frequent involvement of parents on a daily basis, this could increase stress to parents of which the majority is either farming or working and not in position to attend to their child every 4-6 hours as required for clean intermittent catheterization. Prior to self-catheterisation, which can be started from the age of 6 with normal intelligence and fine motor skills, parents are the primary helpers in catheterization practicing catheterization (Robinson, Cockram, & Strode, 1985). The responsibility and anxiety around incontinence has a social and psychological impact on families (Borzyskowski, Cox, Edwards, & Owen, 2004). In our study parents pointed out catheterization interfered with their lives, as they sometimes had to make extra trips to school to assist in catheterization. Similar concerns were raised in Kanaheswari's study in Malaysia (Kanaheswari et al., 2011).

In our study parents in the northern region expressed higher stress levels than those in other areas. Northern Uganda suffered a 22 year long conflict between the Government of Uganda and the Lord's Resistance Army in which 90% of the population was displaced (Muyinda & Whyte, 2011). Together with challenges related to recovery from this conflict, parents in this area face more stigmatization due to negative cultural beliefs about the impairment (Bannink, Stroeken, et al., 2015). This is likely to increase stress and have more need for support from other parents as expressed in the interviews and focus group discussions.

We did not find differences between mothers and other caregivers in stress levels. Mothers are often at higher risk for parenting stress than fathers due to role differentiations in care and work (Vermaes et al., 2008). Kanaweshari (2011) found that mothers who were the sole caregiver had higher parenting stress scores in the P-CDI domain (Kanaheswari et al., 2011).

In our study we did find that having support from another adult contributed to less stress. Support here was described by parents as household support and care giving support at home, e.g. through a house help or relative. This support reduced dysfunctional interaction between parent and child. Parents who were a member of a parent support group felt this helped them in taking better care of their child and felt encouragement from other parents.

Persons with disabilities typically live in poorer than average households, and have lower educational attainment (Emmett, 2006; Filmer, 2008; Lwanga-Ntale, 2003). The monthly income of families in our study ranged from 28 to 689 USD dollars with a median of 82 USD dollars (income derived from all sources includes wages, market sells, cattle, land and other assets). This is much lower than the total national average of 156 US dollars (converted from Ugandan Shillings), though closer to the average rural income of 112 USD and regional variations (Uganda Bureau of Statistics, 2014).

Although no significant effect was seen on parental stress in the regression analysis, looking after a child with spina bifida raises financial costs for families in terms of medical treatment, rehabilitation, and transport. Correlations were found between having higher income and school going, the use of assistive devices, receiving rehabilitation services, using catheterization, and having support from another adult.

Whilst each of these factors on their own may not contribute to parental stress, parents with higher household income are expected to be able to respond to their children's medical needs better than

those who have less. Parents who participated in parent support groups indicated that they benefitted from income generating activities the group organized.

We noted that less than half of the participant's children had both spina bifida and hydrocephalus. Given the earlier established rates of 66% hydrocephalus in children with spina bifida (Warf et al, 2011), this is low. We suspect that more children with hydrocephalus died compared to children with only spina bifida, as shunt complications and failure is high and life threatening, especially in rural areas. Whilst Warf et al (2011) found no relationship between survival and presence of hydrocephalus, we suspect this may have an effect for the older group of children targeted in this study, especially those living in hard to reach areas (Warf, Wright, et al., 2011).

Participation in parent support groups did not make significant changes in parental perceived stress in the quantitative outcomes. However from the qualitative outcomes we see that the sense of belonging and being able to share, was an important support for parents and enabled them to cope with stress better. Malm-Buatsi found that involvement in recreational activities with other families affected by SB was associated with more positive parenting characteristics (Malm-Buatsi et al., 2015). Our study did not study parenting styles, but did receive positive feedback from parents about improved interaction with their child after having participated in parents support groups. Lack of knowledge and acceptance of a child with spina bifida may lead to neglect. Parent support groups aim to raise awareness and support for parents to look after their children positively. Warf, Wright et al earlier described finding situations of child neglect during home visits in the eastern region (Warf, Wright, et al., 2011).

This study was limited by involvement of parents of children only who were receiving or are attending follow up and rehabilitation care. This means we interviewed those who may have been doing 'better' than others, as we only found those whose children accessed surgical care and survived.

Although the impairment factors and support explained some of the parental stress, a large part remained unexplained in our study. We expect a complex combination of child, parent, family, and environmental factors may contribute to parental stress in a low income setting. Earlier Vermaes et al (2005) found a combination of these factors explain parents' psychological adjustment (Vermaes et al., 2005).

Further studies need to be conducted to understand the interaction between poverty, survival, and functioning of children with spina bifida and their parents in low resource settings. Participation in parent support groups could be a protective factor for the child, reducing child neglect. We suggest analyzing this contribution in greater detail and tailoring interventions to raise awareness and prompt action of support networks and state actors to ensure child protection and care. Studies on parental characteristics and coping styles could help inform supportive activities further.

Within the Ugandan setting we argue for more investment in community based rehabilitation to improve mobility, and care for children with spina bifida and their parents. Sensitization, especially in areas where stigmatization appears high such as the northern region, is recommended. We recommend continuous support and creation of peer support networks for parents from government and civil society. We argue for awareness raising on spina bifida to increase engagement of relatives, house helps and community members in the care of children with spina

bifida. We believe this will help reduce parental stress and improve the inclusion of both children with spina bifida and their parents.

Conclusion

Parents of children with spina bifida experience high levels of stress. The degree of such stress is partly influenced by the level of impairment and need for parental involvement in care, support received from other adults, and area of residence of the family. To reduce the parental stress, rehabilitation services should focus on improving mobility, advocacy to reduce stigmatization and strengthening in communities and developing peer support networks.

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Table 1. Demographics of the study population (N=134)

Demographic variable	Number	Percentage
<u>Caregiver relationship</u>		
Mother	104	77.6%
Father	15	11.2%
Grandmother	9	6.7%
Other	6	4.5%
<u>Education level caregiver</u>		
None	6	4.7%
Primary	72	56.2%
Secondary	28	21.9%
Vocational	11	8.6%

University	11	8.6%
<u>Marital status caregiver</u>		
Single	12	8.8%
Married	100	75.2%
Separated	9	8.0%
Widowed	11	8.0%
<u>Occupation parent</u>		
Finance / administration	5	3.8%
Small scale private business	30	22.6%
Teacher / education	9	6.8%
Medical / paramedical	5	3.8%
Civil service / government	3	2.3%
Peasant farmer	65	48.9%
No occupation	16	12.0%
<u>Child is schooling in</u>		
Nursery school	49	37.4%
Primary school	22	16.8%
Secondary school	4	3.1%
Not schooling	56	42.7%
<u>Type of disability of the child</u>		
Spina bifida	73	54.5%
Spina bifida and hydrocephalus	61	45.5%
<u>Location / region</u>		
Central	63	47.0%
East	23	17.2%
West	29	21.6%
North	19	14.2%
<u>Religion</u>		
Christian	101	77.3%

Muslim	26	19.4%
Other	3	2.3%

Monthly household income

< 30 US dollar	25	19.5%
30 - 60 US dollar	28	21.9%
61 - 90 US dollar	26	20.3%
> 90 US dollar	49	38.3%

Table 2. PSI/SF subscale and total scores for 131 parents of children with spina bifida

PSI/SF domain	Range	Mean (SD)	High score (>90% percentile) n (%)
Parental Distress Score	14-52	28.4 (10.0)	19 (14.5%)
Parent Child Dysfunctional Interaction Score	14-55	30.1 (8.7)	63 (48.1%)
Difficult Child Score	14-50	31.6 (8.3)	43 (32.8%)
Total Score	50-153	90.2 (23.9)	69 (52.7%)

Table 3. Spearman rho correlations PSI SF scales (N=134)

	PSI	PSI PD	PSI PCDI	PSICD	Gender	Child is schooling	Location	Household income	Relationship parent child	Education parent	Daily functioning VABS	Hydrocephalus	Able to walk	Assistive Devices	Rehabilitation received	Continent urine	Uses CIC	Continent stool	Uses bowel management	Parent has support of an adult	Parent is a member of a PSG
PSI total score	1	.758**	.748**	.752**	-0.049	0.033	.203*	-0.105	-0.004	-0.14	-0.136	.171*	-.254**	0.091	-.211*	0.103	-.181*	0.026	-.213*	-0.119	0.032
PSI parental distress	.758**	1	.713**	.653**	-0.029	0.101	.298**	-.178*	-0.022	-0.152	0.055	0.077	-.180*	0.202	-0.154	0.063	-0.095	0.021	-0.167	-0.169	0.065
PSI Parent Child Dysfunctional Interaction	.748**	.713**	1	.715**	-0.005	-0.006	.313**	-0.142	-0.037	-.182*	-0.102	0.145	-.200*	0.041	-0.079	.182*	-0.168	0.146	-.185*	-.234**	0.056
PSI Difficult Child	.752**	.653**	.715**	1	-0.092	-0.013	.220*	-0.162	0.022	-0.158	-0.022	0.13	-.231**	-0.008	-.173*	0.114	-.202*	0.069	-.204*	-0.096	0.003
Gender	-0.049	-0.029	-0.005	-0.092	1	-0.091	-0.081	-0.05	.186*	-0.03	0.083	0.043	0.126	0.126	0.002	-0.001	0.146	-0.01	0.125	0.069	-0.041
Child is schooling	0.033	0.101	-0.006	-0.013	-0.091	1	-.231**	.199*	0.006	.198*	.400**	-0.055	.253**	.368**	0.158	.220*	.331**	0.172	.239*	0.106	0.122
Location	.203*	.298**	.313**	.220*	-0.081	-.231**	1	-.269**	-0.144	-.340**	-0.031	0.108	-0.171	-0.08	-0.129	-0.026	-0.171	-0.02	-0.181	-0.114	.277**
Household income	-0.105	-.178*	-0.142	-0.162	-0.05	.199*	-.269**	1	0.004	.373**	-0.002	-0.051	-0.044	.499**	.229**	0.014	.218*	-0.071	0.172	.263**	-0.027
Relationship parent child	-0.004	-0.022	-0.037	0.022	.186*	0.006	-0.144	0.004	1	-0.119	0.024	0.024	-0.008	-0.026	.188*	-0.148	.183*	-0.153	0.16	-0.006	-.185*
Education parent	-0.14	-0.152	-.182*	-0.158	-0.03	.198*	-.340**	.373**	-0.119	1	0.079	-0.027	0.013	0.061	.233**	0.159	0.181	.186*	0.161	0.039	-0.008
Marital status	-0.036	-0.076	-0.114	0.028	-0.078	0.166	-0.112	.285**	0.169	0.129	0.015	0.028	-0.094	0.223	0.105	-0.024	0.132	0.015	0.141	0.092	0.001
Daily functioning	-0.136	0.055	-0.102	-0.022	0.083	.400**	-0.031	-0.002	0.024	0.079	1	-0.052	.507**	0.265	.219*	.184*	.243**	0.167	0.165	0.078	0.133
Hydrocephalus	.171*	0.077	0.145	0.13	0.043	-0.055	0.108	-0.051	0.024	-0.027	-0.052	1	-.255**	-0.066	-0.083	0.051	-0.095	-0.014	-0.099	-0.066	0.128
Able to walk	-.254**	-.180*	-.200*	-.231**	0.126	.253**	-0.171	-0.044	-0.008	0.013	.507**	-.255**	1	.	0.172	.185*	.233*	.224*	0.151	0.015	0.069
Assistive Devices	0.091	0.202	0.041	-0.008	0.126	.368**	-0.08	.499**	-0.026	0.061	0.265	-0.066	.	1	.339*	0.113	.455**	-0.036	.434**	0.224	0.245
Rehabilitation received	-.211*	-0.154	-0.079	-.173*	0.002	0.158	-0.129	.229**	.188*	.233**	.219*	-0.083	0.172	.339*	1	0.012	.505**	0.03	.475**	0.044	0.124
Continent urine	0.103	0.063	.182*	0.114	-0.001	.220*	-0.026	0.014	-0.148	0.159	.184*	0.051	.185*	0.113	0.012	1	-0.006	.762**	-0.131	0.041	0.137

Uses CIC	-.181*	-0.095	-0.168	-.202*	0.146	.331**	-0.171	.218*	.183*	0.181	.243**	-0.095	.233*	.455**	.505**	-0.006	1	0.043	.896**	0.113	.213*
Continent stool	0.026	0.021	0.146	0.069	-0.01	0.172	-0.02	-0.071	-0.153	.186*	0.167	-0.014	.224*	-0.036	0.03	.762**	0.043	1	-0.092	-0.026	0.116
Uses bowel management	-.213*	-0.167	-.185*	-.204*	0.125	.239*	-0.181	0.172	0.16	0.161	0.165	-0.099	0.151	.434**	.475**	-0.131	.896**	-0.092	1	0.163	.207*
Parent has support	-0.119	-0.169	-.234**	-0.096	0.069	0.106	-0.114	.263**	-0.006	0.039	0.078	-0.066	0.015	0.224	0.044	0.041	0.113	-0.026	0.163	1	0.023
Parent is PSG member	0.032	0.065	0.056	0.003	-0.041	0.122	.277**	-0.027	-.185*	-0.008	0.133	0.128	0.069	0.245	0.124	0.137	.213*	0.116	.207*	0.023	1

** . Correlation is significant at the 0.01 level (2-tailed). * . Correlation is significant at the 0.05 level (2-tailed).

Table 4. Linear regression model of PSI/SF total and subscale scores, with demographic, impairment, and support related predictors presenting (only significant predictors for each dependant variable)

Dependent variable	Significant predictors	Beta	R ²	F
Parental distress	Ability to walk <i>a</i>	-.190*	.100	2.254*
	Bowel management <i>b</i>	-.468*		
Parent Child Dysfunctional Interaction	Support of another adult <i>c</i>	-.228*	.184	7.793**
	Location <i>d</i>	.284**		
Difficult child	Ability to walk	-	.154	7.793*
	Location	.218**		
		.260**		
Total Parental Stress	Ability to walk	-.248*	.109	3.114*

Linear regression analysis codes: a child is able to walk unaided is 1, unable to walk unaided is 0; b Practising bowel management is 1, not practising is 0; c. Having support is 1, not having support is 0; Location north is 1, other is 0.

** <0.01; * <0.05

Table 5. Children's mobility and physical impairments

Impairment variable	Yes	No
Child can sit without assistance	131 (97.8%)	3 (2.2%)
Child is able to speak	127 (94.8%)	7 (5.2%)
Child is able to walk without assistive devices	62 (49.2%)	64 (50.8%)
Child is continent of urine	15 (11.4%)	117 (88.6%)
Child uses clean intermittent catheterization	99 (81.1%)*	23 (18.9%)
Child is continent of stool	18 (13.4%)	114 (86.4%)
Child uses bowel management	91 (78.4%)**	25 (21.6%)

* 23 (35.9%) uses a wheelchair, 23 (35.9%) uses crutches, 17 (28.1%) crawls

* 22 (16.4%) practices CIC without assistance of another person

** 7 (6.9%) practices bowel management without assistance of another person

Table 6. Reasons for (not) participating in parents support groups for parents of children with spina bifida and hydrocephalus

Benefits of participating in Parents Support Group (N=56)	Number	Frequency
Learning about taking care of child with SB	28	50.9%
Encouragement from other parents	8	14.5%
Sharing experiences, learning from each other	8	14.5%
Learning how to include my child in school	1	1.8%
Income generating activities / opportunities	7	12.7%
Not sure, just joined	2	3.6%
Reason unknown	2	3.6%
Reason for not participating in Parents Support Group (N=78)		
Not aware of existence of PSG	43	55.1%
Not in their location / far away	23	29.5%
Is planning to join, was not aware	5	6.4%
Does not have time	3	3.9%
Reason unknown	4	5.1%

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5.2 Family relationships, dependency and care: perspectives of children with spina bifida in Uganda⁸

Abstract

In Uganda public health and education services are poorly equipped to respond to needs of children with spina bifida. Such children are highly dependent on their families for care and support. In this study we explored the children's and their siblings' perceptions of family relationships, dependency and care to inform support for the children and their families.

Semi-structured interviews were held with 30 children with spina bifida, and 30 of their siblings in Uganda. Based on the Family Relations Test, we created a set of local dolls and culturally appropriate messages to evaluate the children's perspectives about their family members and themselves.

Our findings confirm a highly dependent family care model in which mothers play a key role in the children's lives, dependency and care; fathers are relatively absent for both siblings and children with spina bifida; siblings, and other household members play an important role in the care and inclusion of children with spina bifida; and children with spina bifida have more negative self-perceptions compared to their siblings.

The internal family networks of children with spina bifida in Uganda are wide and varied, and characterized by higher dependency on mothers and elder siblings and househelps. We argue for an extended family centered approach in health care and social services, in which children with spina bifida, their parents (including their fathers), siblings, house-helps and other family members are included. Through family support, children and adult family members can be strengthened and enabled to demand for inclusive care and support services on community and national level.

⁸ Based on: Bannink, F., Idro, R., and van Hove, G. (in press) Family relationships, dependency and care: perspectives of children with spina bifida in central Uganda. *Knowledge Management for Development Journal, Special Issue Knowledge for Disability Inclusive Development*.

Introduction

Spina bifida is a neural tube defect, a congenital disability, whereby the spinal cord and vertebrae do not form completely and the neural tube fails to develop normally. In Uganda incidence data is not available. Warf et al (2011) estimated a birth incidence of 1 in 1,000, translating into 1,400 children born with spina bifida in the country annually (Warf, Wright, & Kulkarni, 2011). Most children with spina bifida have some degree of paralysis, which affects mobility as well as bowel and bladder control (Northrup & Volcik, 2000). Hydrocephalus is another common complication. Sixty-six per cent of children with spina bifida in low-income countries develop hydrocephalus (Warf et al., 2009). As a result, children with spina bifida and their families must manage medical and rehabilitative care including catheterization, bowel management programs, assistive devices, and skin checks to avoid pressure sores (Lindsay, 2014). Aside from these physical needs, research has shown that children with spina bifida are prone to neurocognitive deficits in executive function (involving organization, impulse control, working memory, and flexible thinking), attention and learning difficulties (Dennis & Barnes, 2010). Children with spina bifida often face social skills and inclusion challenges (van't Veer et al., 2008) and often rely on their family members for social interaction (Holmbeck & Devine, 2010).

Research on functioning and psychosocial adjustment of families of children with spina bifida in high income countries support a resilience–disruption view, whereby the presence of a child with spina bifida disrupts normative family functioning at first, but after a period of time, families adapt and exhibit considerable resilience (Holmbeck et al., 1997; Vermaes, Janssens, & Gerris, 2007). Nevertheless, spina bifida has negative effects on parents' psychological adjustment and functioning (Holmbeck & Devine, 2010; Vermaes, Janssens, Mullaart, Vinck, & Gerris, 2008). Stress levels of parents of children with spina bifida are higher than parents of typically developing children (Holmbeck et al., 1997; Kanaheswari, Razak, Chandran, & Ong, 2011). Caregivers of children with disabilities often felt that they do not have sufficient time to cope with household tasks, and feel isolated (Gona, Mung'ala-Odera, Newton, & Hartley, 2011; Hartley, Ojwang, Baguwema, Ddamulira, & Chavuta, 2004).

A study on adjustment and sibling relations in families with a child with spina bifida in the US (Bellin & Rice, 2009), found children with spina bifida were more likely to feel satisfied with family functioning when they experienced warmth and limited conflict in their relationship with their sibling(s).

In Uganda and other East and Southern African countries formal public rehabilitative and social services are limited for families of children with spina bifida (Mertens & Bannink, 2012). Families in Uganda do not receive government support, and public health and education services are poorly equipped to respond to needs of children with spina bifida (Bannink, Idro, & Van Hove, 2016b). The children's families are often their main source of care and protection (Miles, 2006). A few civil

society organizations aim to provide basic surgery and rehabilitation services at community level for children with spina bifida and involve parents as the key stakeholders (Mertens and Bannink, 2012).

Families of children with neuro-disabilities including spina bifida in Uganda (Bannink, Stroeken, Idro, & Van Hove, 2015), Kenya (van't Veer et al., 2008), Malawi (Paget, Mallewa, Chinguo, Mahebere-Chirambo, & Gladstone, 2015) and South Africa (Coomer, 2013) struggle with the financial implications and social barriers towards care and support for their child. Parents in Uganda have described how they were initially discouraged from taking care of their child by relatives, neighbours, and health workers (Bannink et al., 2015). Although attitudes changed over time, inaccessibility of services and negative perceptions held by the community at large continued to affect the care and inclusion of the children (Bannink et al., 2015). High parental stress was found in mothers and grandmothers looking after children with disabilities in Uganda and Kenya (Gona et al., 2011; Hartley et al., 2004). In Uganda, parental stress is higher in parents of children with spina bifida with limited mobility, those who have no support of another adult in the provision of care, and who require daily continence management exercises (Bannink, Idro, et al., 2016b). Social inclusion of children with spina bifida is limited to micro level and is affected by poverty and stigmatization; children do have a sense of belonging at home in which participation in daily household activities plays an important role (Bannink, Idro, & Van Hove, 2016a). Inclusion in wider society only takes place over time. Participation in social life outside the home setting is often enabled by siblings who accompany the child with spina bifida to play with other children in their communities, or help them to get to school (Bannink, Idro, et al., 2016a).

To further explore family relationships, dependency, and care in families of children with spina bifida in Uganda, we interviewed children with spina bifida and their siblings, using the Family Relations Test (FRT). The FRT is a psychological test which evaluates a child's perspective about their family relationships and themselves, and measures outgoing and incoming negative and positive feelings for each family member, as well as feelings of dependence (Bene & Anthony, 1985). Through the use of the FRT in interviews with children with spina bifida and their siblings in Uganda, we explore both qualitatively and quantitatively how the children perceive their relationships towards their family members. We included both children with spina bifida and their siblings in the study to explore family relationships of, and interdependency between, both groups of children, with siblings functioning both as a control group for understanding the children with spina bifida and as an independent source of valuable insights about their experience, as recognised enablers of inclusion (Bannink et al, 2016b). The study aims to provide recommendations for civil society and government programs on support to families with children with spina bifida.

Method

Sample

Participants in this study were 30 children with spina bifida and 30 siblings aged 4 to 14 years from central Uganda. The sample was taken from a larger social inclusion study sample of 139 families of children with spina bifida in Uganda. The following inclusion criteria were used to select the sample: i) residing in the central region (51 children out of the total 139); ii) cognitive ability to complete the FRT (38 out of the 51 children), based on earlier cognitive test findings (Bannink, Fontaine, Idro, & Van Hove, 2016); and iii) has a sibling who is willing and available to participate (30 out of 38). Elder and younger siblings closest in age to the child with spina bifida in the household were purposefully selected during home visits. The sibling closest in age was selected to enable comparisons between perceived family relationships without a large age bias (e.g. perceptions of relationships between teenagers and parents differ from those of younger children, and older children may attribute more independence cards to themselves compared to younger children). In total 17 boys and 13 girls with spina bifida, 14 brothers (7 elder, 7 younger), and 16 sisters (9 elder and 7 younger) participated. The average age of the children with spina bifida was 9.20 (SD=2.37) years, and for siblings 9.13 (SD=1.98) years. The average household size was 7 (ranging from 3 to 11). The majority (22/30) of the children's parents were married and living together. Data was collected at routine visits of children at a clinic providing neuro-surgical follow up and rehabilitative care in Kampala and at home. All children in this study had received neurosurgery and physiotherapy, and were practising continence management. Both the children with spina bifida and their siblings were in school. The average household income was US\$87 per month.

Data collection

Semi-structured interviews were held with 30 children with spina bifida, and 30 siblings about family relationships, dependency, care and inclusion using the Family Relations Test.

The Family Relations Test (FRT) was developed by Eva Bene and James Anthony in 1957, and revised in 1978 and 1985 (Bene & Anthony, 1985). Translations have been made into various languages, and adaptations made to allow for more quantitative analysis and comparison of the findings (Celestin-Westreich, Baarda, & Ponjaert-Kristoffersen, 1999). The FRT evaluates a child's perspective about their family relationships and themselves, and measures outgoing and incoming negative and positive feelings for each family member, as well as feelings of dependence (Bene & Anthony, 1985). The tests are designed for use with children aged 3 to 7 years and 8 to 15 years respectively. Two versions are available for these age groups (Bene & Anthony, 1985).

From 20 cardboard figures, the child selects one to represent each member of the family, including him or herself. The cardboard figures have folded mailboxes. The child reads (or is read) the statement on each card, and is asked to drop the card bearing a single emotional statement or attitude to one of the figures. The inward positive cards show which person supports the child; the

outward positive cards reflect who the child goes to for support, care, and play. The inward negative cards describe who gives negative feedback and disciplines the child, whilst the outward negative cards describe who the child does not like, is competitive with, and fights with. The dependency cards measure the child's independence in terms of self care. If the statement is not applicable to any of the represented family members, the card is given to the figure "Nobody". After all of the cards have been distributed, scores are tallied on the appropriate form. The FRT does not have standard scores or norm groups and test reliability cannot be calculated. We explored the use of the test to enable us to better understand children's family perceptions.

The FRT test as published by Celestin-Westreich et al (1999) was analyzed, adapted, translated, and administered by the authors - psychologists, educational scientists, and social workers with over 25 years combined working experience with children with spina bifida in Uganda. In this version, a child can only give the card to a single family member, which makes it possible to quantify the number and percentage of cards each family member received and compare results. It has been used in therapy with traumatized children from multi-cultural settings in Belgium and The Netherlands. Advice on adapting this version of the FRT for use in Uganda was requested and received from Prof. Dr. S. Celestin-Westreich.

Subsequently, the text on the cards was translated into Luganda, the local language spoken in the central region. The text on the card 'who would you like to give a kiss' (a positive outgoing message card) was changed into 'who would you like to give a hug' as kissing is not a culturally common form of expressing affection or greetings in Uganda. While in the original version the cards are entered in small mailboxes, the cards in the Ugandan setting were placed next to a doll that represented the relevant family member, since most children are not familiar with a postbox system, but rather with an in person delivery of messages, a 'messenger'. The Caucasian looking cardboard figures were replaced by locally made dolls, dressed in African clothes. In total 20 dolls were made, representing persons of both sexes and different ages (including grandparents and babies).

Image 1. Photo of the dolls



The test was initially piloted with 12 school going children (6 with and 6 without spina bifida) age 4 to 13. All children easily associated the dolls with their family members and enjoyed participating. The average family size in the pilot was 7 (5 children, 2 adults). The cards were read to all children, as the majority could not read or not read fast enough to complete the test on their own. The statements on the cards in the version for younger children were easily understood. However, the statements on the cards in the version for older children often led to confusion and the higher number of cards often resulted in disengagement of the child at the end of the test. When the older children were given the version for the younger ones, these challenges did not occur. Administration of the version for older children took approximately 52 minutes, whilst the version for younger children was completed within an average of 28 minutes. We decided to only use the version for younger children for the children and siblings in this study.

Data analysis

Interviews were transcribed and thematic analysis was conducted using NVIVO16. The interview data included examples and stories told by the child during the FRT administration. For example, a child would assign an FRT card 'who do you like to play with' to a sibling, and narrate a story about playing a game with his brother. FRT results were recorded and entered into a database. For each child, a summary was made of the total number of positive and negative incoming and outgoing feelings, and dependency cards as per test categories and instructions. Positive and negative incoming and outgoing feeling scores were totaled into one positive and one negative score. Summaries and averages were grouped for older and younger siblings. Averages and standard deviations of cards given per family member were calculated based on family size and selected family figures. A two-tailed T-test was utilized to test if there would be a significant difference between children with spina bifida and their sibling's perceptions of feelings related to their family members.

Ethics

Ethical approval and research clearance were obtained from Ghent University, Belgium, the Uganda Virus Research Institute, and the Uganda National Council for Science and Technology. Informed consent was obtained from all parents, and assent from children and siblings eight (8) years or older where possible. Individual results of the FRT and interviews with the children were discussed and shared with the social workers involved in the children's follow up program. The use of dolls as a method to help children talk, describe, and work through difficult situations was discussed with the social workers.

Results

Family composition

Children were requested to describe their families. Most of them named their family members and explained where they live.

'My mum, and my brother, and my sister, and my baby sister, and my father, and my auntie'
Boy with spina bifida (6)

'I have two brothers and three sisters. And my mum and my dad. We live in Nsambya' Girl
with spina bifida (11)

When asked to select a doll for each family member, twenty eight (28) children selected a mother figure. Two (2) children were looked after by a grandmother and had no mother present. Another two (2) children added a grandmother as well as a mother in their family composition. Twenty two (22) children selected a father figure. All children selected at least one sibling; the majority selected 4 or more siblings.

FRT results were entered for each individual family member. Table 1 shows a summary of the findings with the average numbers of FRT cards given to each family member for parents, siblings, and other family members. Sibling results were grouped into younger, older, and total siblings. Findings for other relatives and house helps were summarized in one group.

The family set up children made often differed from the family set up given by the parent. In total 6 (six) children left out fathers who worked upcountry, and twelve (12) children left out siblings who were in boarding school, and were only around during weekends and/or holidays. Twenty six (26) children with spina bifida included 'other' relatives and househelps in their family selection; siblings did not include any.

Table 1. Average number of cards and two tailed T-test results for children with spina bifida and their siblings' family perceptions (N= 30 children with spina bifida and N=30 siblings from the central region)

Person receiving card	Type of card	Average number of cards given (SD)		Levene's Test	t-test
		by CwSB (n=30)	by Siblings (n=30)	F-value	T
Self	Positive	0.52 (1.52)	0.07 (0.25)	7.989**	1.535
	Negative	0.59 (0.89)	0.13 (0.25)	21.080***	2.467**
	Dependence	1.14 (1.36)	2.63 (0.34)	0.006	-3.755***
Father	Positive	1.71 (1.61)	1.20 (1.38)	0.611	1.200
	Negative	1.19 (1.50)	1.33 (1.58)	0.013	-0.318
	Dependence	0.65 (0.79)	0.59 (0.94)	0.296	-0.044
Mother	Positive	4.38 (2.59)	4.40 (2.65)	0.081	0.000
	Negative	1.52 (1.75)	0.67 (0.91)	13.534***	2.300*
	Dependence	3.11 (1.78)	3.86 (1.75)	0.404	-1.599
Grandmother	Positive	0.24 (1.28)	0.67 (1.72)	4.804*	-1.096
	Negative	0.17 (0.91)	0.07 (0.36)	1.332	0.557
	Dependence	1.33 (1.25)	1.50 (0.87)	0.653	-0.176
Older sibling	Positive	3.76 (3.68)	7.90 (2.72)	4.017*	-4.538***
	Negative	5.00 (4.41)	8.07 (3.14)	5.670*	-3.116**
	Dependence	0.93 (1.96)	1.03 (1.05)	7.417**	-0.326
Younger sibling	Positive	3.00 (3.35)	0.53 (1.56)	19.311***	3.455***
	Negative	2.10 (4.04)	0.53 (1.87)	7.261***	1.833
	Dependence	0.72 (1.39)	0.10 (0.40)	17.582***	2.266*
Sibling (total)	Positive	6.76 (3.29)	8.43 (2.67)	1.152	-1.994*
	Negative	7.10 (4.24)	8.60 (3.37)	1.451	-1.599
	Dependence	1.66 (2.06)	1.13 (1.09)	12.109***	1.086
Other	Positive	0.76 (1.72)			
	Negative	0.66 (1.49)			
	Dependence	0.34 (0.88)			
Mr Nobody	Positive	2.10 (2.31)	1.37 (1.08)	2.753	1.495
	Negative	5.10 (3.94)	5.30 (2.61)	6.573**	0.000
	Dependence	0.59 (0.89)	0.27 (0.63)	19.127***	3.837***

* p < .05, ** p < .01, *** p < .001

Family relationships

Children with spina bifida were asked to tell the interviewer about their family members. The children gave examples of situations that had occurred with family members, and described different roles and relationships in their narratives. Most of these were related to care, family disputes or restrictions applied to children's time to play outside or expectations towards school work.

'My parents look after us. My dad pays school fees. My mum cooks and she takes me to the clinic. I play with my brothers and my sister'. Sister (7) to a girl with spina bifida (10)

'My brother always gets into trouble. He never does his homework. My dad gets very angry and beats him when he comes home. He does not beat me, my mum told him it is not good for my head' Girl with spina bifida and hydrocephalus (12) about her elder brother (14)

'Jajja [grandmother] lives with us, she is very old, she likes to tell stories' Sibling (7) of a girl with spina bifida (5)

'On Sundays we all go to church together' Girl with spina bifida (5) about her family

The positive cards were generally distributed amongst three (3) or four (4) family members. The inward positive cards were mostly given to mothers, and elder siblings. Some were given to other family members by children with spina bifida. The outward positive cards were mostly given to mothers, younger siblings for children with spina bifida, and older siblings for the siblings interviewed. No significant differences in positive perceptions about parents were found between children with spina bifida and siblings. Children with spina bifida gave less positive ($t=-4.538$, $p<0.001$) cards to their elder siblings compared to their siblings, but more positive cards to their younger siblings than their siblings ($t=3.455$, $p<0.001$). In discussions about the positive messages children gave more details about what their family would do to make them feel loved, or who they enjoyed to play with. Most of them spoke about their mothers and siblings, only a few spoke about their fathers.

'I like to play football. My brother watches. I push his wheelchair to the field'. Older brother (14) about his younger brother with spina bifida (12)

'I like to play with my sister, she likes to play meso [a board game] with me'. Boy with spina bifida (10) about his younger sister (8)

'My mum carries me and she tells me stories'. Girl with spina bifida (6)

'I help my little brother with his homework' Boy with spina bifida (11)

'My little brother comes and sits on my lap and gives me hugs'. Girl with spina bifida (6)

'My brother helps me when I am scared. He takes me with him and protects me'. Elder sister (11) of a girl with spina bifida (6) about her brother (15)

'My sister pushes me to school' Girl (8) with spina bifida using a wheelchair

'My aunt [househelp] gives me food' Boy with spina bifida (4)

Negative inward and outward cards were mostly given to elder siblings. Children with spina bifida gave significantly less negative ($t=-3.116$, $p<0.001$) cards to their elder siblings compared to their siblings. Children with spina bifida perceived more negative interactions with their mothers than their siblings ($t=2.30$ $p<.05$), e.g. 'this person does not like me'. Similar perceptions were found by the children living with their grandparents. In discussions with the children, they would describe what they did not like in the relationship to the family member they gave their card to:

'My mum does not let me play with the other children'. Boy with spina bifida (6)

'Mum shouts at me' Girl with spina bifida (4)

'My sister beats me when I don't listen to her' Older sister (8) of a boy with spina bifida (6)

Some siblings expressed negative feelings towards their sibling with spina bifida:

'He needs to be helped all the time. My mum has no time to help me with my homework because she is always busy with him' Elder sibling (14) of a boy with spina bifida (9)

No differences were seen in perceived family relations with fathers between children with spina bifida and their siblings. When asked about their fathers, most children described their profession. The most common positive outward going card children gave to the father figure in the FRT was 'who repairs your toys when they are broken'.

Dependency and care

Children with spina bifida assigned themselves more negative cards than their siblings did ($t=2.467$, $p<0.01$). These cards were related to not liking themselves, and being stubborn. When asked for clarification most of the children referred to their difficulty in daily functioning:

'I just have to sit around and wait for her [mum] to finish her work, then I can get out of the house'. Girl with spina bifida, 7 years old

'I don't want to do CIC [clean intermittent catheterization used for continence management], my mum says I have to learn to do it myself but it takes long and I just want to play. She says I am stubborn' Boy with spina bifida, 10 years old

These statements relate to the lower number of independence cards in terms of care (e.g. 'who helps you to dress') children with spina bifida gave themselves compared to their siblings ($t=-3.775$, $p<.001$). The highest average of dependency cards of children with spina bifida were given to mothers. Children with spina bifida gave more dependency cards to their younger siblings compared to their siblings too ($t=2.266$, $p<0.05$). As some of the children with spina bifida and their siblings explained:

'My mum carries me to school; and comes to school to do CIC' Boy with spina bifida 8 years old

'I can't dress myself, I can't move my legs. I can put on a shirt, but the trousers I can't. My brother has to help me.' Boy with spina bifida 11 years old

'I help my sister to fill her basin with water. My mum then puts her down in the bathroom and then she baths herself'. Younger sister of a girl with spina bifida 7 years old.

Children with spina bifida attributed more dependence ($t=3.837$, $p<.001$) cards to Mr Nobody than their siblings. The most common card which was given to Mr Nobody was 'who likes to lift you?' Children with spina bifida explained the dependency in mobility:

'Sometimes I want to get out of my wheelchair and crawl around. If I have pressure sores my mum does not allow me. Then I get bored because I am too heavy to carry and I can't move my wheelchair on my own, so then I am stuck where she parked me'. Girl with spina bifida 13 years old

Discussion

Family perceptions of the children using the Family Relations Test enabled us to explore their views on family relationships, dependency, and care. The adjustments we made to the tests helped us to successfully engage the children and administer the test in a culturally appropriate and understandable manner.

We identified the following key elements in family relationships, dependency and care:

- Mothers play a key role in the children's lives, dependency and care
- Fathers are relatively absent for both siblings and children with spina bifida
- Elder and younger siblings play an important role in the care and inclusion of children with spina bifida
- Importance of other relatives in the care for children with spina bifida
- Children with spina bifida have more negative self-perceptions compared to their siblings

Our study was limited by a small non randomized but purposely selected sample size, and the small number of siblings we could involve. Family relationships are varied and fluid. Our findings captured perceptions of children at a certain point in time. Larger studies with assessments at different times and in-depth sibling-sibling analysis are recommended to understand the way relationships evolve over time. We did not find consistent differences in family relationships between younger and older children with spina bifida. However the need for and understanding of care and dependency did change over time. Older children gave more feedback on the impact of care on family relationships, e.g. their mother spending a lot of time in ensuring they would practice their continence management, and the disagreements about their freedom and possibilities to interact with peers outside their homes.

Mothers were given the highest average of positive feeling and dependency cards of all relatives. Children with spina bifida in our study perceived more negative interactions with their mothers than their siblings, but equal positive and dependency ones. Most likely the increased amount of time spent with their mothers due to their care needs, increases negative interaction and frustration in the child-parent relationship compared to the sibling-parent relationship, as siblings may have less care needs and less time interacting with their parent. In high income countries, spina bifida affected the parent-child relationship more negatively for mothers than fathers, possibly due to the mothers continuous exposure to spina bifida related demands (Vermaes et al., 2007). In Malaysia dysfunctional parent-child interaction was higher if mothers were the sole caregiver (Kanaheeswari et al., 2011). Despite the burden and higher negative interactions, overall relationships with mothers are positive and children feel close to them. These findings are in line with studies from other countries, e.g. in Canada where Antle et al. (2009) found that mothers were an important source of support for children with spina bifida followed by fathers, siblings, and other family members.

The low average number of cards given to fathers (positive and negative) by both children with spina bifida and their siblings in our study, could indicate children perceive their fathers to be less present in their lives. Traditionally the role of a father was more one of authority and the provider of a family in sub-Saharan Africa (Shwalb, Shwalb, & Lamb, 2013). Although society is changing, fathers may still be less involved in direct physical care of a child or being emotionally close: typically considered the mothers' role (Lamb, 2004; Shwalb et al., 2013).

In our study we found grandmothers and elder siblings played important roles in the children's lives. Traditionally children in Uganda live with their extended families (Rutakumwa, Zalwango, Richards, & Seeley, 2015). Although family sizes have reduced, especially in urban areas, the presence of a grandparent, aunt, uncle or cousins in addition to the nuclear family is still common. Families in our study had an average of seven household members. In the families where mothers were absent, other relatives would take up the care for the child with spina bifida, e.g. grandmothers and elder siblings. Grandmothers have played a key role in child care in Uganda over the past decades especially for children orphaned by AIDS (Rutakumwa et al., 2015).

In all families, elder siblings had a general responsibility for looking after younger siblings. We found that children with spina bifida perceived less positive and less negative relationships with their elder siblings compared to their siblings. Children with spina bifida perceived more positive relationships and dependency with their younger siblings, than their younger siblings did. These findings could indicate less interaction between children with spina bifida and their elder siblings, compared to the interaction between their siblings. Earlier we found siblings noted their sibling with spina bifida was treated differently in school and the community (Bannink, Idro, et al., 2016a). Elder siblings in our study did make an effort to include their sibling with spina bifida in daily and social activities. They often played an important role in getting the child to and from school. They may have had less negative interactions with their sibling with spina bifida compared to their other siblings, as they have a care responsibility and may protect their sibling.

Elder siblings are more likely to have their own social interactions outside the home, and are often expected to help in household chores such as fetching water and going to the shop compared to younger siblings. Relationships with younger siblings were referred to in terms of play and hugs. As children with spina bifida are less mobile than their peers, they tend to spend more time at home, and interact with younger siblings more than their elder siblings do.

Our findings indicate the importance of sibling relationships and the important role siblings play in day to day care and inclusion of children with spina bifida. In care and support programs, the current focus is on parents, and could be expanded by including siblings to build positive interactions and support both children with spina bifida and their siblings. This is in line with earlier findings of studies conducted in USA in which inclusion of siblings in support programs has been argued for (Bellin & Rice, 2009) (Lindsay, 2014).

Most children with spina bifida included 'other' relatives and househelps in their family selection; siblings did not do this. Having a househelp or 'maid' is a common practice in Uganda's central where this study was conducted (UBOS, 2014). Househelps are often relatives of the parents, and are paid a very small amount of money. In our previous study of parental stress in parents of children with spina bifida in Uganda, parents pointed out that they often felt alone in taking care of their child and found it hard to meet all the care demands. This was moderated by receiving support from other adults (Bannink, Idro, et al., 2016b). In this study children with spina bifida acknowledged the presence and role of other adults in their home: they are involved in the care for children with spina bifida on a daily basis. They may be less present in the lives of the siblings as they have different care needs, and therefore not be perceived as important persons to display in family set up for the FRT by siblings.

In comparing family perceptions of children with spina bifida and their siblings, we found children with spina bifida had a more negative perception of themselves than their siblings, and showed less independence in terms of self care. This is in line with studies from high income countries, which found lower self esteem and high dependence in children with spina bifida (Bellin & Rice, 2009;

Rofail et al, 2013). The challenges with independence are mostly caused by mobility challenges. The self perception of children with spina bifida was negatively affected by not being able to participate in household tasks. In an earlier study parents explained children were sometimes left out from larger family gatherings due to mobility problems (Bannink, Idro, et al., 2016a). In this study mobility and physical care such as continence management were mentioned too.

Children with spina bifida did attribute more dependence cards to Mr Nobody than their siblings, which raises a concern about the care they receive as they may not be receiving the help they need completing daily tasks such as dressing and bathing. Warf, Wright et al earlier described finding situations of child neglect during home visits in the eastern region (Warf et al., 2011). It may refer to anxiety around accessing the help they need in a broader way.

Conclusion

Family relationships, dependency and care of children with spina bifida in Uganda needs to be understood within the cultural context. Our study shows the internal family networks of children with spina bifida in Uganda are wide and varied and family relationships are characterized by higher dependency on mothers and elder siblings and househelps.

The culturally adapted FRT was a helpful measure to assess family relations, dependency and care from the children's perspective. In some cases, the dolls produced were further utilized in counseling sessions, and used to act out stressful situations and integrate possible coping strategies. The combined use of the dolls as an assessment and therapy tool, works well in low resource settings such as Uganda, and should be further explored in intervention programs and studies.

To further disability inclusive development, we argue for an extended family centered approach in health care and social services, in which the children with spina bifida, their parents (including the fathers), siblings and house-helps are included.

Siblings play a key role in the day to day interaction and development of children with spina bifida; by promoting positive interaction and interdependence, children with spina bifida are more likely to be included in daily life activities, school, and communities. At the same time siblings need to be protected from taking on the sole responsibility for the care of their sibling with spina bifida.

Although mothers are the target group for most care and support programs in Uganda, they require support from other adults too, and may benefit more from programs which engage others in caring for their child with spina bifida to reduce their sole care responsibility.

Programs which empower fathers to become role models in child care and examples in their communities fighting for disability inclusive development are encouraged.

House helps need to be involved in community based rehabilitation trainings to ensure children with spina bifida are assisted in their daily care needs including continence management.

A balance of task division with interdependence is important to ensure all family members develop to their full potential and are cared for. By enabling families to form interdependent units, advocacy will follow. Families will speak up about and ask for disability inclusive development in their communities and country.

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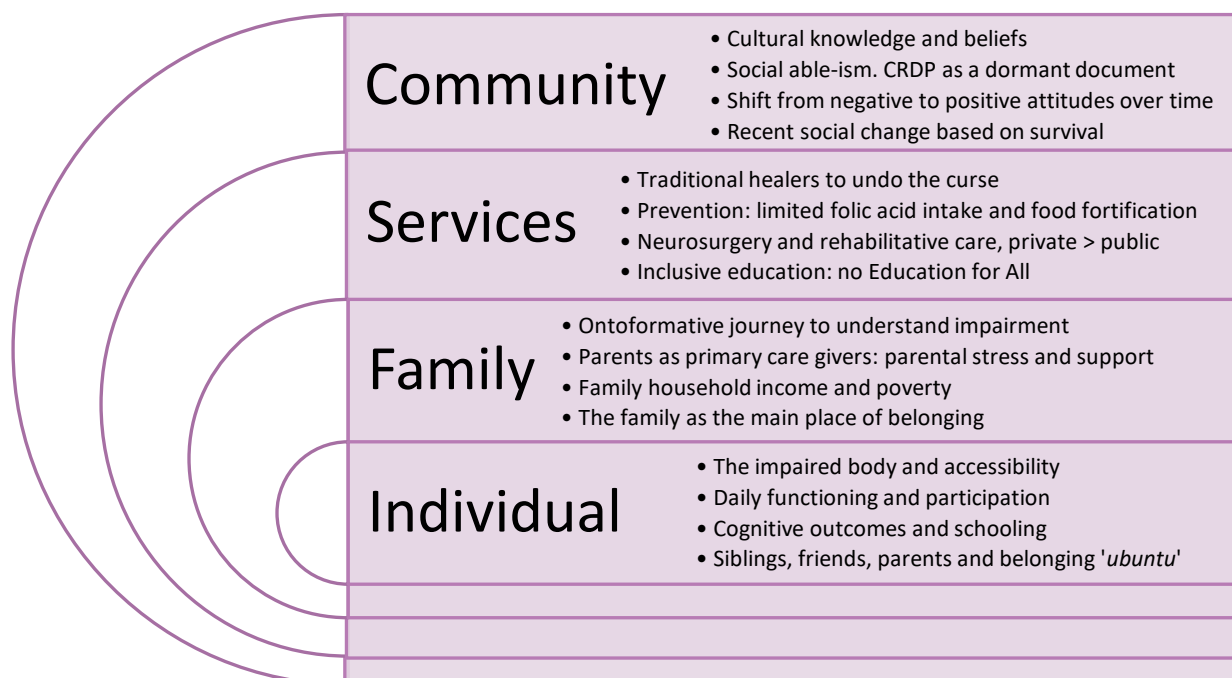
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Chapter 6 Discussion



6.1 Main findings

This study presented a situation analysis on social inclusion, care and belonging of children with spina bifida in Uganda. The framework below explains the different layers, and factors of social inclusion, following Reindal's socio-relational model (Reindal, 2008). It includes the bodily experiences and limitations experienced due to reduced functioning on one hand and poverty, cultural, and social barriers and the experience of these on the other hand.



6.1.1 Knowledge, attitudes, practices

The first research question asked about the *knowledge, attitudes, and perception of children with spina bifida in Uganda*. The descriptions of disability and spina bifida found in our study reflect cultural interpretations of the cause and refer to the physical appearance of the child. A wide variety in explanations was found and regional and individual differences were noted. Most commonly communities believed spina bifida was caused by witchcraft, bad luck, and the use of family planning methods. Whilst a large number of parents consulted traditional healers for treatment after birth who would provide explanatory models and support to undo a 'curse', most parents remained searching for a cure and accessed biomedical treatment and rehabilitative care to treat the impairment. Their search for meaning and understanding of their child's impairment was and is an ontoformative journey through indigenous, colonial, and global knowledge, reflecting dynamics of current Ugandan communities, embracing traditional, colonial, and global cultural beliefs and practices.

Prior to the existence of neurosurgery and rehabilitation, children with spina bifida in Uganda would die shortly after birth. We expect many still do as approximately 1,400 children are born with spina bifida (Warf, Wright, et al., 2011), and only 300 receive neurosurgical closure of the spine annually. The cultural explanations of the cause and prognosis reflect what used to happen: a child with a 'swelling on the back' would die. Parents were cursed, had bad luck, as their child would not survive. The community feels parents are 'wasting their time and income' investing in this 'dead' child. The community response is to 'help' the parents to get rid of the child, by 'accidentally' dropping the child in the river, or by neglecting the child and excluding the child and parent from social life so that they will eventually give up. In a few cases the children were seen as having supernatural powers. Despite being treated well out of fear, they were still outcasts. Not only children, but parents too, would be called names.

Most parents in our study were not supported in accessing medical care; many were not aware or if they were, family members felt it was a waste of resources to send the child for neurosurgery and rehabilitation. Health workers of government health facilities held similar beliefs and were often not informed about the biomedical causes and treatment options. The exclusion has a function of protection of other community members from curse or bad luck, mitigating effects of poverty, ensuring that scarce resources are used for socially agreed priorities.

Communities often assumed the mother did something wrong resulting in this misfortune. However the concept of being cursed or being given by God also placed the responsibility with an outside source, something which could take away the dilemma and responsibility from the parents (Stroeken, 2010). Mothers in our study indicated their fight against these assumptions and efforts made to show others their child is alive and should belong.

Over time community attitudes changed; after seeing the child survived surgery, and started participating in day to day family life, community members change their way to 'support' the family. The child is gradually accepted as part of the community, and social life. Whilst this was a common trend, not all parents had this experience, especially those in the northern region struggled with exclusion due to the belief spina bifida is infectious. The number of children found and average age of the children in this region was lower compared to other regions, and attitudes were largely negative. This could be attributed to a combination of cultural beliefs and lack of access to basic services during the 22 year long conflict between the Government of Uganda and the Lord's Resistance Army in this region in which 90% of the population was displaced (Muyinda and Whyte, 2011).

The social and cultural change in survival and responses to the survival is recent, and is a complex play of traditional society and cultural values and globalization and access to biomedical care. Looking at the knowledge, attitudes, and perceptions from a right based approach, attitudes and practices towards children with spina bifida and their families can be defined as child neglect, abuse, and discrimination. However individual rights are not a universal concept (Meekosha & Soldatic, 2011). By looking at the diversity of factors, including the history of tribal cultures, colonial past, poverty, and globalization, a deeper understanding and discussion of knowledge, attitudes, and practices towards social inclusion and belonging can take place.

6.1.2 Health and rehabilitative services

The second research question was set to find out *which biomedical health and rehabilitation services children with spina bifida and their parents access* as these closely relate to their survival and daily functioning. We found children had poor access to general health services, and difficulties accessing specialized health services for neurosurgery and rehabilitation. The children generally expressed excitement and happiness over attending the specialized rehabilitation services. Most children knew the rehabilitation workers by name, and felt close to them. Although the rehabilitation centers encourage families to attend general health services for unrelated conditions, the children and parents expressed dissatisfaction with these services. Some were denied care by health workers. Health workers had limited biomedical knowledge and understanding of the impairment.

In our study the majority of the children received neurosurgery to close the spine, and those with hydrocephalus received ETV, followed by VP shunting. Over 90% had received physio- and occupational therapy, and continence management training. The majority could sit unaided; about half could walk without assistive devices. Of those in need of assistive devices, almost a third did not have access to these. The majority of children mentioned mobility and practicing Clean Intermittent Catheterization (CIC) were the most stressful areas of life.

In low income countries like Uganda the availability and access to specialized health care is challenging. The high numbers accessing neurosurgery and rehabilitative services in our studies reflect the bias caused by the selection method. Most of the children in our study were found through rehabilitation centers. As described above, this is most likely less than 30% of all children with spina bifida; the majority of those who do not access care die. To access care, one needs to be aware the service exists, and if available, needs time and financial and social support to access the services. The majority of the parents of children with spina bifida in our study were identified by other parents, and community based rehabilitation workers, and informed about neurosurgical options and rehabilitation. A few were referred from the health center they delivered their baby from. In Uganda the International Federation for Spina Bifida supports CURE Children's hospital and three rehabilitation centers in Kampala, Mbarara, and Gulu in the provision of low cost neurosurgery and (community based) rehabilitative care. This support starts from the creation of self-help groups of parents, and builds up via physio- and occupational therapist support using locally available and low cost materials, to the training of neurosurgeons and provision of innovative neurosurgical procedures such as ETV.

In our study we found that folic acid intake prior to and during pregnancy is very limited. This is explained by limited education and understanding of women and health workers about the importance of early folic acid intake, late presentation of women at ANC, poor supply chain and dilapidated health services caused by war and poverty in the northern region.

It was clear from these studies that poverty plays a major role in the availability and presence of health and rehabilitative services for children with spina bifida in Uganda. Without the provision of basic needs and services, the implementation of the by Uganda ratified United Nations Convention of the Right of Persons with Disabilities (2006) and the United Nations Convention on the Rights of the Child (1989) cannot be enacted.

6.1.3 Experiences of inclusion and belonging

The third research question assessed the *factors that promote social inclusion and belonging of children with spina bifida*. After setting out the community and service context of the children and their families above, we studied participation, inclusion, and sense of belonging of children with spina bifida. Belonging to a family is a central and key concept in African societies (Bigombe & Khadiagala, 2003; Chataika & McKenzie, 2013; Guyer, 1981; Malinowski, 1929).

In our study children described their households as extended family units, often not only including direct relatives such as parents, siblings, aunts, uncles, and grandparents, but also house helps and neighbours. Whilst some of the children stayed alone with one parent, the majority lived in larger households. Children, siblings and parents described situation of play, daily activities, and interaction in and around their home compounds involving each of them. Children with spina bifida had a more negative perception of themselves compared to their siblings, and showed less independence in terms of self care. Participation in household activities was a key area in daily life and belonging. Children who participated in these, expressed a sense of accomplishment. The activities provide social interaction between siblings and neighbouring children. Full participation in daily activities was hampered by mobility challenges, lack of assistive devices and poor infrastructure. Children with both spina bifida and hydrocephalus with more severe cognitive and motor skills difficulties were less likely to be involved in household activities, and were more often neglected.

Children with spina bifida in Uganda had lower cognitive outcomes compared to their siblings. The effect is larger than in children with spina bifida and their siblings in high income countries. Cognitive outcomes in our study population were predicted by age, household income, motor functioning, and schooling. Schooling was predicted by age, household income, and motor functioning. Motor functioning and continence management were predicted by age, the presence of hydrocephalus, and parental support. Starting from the end of the chain, ensuring support for parents, in terms of social and practical support in care, and livelihoods activities to increase household income is key to improving cognitive outcomes and access to school.

The next step is the creation of an inclusive environment in school and communities. The majority of children, siblings, and parents experienced negative attitudes, verbal abuse, and felt unable to participate in school and community life at some point in time. This made some children avoid or feel anxious about going to school and public places. However of children in school most enjoyed going to school and were able to develop a sense of belonging at school over time.

Physical accessibility to schools for children with spina bifida is very limited. Classroom participation is affected by lack of space, materials, knowledge and experience of teachers to use diversified teaching methods. Education performance is rated lower by teachers than parents. Inclusive policies to include the children and prevent bullying are in place but lack implementation.

Impairment, disease, and disability cannot so easily be separated (Shuttleworth & Kasnitz 2005). The lack of an enabling environment hinders children from participation. In the children's experiences their impairments are restrictive as it prevents them from engaging in daily activities such as fetching water, accessing school, or play, which they see as a missed opportunity to socialize. The children experience the world with their impaired body (Campbell, 2009; Hughes, 2007). Their experience of

societal ableism affects the sense of self worth (Daalen-Smith, 2007). Inclusion can only take place on able-bodied terms (Meekosha 2011). No mention or reference was made to policies, and human rights including the CRPD by children, siblings or parents. Only a few health workers and policy makers referred to government's responsibility to facilitate inclusion. In many communities the entire population lived in poverty with limited access to health care, education, water and other services. Our findings argue for the universal use of the biopsychosocial International Classification of Functioning, Disability and Health model of the WHO (WHO, 2001), in which body function, activity, participation, and contextual factors are mapped out and addressed in care and support programs. These can then include basic needs, as well as impairment specific needs. When mapping and addressing contextual factors in the WHO model, we follow the suggestions of Reynaert et al (2009) to examine the context of children's rights and the application of the United Nations Convention of the Rights of the Child (UNCRC), taking into account the social, economic and historical environment (Reynaert, Bouverne-de Bie, et al, 2009), and Mortier's (2002) suggestion to focus on opportunities for learning (cognitively, socially, practically etc) to empower children, and approach autonomy as a verb which facilitates discussion and interaction between parents and children (Mortier, 2002). In our context, between family and community members, and children in the home, school, and public settings.

We believe a shift can be made towards inclusion on disability terms, starting from the place where children belong: the family. Through family support children have better chances to participate and develop communities in which they belong, and create a shift in thinking about impairment, disease, and disability.

6.1.4 Family relationships, care, and belonging

The fourth research question investigated *the role of the family in social inclusion, care, and belonging of children with spina bifida*. Given the limited services available in our setting, and absence of institutional care, the families are the key care givers and persons to socially relate to in the child's life.

Stress levels of parents and caregivers of children with spina bifida are higher than parents of typically developing children (Holmbeck et al., 1997; Kanaheswari et al., 2011; Ong et al., 2011; Wallander et al., 1990). In our study parents had high stress levels and high levels of dysfunctional interaction between parents and children. In studying the perceptions of children and siblings of their families, we found mothers are the main caregivers; only a few fathers were involved in the primary care of both children with spina bifida and their siblings. This is in line with findings from high income countries (Engle & Breau, 1998; Hewlett, 2000; Lamb, 2004; Shwalb et al., 2013). In our study children with spina bifida perceived more negative interaction with their mothers compared to their siblings, but equal positive interaction and care dependency as their siblings. Most likely the increased amount of time spent with their mothers due to their physical care needs, increases negative interaction and frustration in the child-parent relationship compared to the sibling-parent relationship, as siblings may have less care needs and less time interacting with their parent.

Parental stress outcomes were related to mobility, incontinence management, access to rehabilitative care, household income, geographical region, and having support from another parent in taking care of the child. Parental stress was mostly explained by the child's inability to walk, incontinence management, and having another adult to provide support in caring for the child. Parents in Uganda felt catheterization was disruptive as assisting in catheterization, especially if this involved going to school, interfered with daily work and activities. This is in line with earlier studies in which the responsibility and anxiety around incontinence had a social and psychological impact on families (Borzyskowski et al., 2004; Kanaheswari et al., 2011).

Siblings play an important role in belonging of children with spina bifida. Children in our study perceived less positive and less negative relationships with their elder siblings, and more positive relationships and dependency with their younger siblings, compared to their brothers and sisters. Older siblings were often in school and involved in care, whilst younger siblings were more likely to be home and be able to interact and play with the child with spina bifida.

Children with spina bifida had more interaction with househelps and other in-living relatives than their siblings. Parents in northern Uganda had less support of househelps and relatives, and had significantly higher stress scores compared to parents in other regions. As outlined in the knowledge, attitudes, and practices section above, negative cultural beliefs about the impairment and conflict history may have contributed to this.

In some cases we observed child neglect, and had concerns about the child's responses on dependency and care. Whilst most parents and families were supportive and keen to include their child, some neglected their child. As Ryan and Runswick-Cole (2008) pointed out, we should be careful to describe all parents and families as mythical allies, teachers, facilitators, and advocates. Some families or parents are unable to fulfil these roles. Earlier child neglect was observed during home visits in eastern Uganda (Warf et al., 2011). Curren and Cole (2014) argue for an approach in which parent child relationships are appreciated outside the contexts of 'care' and 'normativity'. This in the Ugandan setting without external help in care is difficult, as all are intertwined and families are expected to fulfil these roles in society as institutional care is absent.

Although national policies in relation to the Universal Convention of the Rights of the Child do exist in Uganda, execution and implementation of the related legal framework is limited. The historic model of children's rights which were initiated as a response to social problems, aiming to create autonomous responsible citizens in Europe (Mortier, 2002; Bouverne-de Bie, De Vos, & Bradt, 2014), is not applicable in the Ugandan context where '*ubuntu*' ('*obuntu bulamu*' in the central region, or '*kit dano*' in the northern region), rather than social institutions still define how children are raised, protected, and belong. Families are the agents and experts of their children, the impairment, care and belonging in our context.

6.2 Limitations

Our study was limited by a small non randomized but purposely selected sample size, and small number of siblings we could involve due to logistic constraints. Due to the sampling issues at the beginning of our study, and the alternative selection method applied, our study population mostly consisted of children who received follow up and rehabilitative care. We could therefore not compare children who were in with those out of care. The children included in the interviews and assessments were selected on the ability to speak, leaving out those who may have had more significant cognitive and motor impairments. To mitigate this we did include views and experiences of parents and health care workers of children with more severe impairments in the knowledge, attitudes, and health services parts of the study.

Comparing regional differences was limited by the low numbers of children with spina bifida found in Northern Uganda. Due to the conflict and cultural practices, very few children in the age group 4 to 14 accessed surgery and survived. Of the children a large number had severe impairments and could not speak, thereby excluding them for the cognitive assessments and individual interviews.

Limitations of our study included the lack of a validated and normed cognitive assessment tool for Ugandan children with spina bifida to define cognitive assets and deficits in detail and make recommendations on neurocognitive learning goals. The feasibility and meaning of performing psychometric procedures in children with severe impairment has been questioned (Fletcher, 2014). Equally other measures of social functioning and quality of life – despite adjustments and translations to fit the socio-cultural context and impairment - were not applicable for the study population.

We attempted to present an enactment of voices of the children, siblings, and parents. We took into account observations, and used non-verbal interview methods such as play and drawing. We were however limited by translating these into verbal language. We had an expert-child bias in interpretation of the text and non-verbal communication in our study as it consisted of adult-child and expert-child interaction (Tisdall, 2012).

The main weakness was the absence of longitudinal measurements and a strong cohort of children to enable us to effectively measure the impact of surgery, rehabilitative care, and family care. No intervention effects could be tested due to this limitation.

6.3 Implications for interventions

The findings of our study have implications for interventions for children with spina bifida and children with other physical and neuro-disabilities. The results and recommendations here below will be shared with the International Federation for Spina Bifida and Hydrocephalus, the Ugandan Ministry of Health – disability, rehabilitation, and maternal and child health departments, Ministry of Education and Sports – special and inclusive education department, Ministry of Gender, Labour, and Social Development – orphans and vulnerable children’s section, the National Union of Disabled Persons in Uganda (NUDIPU), country offices of the World Health Organization (WHO), United Nations Children’s Fund (UNICEF), European Commission (EC), the Ugandan Human Rights Commission (UHRC), the Uganda National Council for Science and Technology (UNCST), the Non-Governmental Organizations (NGO) forum, and disability specific organizations.

Provision of neurosurgery, rehabilitation, and assistive devices are key services which should be provided and do remove certain barriers to inclusion. However cultural attitudes and the strong perception of social able-ism in Uganda, still override the possibilities of children being included and access services in their wider communities. We argue for building onto a network at family level where the environment is more enabling for the children to find a place of belonging and support, and expand this at community and national level. Only through a bottom up approach the paradigm at community and national level can shift from an approach of excluding children with disabilities or making them ‘fit in’ the norm, to valuing them as unique persons with a sense of belonging and ability to create a society in which they are considered as participants and actors of change.

At the family level we argue for an extended family centered approach, in which siblings and house-holds are included, and male involvement is encouraged. This approach needs to acknowledge the impact of the families’ relationships on the child with spina bifida and the effect of inclusion of the child on siblings and other family members. Parents, siblings, and other household members play an important role in actively engaging their community to see their child belongs. Parent or rather family support groups can help family members to encourage each other and feel strong in fighting for this in their communities. Rather than focusing on individual children’s rights in terms of autonomy and participation rights, we argue to look at the context in which children’s rights can emerge in an interdependent context.

At community level awareness raising and advocacy on disability to address inclusion is needed to change attitudes and involvement, and enable children with spina bifida and their families to access care and support services at community level. Early intervention and sensitisation of health workers, teachers, and communities may help to reduce waiting times and preventable deaths of new born children with spina bifida and other congenital disabilities. Knowledge about the impairments in itself may not change attitudes right away, as change was gradual and mostly reported after the child visibly developed. Alongside the existing ‘before’ and ‘after’ posters used to create awareness, we urge for the expert client approach involving parents, children and youth and eventually adults. This may be the most appropriate and successful method to shift expectations and develop a more inclusive society. We argue for more investment in community based rehabilitation to improve mobility, and care for children with spina bifida and their parents. Community Based Rehabilitation

programs should benefit children with other physical and neuro-disabilities too, as the distance and costs to reach health facilities with the right care is restrictive to many families.

In schools we argue for awareness raising on the cognitive profiles of children with spina bifida and strategies to help them in learning rather than not affordable or accessible individual testing of the child as a strategy to address their 'slow learning'. The International Federation for Spina Bifida has sensitization materials available on inclusion of children with spina bifida in schools (IFSBH, 2010). These could be adjusted and translated for use in low income countries. Further support and activities to increase participation in sports are recommended. These efforts could be embedded in a programme with a wider scope which aims at inclusion of children with physical and neuro-disabilities in school. Teachers' information on various impairments, accessibility, and learning needs combined with practical basic inclusive teaching ideas for low resource settings could be explored and piloted in schools to enhance participation and inclusion.

At national level implementation of the CRPD and policies to promote inclusion should be followed through and budgeted for. Special attention should be paid to children in the northern region.

Mandatory food fortification, sensitization of health workers, women, and improving folic acid supply at national level is required to reduce the number of children born with neural tube defects. Aside assuring folic acid supplies to health facilities and pregnant women, we recommend sensitization on the correct dosage and duration this should be taken.

Private non for profit services are currently key in service provision for children with spina bifida, creating a parallel system, taking over governmental responsibilities. We urge the Ministry of Health and government health workers to take ownership and private non for profit organizations to work hand in hand with government facilities. More efforts need to be made towards addressing physical and neuro-disabilities in the general health system, and nationally budget and staff needs to be made available to implement low cost but specialized services. Data on spina bifida and other impairments should be collected in the national health system. Currently neural tube defects are not listed as a separate category health workers can select in the national Health Management Information System (HMIS). To effectively implement the above, general health system reform and strengthening is needed.

Awareness raising on disability, inclusive education, and more active implementation of inclusion and child protection policies in school by teachers, the Ministry of Education and Sports, and Ministry of Gender, Labour and Social Development and its district offices are needed to stop bullying. To diversify teaching methods, a revision of the national curriculum and method of teaching is required, as use of visual and image activities, play, art, and use of assistive electronic devices in schools is currently not available. Having embraced the Education for All objectives, there is need for the Ministry of Education and Sports, and Ministry of Gender, Labour and Social Development to critically look at the way children are involved and taught in class.

Social inclusion and inclusive services need to be integrated in general poverty reduction. Current health, education, and social services are lacking for the entire population.

6.4 Implications for further research

Despite radio announcements and searches for children with spina bifida in the communities by local leaders, we did not find any children who had not received neurosurgery and rehabilitation. Most likely those who did not receive this died. In future a cohort study following children from birth is recommended to understand mortality, survival, development, and inclusion over time. The number of siblings included in the study was limited due to logistic constraints in follow up of children upcountry. In future studies we recommend selecting families in one or two geographical areas and follow the child, siblings, parents, and other caregivers over time.

Longitudinal studies measuring attitudes and practices at different points in time, with exposure to a child with spina bifida and/or hydrocephalus, is the preferred method of studying inclusion in society. In non-longitudinal studies, researchers can systematically ask about evolution of knowledge, attitudes, and practices over time, taking into account poverty and other environmental factors.

With regard to prevention and biomedical aspects of the impairment, we recommend further studies involving women prior to conception, during pregnancy and after birth with frequent measurements of folic acid intake to understand the actual levels of folic acid intake in the population, and evaluation studies to measure possible effects of sensitization campaigns and training of health workers in future. Further biomedical research is necessary on the incidence, causes and treatment of spina bifida and hydrocephalus in low resource settings to provide the most accessible and affordable treatment. In our study no effect was seen between children treated with ETV or VP shunts. However it could be argued that those with VP shunts were more likely to die from infections, and were therefore not included in our study group. Cohort studies may be more applicable to study the effect of medical interventions.

There is evidence for successful adaptation of assessment batteries for Ugandan children (Bangirana et al., 2009; Bangirana, Sikorskii, Giordani, Nakasujja, & Boivin, 2015; Nampijja et al., 2010). Rather than developing norm groups for specific populations including children with neuro-disabilities, we argue for a strengths based approach, in which low costs interventions rather than detailed diagnostic testing are developed. The use of narrative assessments has been suggested by Guerin for children with disabilities in New Zealand (Guerin, 2015). Goodley and Van Hove (2005) suggested the use of research design not reliant on language for children with learning disabilities, which is relevant for children in our setting with more significant cognitive impairments, and children who may be too shy to engage in interviews and assessments (Goodley & Van Hove, 2005). Pameyer argues for less individualized plans and more action oriented approaches in classrooms to increase participation of students with different learning abilities in group settings (Pameijer & van Beukering, 2010). In Flanders De Schauwer et al (2009) found that children are often helped by classmates, which either happens spontaneously or sometimes within a structured system (De Schauwer, Van Hove, Mortier, & Loots, 2009). Intervention studies in low resource settings could consist of the use of low cost inclusive action plans relevant for large class sizes in schools, sports in communities and its effect on inclusion and belonging. Studies should inform evidence-based programming for low resource settings.

To enact the United Nations Convention of the Right of Persons with Disabilities and the United Nations Convention on the Rights of the Child children with disabilities need to participate in mainstream and disability specific research (Wickenden & Kembhavi-Tam, 2014). More studies in African countries with a focus on children's perceptions and experiences on social inclusion, care, and belonging are recommended to develop African Childhood Disability Studies. In our study children described their day to day lives in terms of belonging at micro level, participating in household tasks and friendships. Whilst the individual children were interviewed, close family relationships and the importance of belonging to a family and culture, were concepts which emerged. Social change around impairment and disability is recent and complex. There are no simple solutions to create inclusion or meet the CRPD and CRC goals. Using child friendly interview tools and asking about their lives rather than impairment can open the doors to a new understanding of childhoods and disability. We argue for a family approach rather than individual quality of life approach in which childhood is studied from the child's, siblings', parental, and other household members' perspective. An individual child based approach is inappropriate in a setting where '*ubuntu*' is a key concept to inclusion, care, and belonging.

The concepts developed by Woodill et al (Woodill, Renwick, Brown, Raphael, & Goode, 1994) and Brown and Brown (2003) of being (who the person is), belonging (the people and places in a person's life), and becoming (things the person does through life) are key in studying childhood disability (Brown, 2003). However in our setting we argue for a change in order of these concepts, as belonging comes first, through '*ubuntu*' the person is defined and becomes, the individual 'is' not without the family and context. The physical, psychological and spiritual components of being are intertwined with belonging. Hall defined belonging as something much more than being socially included, he said: "To belong is to feel attached, to feel valued, and to have a sense of insidership and proximity to 'majority' people, activities, networks and spaces" (Hall, 2010). After belonging and being, a child can become.

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Summary in English

Introduction

In sub-Saharan Africa Disability Studies have largely focused on adults with disabilities and the caregivers of children with disabilities, and have primarily been conducted in South Africa. This study presents a situation analysis on social inclusion, care and belonging of children with spina bifida in Uganda. Spina bifida is a congenital disability, whereby the spinal cord and vertebrae do not form completely and the neural tube fails to develop normally. Most children with spina bifida have some degree of paralysis, which affects mobility as well as bowel and bladder control. The majority of children with spina bifida develop hydrocephalus, a condition in which the circulation of cerebrospinal fluid in the brain is obstructed, the fluid accumulates, and the head enlarges. In Uganda an estimated 1,400 children are born with spina bifida annually; about 30% receive neurosurgery, which is available in 3 locations in the country. Without surgery, most children with spina bifida die. This study focuses on the children who have survived. It describes the bodily experiences and limitations experienced by the children due to reduced functioning on the one hand and poverty, cultural, and social barriers on the other hand. The study was supported by the International Federation for Spina Bifida and Hydrocephalus and Child-Help.

Method

Between 2011 and 2015 children with spina bifida and their families were selected from the following rehabilitation centers where specialized outreach clinics and rehabilitative care are offered: CURE Children's hospital of Uganda in Mbale, Katalemwa Cheshire Home in Kampala, Gulu Regional Rehabilitation Centre in Gulu, Lira Government Hospital in Lira, and Organised Useful Rehabilitation Services in Mbarara. Semi-structured interviews, observations, and focus groups discussions were held with children, parents, hospital and rehabilitation centre staff, teachers, community members, and local leaders to study knowledge, attitudes, perceptions, access to and use of health and rehabilitation services, and experiences of belonging of children with spina bifida. A set of assessment scales to measure gross and fine motor function, daily functioning, and cognitive outcomes was compiled using existing instruments, adjusted for the Ugandan setting and study population. The Index for Inclusion was used as a guideline to measure the inclusion levels and attitudes of school administrations and teachers. The Ugandan National Accessibility Standards were used to assess physical accessibility to schools. To investigate parent child interaction, parental distress, and perceived family relationships, the Parenting Stress Index was administered and focus group discussions were held with parents. To understand belonging in the family, and the role of the family in care, the Family Relations Test was adapted and used in semi-structured interviews with children with spina bifida and their siblings. In addition home visits and observations were conducted. Data was entered and analysed using SPSS16 and Nvivo version 10.

Findings

The first research question asked about the *knowledge, and attitudes about, and perception of children with spina bifida in Uganda*. Most commonly communities believed spina bifida was caused by witchcraft, bad luck, and the use of family planning methods. Communities often assumed the mother did something wrong resulting in this misfortune. Parents would consult traditional healers, biomedical treatment and rehabilitative care. Most of them were not supported by family members in accessing care, as family members assumed the child would die and found it a waste of time and resources to invest in the child. Health workers of government health facilities held similar beliefs and were often not informed about the biomedical causes and treatment options. After communities noticed a child survived surgery, and could participate in day to day family life, attitudes changed. The child then gradually became accepted as part of the community, and social life. Whilst this was a common trend, not all parents had this experience; those in the northern region especially struggled with exclusion due to the belief that spina bifida is infectious. The social and cultural change in survival and responses to the survival is recent, and is a complex play of traditional society and cultural values, globalization and access to biomedical care.

The second research question was set to find out *which biomedical health and rehabilitation services children with spina bifida and their parents access*, as these closely relate to their survival and daily functioning. Children had poor access to general health services, and difficulties accessing specialized health services for neurosurgery and rehabilitation in our study. The majority could sit unaided; about half could walk unaided. Of those in need of assistive devices, 1/3 did not have access to them. The majority of children practiced catheterization and bowel management to manage incontinence. Incontinence management and mobility constraints were described as the most stressful areas in daily functioning. The intake of folic acid through supplements and food fortification before and during pregnancy can reduce the incidence of spina bifida. In our study only 8% of pregnant women took folic acid due to limited education and understanding of women and health workers about the importance of early folic acid intake, late presentation of women at antenatal care, poor supply chains and dilapidated health services. Poverty played a major role in the availability and presence of preventative, curative and rehabilitative health services for children with spina bifida in Uganda.

The third research question assessed the *factors that promote social inclusion and belonging of children with spina bifida*. In our study, children described their households as extended family units. Participation in household activities is a key area in daily life and belonging. In the children's experiences their impairments are restrictive as they prevent them from engaging in daily activities such as fetching water, accessing school, and play, which they see as a missed opportunity to socialize. Children with spina bifida had a more negative perception of themselves, were less likely to be in school, had lower cognitive outcomes, and showed less independence in terms of self care compared to their siblings. Children with both spina bifida and hydrocephalus with more severe

cognitive and motor skills difficulties were more likely to be neglected. The majority of children, siblings, and parents experienced negative attitudes, verbal abuse, and felt unable to participate in school and community life at some point in time. Higher household income and having support from another adult in taking care of the child improved the child's motor skills and eventually schooling and cognitive outcomes. The lack of an enabling environment hindered children from participation, e.g. for those in school, physical accessibility was very limited and classroom participation was affected by lack of space, materials, knowledge and experience of teachers to use diversified teaching methods. Inclusive policies to include the children and prevent bullying are in place but lack implementation. In many communities the entire population lived in poverty with limited access to health care, education, water and other services.

The fourth research question investigated *the role of the family in social inclusion, care, and belonging of children with spina bifida*. Given the limited services available, and absence of institutional care, the families are the key care givers and persons to socially relate to in the child's life. In our study, parents had high stress levels with parent child dysfunctional interaction. Mothers were the main caregivers; only a few fathers were involved in child care. Children with spina bifida perceived more negative interaction with their mothers compared to their siblings, but equal positive and dependent interaction. Parental stress was mostly explained by the child's inability to walk, incontinence management, and lack of another adult to provide support in caring for the child. Parents in Uganda felt catheterization was disruptive as assisting in catheterization, especially if this involved going to school, interfered with daily work and activities. Siblings and househelps played an important role in belonging of children with spina bifida. Parents in northern Uganda had less support of househelps and relatives, and had significantly higher stress scores compared to parents in other regions. Negative cultural beliefs about the impairment and the recent conflict in this region could explain this. Whilst most parents and families were supportive and keen to include their child, some neglected them.

Implications for interventions

Provision of neurosurgery, rehabilitation, and assistive devices are key services which should be provided, and can help remove certain barriers to inclusion. However, cultural attitudes and the strong perception of social able-ism in Uganda, still limit the possibilities of children being included and their ability to access services in their wider communities. We propose strengthening the network at family level, where the environment is more enabling for the children to find a place of belonging and support, and expanding investment and awareness at community and national level. At the family level, we argue for an extended family centered approach in interventions, in which family support groups include siblings and house-helps, and focus on male involvement. Rather than focusing on individual child rights in terms of autonomy and participation rights, we argue to look at child rights in an interdependent context.

At community level, awareness raising and advocacy on disability using expert children, youth, and adults to address inclusion is needed to change attitudes and involvement, and enable children with spina bifida and their families to access care and support services at community level. We argue for more investment in community based rehabilitation to improve mobility, and care. In schools, we recommend awareness raising on the cognitive profiles of children with spina bifida and low cost practical inclusive teaching pilots for teachers to enhance participation and inclusion.

At national level, implementation of the Convention of the Rights of Persons with Disabilities and policies to promote inclusion should be followed through and budgeted for. Special attention should be paid to children in the northern region. Mandatory food fortification; sensitization of health workers and women on folic acid intake; and provision of supplies to health facilities are required to prevent neural tube defects. We urge the Ministry of Health to make more efforts towards addressing physical and neuro-disabilities in the general health system, and make budget and staff available to implement low cost but specialized services in collaboration with development agencies. Having embraced the Education for All objectives and Child Protection Guidelines, there is need for the Ministry of Education and Sports, and Ministry of Gender, Labour and Social Development to critically look at the way children are involved and taught in class, and to take greater steps to ensure bullying is prevented and responded to for all children. Social inclusion and inclusive services need to be integrated in general poverty reduction.

Implications for further research

In future a cohort study following children from birth is recommended to understand mortality, survival, development, and inclusion over time. Longitudinal studies measuring attitudes and practices at different points in time, with exposure to a child with spina bifida and/or hydrocephalus, is the preferred method of studying inclusion in society. We recommend further studies involving women prior to conception, during pregnancy and after birth with frequent measurements of folic acid intake to understand the actual levels of folic acid intake in the population, and evaluation studies to measure possible effects of sensitization campaigns and training of health workers in future. Further biomedical research is necessary on the incidence, causes and treatment of spina bifida and hydrocephalus in low resource settings to provide the most accessible and affordable treatment. Intervention studies in low resource settings could consist of the use of low cost inclusive action plans relevant for large class sizes in schools, and sports in communities and assess their effect on inclusion and belonging. More studies in African countries with a focus on children's perceptions and experiences on social inclusion, care, and belonging are recommended to further develop African Childhood Disability Studies. We argue for the use of child friendly interview tools, and using a family approach in which childhood is studied from the child's, siblings', parental, and other household members' perspective. In African Childhood Disability Studies, we argue to focus on belonging first - through '*ubuntu*' ('I am because we are' or 'humanity to others') the person is defined and becomes – before looking at being, and becoming.

Nederlandse samenvatting

Introductie

Eerder onderzoek in Disability Studies in sub Sahara Afrika vond voornamelijk plaats in Zuid-Afrika en richtte zich op volwassenen met en ouders en verzorgers van kinderen met een beperking. In deze studie presenteren wij studieresultaten over sociale inclusie, zorg, betrokken- en geborgenheid van kinderen met spina bifida in Oeganda. Deze studie werd mede mogelijk gemaakt door de Internationale Federatie voor Spina Bifida en Hydrocefalie en Child-Help.

Spina bifida (open ruggetje) is een aangeboren aandoening waarin één of meerdere ruggewervels zich niet sluiten. Kinderen met spina bifida hebben in min of meerdere mate problemen met beweging (motoriek) en gevoel (sensorisch). Zij zijn aangewezen op hulpmiddelen omwille van mobiliteitsproblemen en incontinentie. De meeste kinderen met spina bifida ontwikkelen tevens hydrocefalie (waterhoofd), waarbij er een verstoorde balans is tussen productie en absorptie van hersenvocht. Spina bifida komt wereldwijd tussen 1 en 6 op de 1000 babies voor.

In Oeganda, in Oost Afrika, worden ieder jaar ongeveer 1400 kinderen met spina bifida geboren, hiervan ontvangt ongeveer 30% de noodzakelijke operatie. Voor deze operatie moeten ouders en kinderen ver reizen. Zonder operatie overlijden de meeste kinderen. Deze studie beschrijft kinderen die een operatie hebben ondergaan, de manier waarop zij hun lichaam en beperkingen die dit hen oplegt ervaren en de culturele, sociale en economische barrières waar zij mee leven.

Methode

Tussen 2011 en 2015 werden in een vijftal rehabilitatiecentra, waar maandelijks gespecialiseerde mobiele klinieken worden georganiseerd voor kinderen met spina bifida, ouders en kinderen uitgenodigd mee te werken aan deze studie. Het betrof de volgende rehabilitatiecentra: CURE Children's hospital of Uganda in Mbale, Katalemwa Cheshire Home in Kampala, Gulu Regional Rehabilitation Centre in Gulu, Lira Government Hospital in Lira en Organised Useful Rehabilitation Services in Mbarara. Om de kennis, attitudes en percepties over, toegang tot medische zorg voor, en inclusie en betrokkenheid van kinderen met spina bifida te onderzoeken werden semi-structureerde interviews, observaties en focusgroepdiscussies gehouden met kinderen, ouders, medisch en para-medisch personeel, docenten, burens, en lokale en religieuze leiders. Om motorische, dagelijkse en cognitieve vaardigheden te meten werden bestaande schalen en vragenlijsten aangepast voor kinderen met spina bifida rekening houdend met hun beperking en de

Oegandese context. De *Index voor Inclusie* werd gebruikt om inclusie in de klas te meten. De *Ugandan National Accessibility Standards* werd gebruikt om fysieke toegankelijkheid te meten. Kind-ouder interactie, stress en familie relaties werden bevraagd in interviews gecombineerd met de vragenlijst *Parenting Stress Index* voor ouders en de interactieve *Familie Relatie Test* voor kinderen. Tevens vonden huisbezoeken en observaties plaats. De verzamelde data werd ingevoerd in databestanden en geanalyseerd met behulp van SPSS16 en Nvivo 10.

Resultaten

In de eerste onderzoeksvraag onderzochten we de kennis en attitudes over en perceptie van kinderen met spina bifida in Oeganda. Spina bifida werd volgens de meeste respondenten veroorzaakt door hekserij of anticonceptiemiddelen. De schuld werd vaak bij de moeder gelegd: zij zou iets verkeerd gedaan hebben tijdens haar zwangerschap waardoor dit 'ongeluk' haar is overkomen. Ouders zochten vaak hulp in traditionele en moderne geneeswijzen. De meeste ouders ontvingen geen steun van familieleden wanneer hun kind nog jong was. De familie dacht vaak dat het kind zou komen te overlijden en vond het niet nodig geld en tijd te investeren in een kind dat toch zou sterven. Het meeste medisch personeel had dezelfde overtuiging en was niet op de hoogte van de biomedische verklaringen en behandeling van spina bifida. Wanneer het kind toch de neurochirurgische ingreep onderging en begon deel te nemen aan het dagelijks leven, werd het kind langzaam maar zeker geaccepteerd in de samenleving en ontvingen ouders meer steun van familieleden en burens. In het noorden van Oeganda bleven ouders verstoken van deze steun, omdat men hier gelooft dat spina bifida overdraagbaar is. De sociaal-culturele verandering in reactie op het overleven van kinderen na de operatie is recent en is een complex samenspel van traditionele en culturele normen en waarden, globalisering, armoede en toegang tot medische zorg.

In de tweede onderzoeksvraag bestudeerden we welke biomedische gezondheidszorg er aanwezig en toegankelijk was voor kinderen met spina bifida en hun ouders. De toegang tot tijdige neurochirurgie is direct gerelateerd aan de kans op overleven en niveau van dagelijks functioneren. De kinderen in onze studie hadden slechte toegang tot de algemene gezondheidszorg en woonden vaak op grote afstand van de gespecialiseerde gezondheidscentra voor neurochirurgie en rehabilitatie. Het grootste gedeelte van de kinderen in onze studie kon zelf zonder hulp zitten, de helft kon zelf lopen. Van de kinderen die hulpmiddelen nodig hadden om zich voort te bewegen had een derde geen toegang tot deze middelen. De meeste kinderen waren getraind in het gebruik van *Clean Intermittent Catheterization (CIC)*, een eenvoudige manier om de blaas een aantal keer per dag te ledigen met een eenvoudige, herbruikbare katheter. Kinderen met spina bifida en hun families beschreven problemen met mobiliteit en incontinentie als de grootste stressfactoren in hun dagelijks functioneren.

Het tijdig innemen van foliumzuur tijdens de zwangerschap kan spina bifida helpen voorkomen. In onze studie nam slechts 8% van de vrouwen foliumzuursupplementen tijdens de zwangerschap. Geen van de vrouwen nam deze voor de zwangerschap. Het ziekenhuispersoneel was niet op de hoogte van het belang van vroege inname van foliumzuur en de relatie met preventie van spina bifida. Naast het gebrek aan kennis, werd de inname van foliumzuur gehinderd door slechte distributiesystemen en het vrij late eerste bezoek aan de verloskundige van zwangere vrouwen. Armoede speelde een grote rol in de beschikbaarheid en toegankelijkheid tot gezondheidszorg voor kinderen met spina bifida en hun families. In de meeste gebieden waar de kinderen met spina bifida woonden, had de gehele bevolking beperkte toegang tot gezondheidszorg, onderwijs, water en andere basisvoorzieningen.

De derde onderzoeksvraag analyseerde de factoren die sociale inclusie en geborgen- en betrokkenheid van kinderen met spina bifida beïnvloeden. De kinderen in onze studie beschreven hun huis als familiehuishouden. Deelname in huishoudactiviteiten was heel belangrijk voor de kinderen om zich betrokken en geborgen te voelen. De kinderen beschreven hun beperking als sociaal beperkend omdat zij hierdoor niet mee konden doen in spel, op school, of bij het water halen bij de bron. Kinderen met spina bifida hadden een negatiever zelfbeeld, zaten minder vaak op school, hadden lager motorisch en cognitief functioneren en waren afhankelijker van ouders in vergelijking met hun broertjes en zusjes. Kinderen uit gezinnen waarin ouders hulp hadden van een andere volwassene en een hoger inkomen hadden, hadden meer kans op beter motorisch functioneren, scholing en cognitief functioneren. Kinderen met spina bifida en hydrocefalie met lager cognitief en motorisch functioneren werden vaker verwaarloosd.

De meeste kinderen met spina bifida en hun broers, zussen en ouders werden wel eens nageroepen, uitgescholden en gediscrimineerd vanwege de beperking; op school werden kinderen vaak gepest. De schoolomgeving legde veel beperkingen op voor kinderen met spina bifida: weinig tot geen toegankelijkheid voor rolstoelgebruikers, geen toiletten waar een kind katheterisatie kon uitvoeren, hoog aantal (60+) kinderen per klas, gebrek aan kennis over en materialen om docenten de mogelijkheid te geven diverse leermethodes toe te passen.

In de vierde en laatste onderzoeksvraag bestudeerden we de rol die de familie speelt in sociale inclusie, zorg, geborgen- en betrokkenheid van kinderen met spina bifida. Gezien de beperkte voorzieningen en afwezigheid van geïnstitutionaliseerde zorg, ligt de zorg voor een kind met spina bifida geheel bij de familie. Hoewel de meeste families zo goed mogelijk voor hun kinderen probeerden te zorgen, waren er ook een aantal families die het kind met spina bifida verwaarloosden. We vonden hoge stresswaarden bij ouders van kinderen in onze studie en dysfunctionele interacties tussen ouders en kinderen. Deze waarden waren hoger bij ouders met kinderen met meer mobiliteits- en incontinentie problemen, en bij ouders die geen hulp hadden van een andere volwassene in de zorg voor het kind. Moeders namen de voornaamste rol in in de zorg voor kinderen met spina bifida; er waren maar enkele vaders betrokken in de zorg. Broers, zussen en

huishoudelijke hulpen speelden een belangrijke rol in de zorg en geborgenheid van de kinderen. Ouders in noord Oeganda hadden meer stress vergeleken met ouders in andere regio's; dit is waarschijnlijk gerelateerd aan het geloof dat de beperking besmettelijk is en wederopbouw na de recente oorlog die in dit gebied nog in volle gang is.

Aanbevelingen voor interventies

Gezondheidszorg, neurochirurgie, fysiotherapie, en het aanbieden van hulpmiddelen zijn basis gezondheidsvoorzieningen nodig voor kinderen met spina bifida. Hoewel deze voorzieningen meehelpen inclusie te bevorderen, is vooral ook sociale verandering nodig, zodat kinderen mee mogen en kunnen doen. Dit begint thuis, waar kinderen zich veilig en geborgen voelen, zich kunnen ontwikkelen en zich vervolgens welkom voelen in hun buurt, op school en het publieke leven.

Op familieniveau is steun voor families van kinderen met spina bifida belangrijk. Ondersteuning kan plaatsvinden door middel van familiegroepen waarin niet alleen ouders, maar ook grootouders, broers, zussen, andere familieleden en huishoudelijke hulpen worden betrokken. In plaats van ons te richten op kinderrechten in termen als autonomie en participatie, bevelen wij aan te kijken naar kinderrechten in de context van afhankelijkheid en armoede en positieve zorgsystemen die hieruit voortvloeien te versterken.

Het verspreiden van informatie en het voorlichten van de bevolking over spina bifida en andere beperkingen is nodig om attitudes en gedrag te veranderen en kinderen de mogelijkheid te geven deel te nemen aan het dagelijks leven. Huisbezoeken en opvolging van het kind in zijn of haar omgeving is belangrijk om mobiliteit en zorg te verbeteren. Op scholen is voorlichting nodig over spina bifida en gerelateerde cognitieve beperkingen. Simpele, goedkope en laagdrempelige methodes voor docenten om kinderen meer te betrekken in de klas en groepsonderwijs aan te bieden voor kinderen met verschillende leerniveaus kunnen worden getest om inclusie te bevorderen.

Op nationaal niveau is de navolging en praktische implementatie van de ondertekende Conventie voor de Rechten van Mensen met een Beperking (Convention of the Rights of Persons with Disabilities) noodzakelijk om inclusie mogelijk te maken. Specifieke aandacht is nodig voor kinderen en families in noord-Oeganda. Het toevoegen van foliumzuur aan voedingsmiddelen, voorlichten van medisch personeel, en het verbeteren van distributie van foliumzuursupplementen aan ziekenhuizen en lokale klinieken zijn belangrijk in de preventie van spina bifida. Aanpassingen in de algemene gezondheidszorg zijn nodig om budget en personeel vrij te maken om kinderen met een beperking de zorg te bieden die zij nodig hebben. Deze zorg kan met behulp van goedkope en effectieve middelen aangeboden worden. Samenwerking met privé-instanties die gespecialiseerde zorg aanbieden is hoogstnoodzakelijk om het parallele bestaan en de kloof tussen publieke en privé-zorg te verkleinen. In navolging van de *Education for All* (EFA) doelen en kinderbeschermingsrichtlijnen is het tijd voor de ministeries van onderwijs, gezondheid, gender en sociale ontwikkeling, te zorgen dat deze

richtlijnen toegepast worden door docenten, medisch personeel en maatschappelijk werkers. Sociale inclusie en inclusieve zorg is een onderdeel van algemene armoedebestrijding.

Aanbevelingen voor onderzoek

In toekomstig onderzoek bevelen we aan een cohortstudie op te zetten die kinderen vanaf hun geboorte volgt. Op deze manier kunnen we sterfte, ontwikkeling en inclusie beter begrijpen. Longitudinale studies om attitudes en gedrag te meten op verschillende meetmomenten kan ons meer inzicht geven in interventies die cruciaal zijn voor inclusie op verschillende tijdstippen.

Meer studies zijn nodig om de inname van foliumzuur en voorlichtingsprogramma's te bestuderen. Verder biomedisch onderzoek is nodig om de incidentie, oorzaken en het verloop van spina bifida en hydrocefalie te bestuderen in de Afrikaanse context in relatie tot neurochirurgische ingrepen en rehabilitatievoorzieningen.

Wij bevelen interventiestudies aan om inclusief onderwijs te bevorderen, evenals participatie via sport. Meer studies die kinderen met een beperking centraal stellen in sub-Sahara Africa zijn noodzakelijk om het perspectief van de kinderen meer naar voren te brengen en African Childhood Disability Studies verder te ontwikkelen. Door middel van het gebruik van kindvriendelijke onderzoeksmethoden en te beginnen waar kinderen zich het meest geborgen voelen, thuis, kan een familieperspectief beschreven worden. Dit moet worden gerelateerd aan 'ubuntu': 'ik ben omdat wij zijn' of 'het diep gewortelde mensbesef, dat wij allen onlosmakelijk met elkaar verbonden zijn'. In *African Childhood Disability Studies*, komt betrokkenheid en geborgenheid eerst, daarin is en ontwikkelt het individu zich, alle fasen in de context van 'ubuntu'.

Data sheets

% Data Storage Fact Sheet

% Name/identifier study: children with spina bifida in Uganda

% Author: Femke Bannink

% Date: 19 May 2016

1. Contact details

=====

1a. Main researcher

- name: Femke Bannink
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2. Information about the datasets to which this sheet applies

=====

* Reference of the publication in which the datasets are reported:

1. Bannink, F., Stroeken, K., Idro, R, and van Hove G. (2015) Community Knowledge, Beliefs, Attitudes, and Practices Towards Children with Spina Bifida and Hydrocephalus in Uganda. *Journal of Disability, Development and Education* 62: 2, 182-201, 2015. DOI: 10.1080/1034912X.2014.998174.
2. Bannink, F., Idro, R., and van Hove, G. (2016) 'I like to play with my friends: children with spina bifida and belonging in Uganda. *Social Inclusion*.
3. Bannink, F., Idro, R., and van Hove, G. (2016) Parental stress and support of parents of children with spina bifida in Uganda. *African Journal of Disability*,

* Which datasets in that publication does this sheet apply to?:

This sheet applies to all datasets used in the publication 1 and all narrative interview data from publications 2 and 3, and chapters 1, 3, and 4 of this dissertation.

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3a. Raw data

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* Who has direct access to the raw data (i.e., without intervention of another person)?

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* Reference of the publication in which the datasets are reported:

Bannink, F., Larok, R., Bauwens, L., Kirabira P., and van Hove, G. (2015) Prevention of spina bifida: folic acid intake during pregnancy in northern Uganda. The Pan African Medical Journal 20: 90. DOI: 10.11604/pamj.2015.20.90.533.

* Which datasets in that publication does this sheet apply to?:

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* Reference of the publication in which the datasets are reported:

1. Bannink, F., Fontaine, J.R.J., Idro, R., and van Hove, G. (in press) Cognitive abilities of pre- and primary school children with spina bifida in Uganda. *International Journal of Educational Psychology*.
2. Bannink, F., Idro, R., and van Hove, G. (2016) Parental stress and support of parents of children with spina bifida in Uganda. *African Journal of Disability*. In print.
3. Bannink, F., Idro, R., and van Hove, G. (2016) 'I like to play with my friends: children with spina bifida and belonging in Uganda. *Social Inclusion*.

* Which datasets in that publication does this sheet apply to?:

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3. Information about the files that have been stored

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* Who has direct access to the raw data (i.e., without intervention of another person)?

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- responsible ZAP
- all members of the research group
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- file(s) describing the transition from raw data to reported results. Specify: see methods section of the articles
- file(s) containing processed data. Specify: An SPSS file contains the processed data
- file(s) containing analyses. Specify: see findings section of the articles
- files(s) containing information about informed consent. Blank copy is saved on my PC.
- a file specifying legal and ethical provisions. The documents submitted and approved by the Ethical and Research Commissions in Belgium and Uganda are filed on my and the co-researcher's PC
- file(s) that describe the content of the stored files and how this content should be interpreted.

Specify: ...

- other files. Specify: ...

* On which platform are these other files stored?

- individual PC
- research group file server
- other: ...

* Who has direct access to these other files (i.e., without intervention of another person)?

- main researcher
- responsible ZAP
- all members of the research group
- all members of UGent
- other (specify):

4. Reproduction

=====

* Have the results been reproduced independently?: YES / NO

* If yes, by whom (add if multiple):

% Data Storage Fact Sheet

% Name/identifier study: children with spina bifida in Uganda

% Author: Femke Bannink

% Date: 19 May 2016

1. Contact details

=====

1a. Main researcher

- name: Femke Bannink
- address: PO BOX 9850 Kampala, Uganda
- e-mail: femke.bannink@ugent.be

1b. Responsible Staff Member (ZAP)

- name: Prof.dr. Geert van Hove
- address: Henri Dunantlaan 2, 9000 Gent
- e-mail: Geert.VanHove@UGent.be

If a response is not received when using the above contact details, please send an email to data.pp@ugent.be or contact Data Management, Faculty of Psychology and Educational Sciences, Henri Dunantlaan 2, 9000 Ghent, Belgium.

2. Information about the datasets to which this sheet applies

=====

* Reference of the publication in which the datasets are reported:

Bannink, F., Idro, R., and van Hove, G. (in press) Family relationships, dependency and care: perspectives of children with spina bifida in central Uganda. Knowledge Management for Development Journal. Special issue Knowledge for Disability Inclusive Development.

* Which datasets in that publication does this sheet apply to?:

This sheet applies to all datasets used in the publication, and chapter 4 of this dissertation.

3. Information about the files that have been stored

=====

3a. Raw data

* Have the raw data been stored by the main researcher? YES / NO

If NO, please justify:

* On which platform are the raw data stored?

- researcher PC
- research group file server
- other (specify): computer of the International Federation for Spina Bifida and Hydrocephalus Uganda office. Hardcopies in the archive of the same office.

* Who has direct access to the raw data (i.e., without intervention of another person)?

- main researcher
- responsible ZAP
- all members of the research group
- all members of UGent
- other (specify):

3b. Other files

* Which other files have been stored?

- file(s) describing the transition from raw data to reported results. Specify: see methods section of the article

- file(s) containing processed data. Specify: An SPPS file contain the processed data

- file(s) containing analyses. Specify: see findings section of the article

- files(s) containing information about informed consent. Blank copy is saved on my PC.

- a file specifying legal and ethical provisions. The documents submitted and approved by the Ethical and Research Commissions in Belgium and Uganda are filed on my and the co-researcher's PC

- file(s) that describe the content of the stored files and how this content should be interpreted.

Specify: ...

- other files. Specify: drawing made by the children

* On which platform are these other files stored?

- individual PC

- research group file server

- other: ...

* Who has direct access to these other files (i.e., without intervention of another person)?

- main researcher

- responsible ZAP

- all members of the research group

- all members of UGent

- other (specify): co-researcher

4. Reproduction

=====

* Have the results been reproduced independently?: YES / NO

* If yes, by whom (add if multiple):

PhD portfolio

Publications

Mertens, P. and Bannink F. (2012) Interdisciplinary care for children with spina bifida in East and Southern Africa. *Journal of Medicine and the Person*, 10, 3: 129-135. DOI 10.1007s/12682-012-0126x.

Bannink, F., Idro, R., and van Hove, G. (2015) Community knowledge, attitudes, and practices in caring for children with spina bifida in Uganda. *Journal of Disability, Development and Education* 62:2, 182-201, DOI: 10.1080/1034912X.2014.998174.

Bannink, F., Larok, R., Bauwens, L., Kirabira P., and van Hove, G. (2015) Prevention of spina bifida: folic acid intake during pregnancy in northern Uganda. *The Pan African Medical Journal* 20: 90. DOI: 10.11604/pamj.2015.20.90.533.

Bannink, F., Idro, R., and van Hove, G. (2016) Parental stress and support of parents of children with spina bifida in Uganda. *African Journal of Disability*, 5(1). DOI: 10.4102/ajod.v5i1.225.

Bannink, F., Idro, R., and van Hove, G. (2016) 'I like to play with my friends': children with spina bifida and belonging in Uganda. *Social Inclusion*, 4 (1). DOI: 10.17645/si.v4i1.630.

Bannink, F., Idro, R., and van Hove, G. (2016) Teachers' and parents' perspectives on inclusive education for children with spina bifida in Uganda. *Journal of Childhood & Developmental Disorders*, 2: 10. DOI:10.4172/2472-1786.100026.

Bannink, F., Fontaine, J.R.J., Idro, R., and van Hove, G. (in press) Cognitive abilities of pre- and primary school children with spina bifida in Uganda. *International Journal of Educational Psychology*.

Bannink, F., Idro, R., and van Hove, G. (in press) Family relationships, dependency and care: perspectives of children with spina bifida in central Uganda. *Knowledge Management for Development Journal. Special issue Knowledge for Disability Inclusive Development*.

Presentations

“Stigmatization, discrimination and exclusion of children with spina bifida and hydrocephalus in northern Uganda” oral presentation at the International Conference Children and Youth Affected by Armed Conflict. Kampala, Uganda, September 26 – 28, 2013.

“Mobility, incontinence, and social functioning of children with spina bifida in Uganda” oral presentation at the Disability Studies International Conference, organized by Disability Studies Netherlands. Amsterdam, The Netherlands, November 1 – 3, 2013.

“Experiences of children living with spina bifida and their parents in Uganda” poster presentation at the Disability Studies International Conference, organized by Disability Studies Netherlands. Amsterdam, The Netherlands, November 1 – 3, 2013.

“Adaptation of the Family Relations Test for use in children in Uganda”, oral presentation at the East African International Conference of Psychology, organized by the East African Psychology Centre. Kampala, Uganda, November 6 – 8, 2013.

Courses*Faculty courses*

Disability Studies, Prof. Dr. Geert Van Hove, 2012

Interpretatieve Onderzoeksmethoden, Prof. Dr. Geert Van Hove, 2012

Medische antropologie: Afrika en de diaspora, Prof. Dr. Koenraad Stroeken, 2012

Onderzoekseminarie afrikaanse talen en culturen, Prof. Dr. Koenraad Stroeken, 2012

Orthopedagogiek en ontwikkelingssamenwerking, Prof. Dr. Geert van Hove, 2012

*Doctoral school courses**Transferable skills*

Cluster Communication - Poster and Paper, waived based on work experience, 2012

Cluster Research and valorisation - Basis Statistics - Uganda Virus Research Institute, Entebbe 2012

Cluster Career management - Project Management, waived based on work experience, 2012

Specialist courses

African workshop International Federation for Spina Bifida and Hydrocephalus, Kampala 2011

Using Regression Wisely – Uganda Virus Research Institute, Entebbe 2012

NVIVO Qualitative Software – University of California San Francisco, Kampala 2013