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Cover page

Specialist hospital treatment and care as reported by children with intellectual disabilities and a cleft lip and/or palate, their parents, and healthcare professionals.

Abstract

Background

Research into hospital treatment and care of children with intellectual disabilities (IDs) is extremely limited but available literature points to difficulties. Some children have a co-occurring condition alongside an ID which requires ongoing treatment, such as a cleft lip/palate. To date, their experiences remain untapped.

Method

Semi-structured interviews with 23 participants comprising children (n=5) (aged 11-16) with intellectual disabilities, their parents (n=9) and healthcare professionals (n=9) working in cleft care. Thematic Analysis determined patterns across the data.

Results

Three key themes were found; struggles (stress and distress, power imbalance) tensions (perceived levels of choice and control in decision-making, lack of training around IDs assumptions and jargon) and good practice (appropriate communication and information, tailored treatment).

Conclusion

Good practice was evident, but was ad-hoc. Individualised treatment and communication based upon children's needs is required as is further investigation into general anaesthetic induction for children with IDs.

Keywords

Children, intellectual disabilities, cleft, healthcare, hospital

Introduction

People with IDs have greater healthcare needs compared with the general population (Perry et al., 2014) and disabled children have higher hospital admission rates compared to non-disabled peers (Mahon & Kiburge, 2004). The United Nations Convention of the Rights of the Child (UNCRC, 1989) and the Rights of Persons with Disabilities (2006) asserts that disabled children/adults should be involved in their care and in decision-making. However, literature on the healthcare experiences of people with IDs predominantly highlights difficulties. Krahn, Hammond and Turner's (2006) review (including research from the United States (US), the United Kingdom (UK) and Israel) captured the 'cascade of disparities' (p.70) for people with IDs in healthcare such as limited attention to care needs and health promotion, and insufficient access to healthcare. Increased familial support and healthcare co-ordination were subsequently recommended. In Backer, Chapman and Mitchell's (2009) review of healthcare for people with IDs (including research from Australia, the UK and Northern Ireland), themes included fear of hospitals, lack of clear information and communication and an absence of choice in decision-making. Recent international research does not demonstrate improvements. Staff attitudes, communication problems and consent issues were cited as barriers to adequate healthcare for people with IDs in Ireland (Doyle et al., 2016). An Australian study reported on the hospital experiences of older adults with IDs (living in group homes) from carer/group home staff perspectives. Although positives were reported such as calm, patient healthcare professionals (HCPs) who allocated more time to procedures, participants referred to communication failures, hospital staff seeming uncomfortable around those with IDs, and suggested some people with IDs were considered unworthy of further treatment (Webber, Bowers, & Bigby, 2010). Lunsky, Tint, Robinson, Khodaverdian, & Jaskulski, 2011) described a Canadian study with 20 people with IDs who

Intellectual disabilities cleft hospital

had experienced a psychiatric crisis and consequently visited the emergency department. Key concerns raised were lack of consultation with caregivers and lack of staff training. In a Swedish study on childbearing experiences of ten Mothers with IDs, participants reported that the hospital was confusing and associated routines challenging (Hoglund & Larrsson, 2013). Pain relief was also inadequate. Iacano, Bigby, Unsworth, Douglas and Fitzpatrick's (2014) systematic review extended Backer's review but revealed 'little additional insight' (p.4). Therefore, the international picture, although scant, portrays a bleak view of hospital experiences for adults with IDs. Research into children's experiences is further limited.

In one of the few published studies about children with IDs' hospital experiences, Brown and Guvenir (2009) carried out UK research with 13 parents/carers, 13 nursing staff and two children. Children reported anxieties about hospital, using the term 'scary' to describe their emotions. Similarly, their parents spoke of feeling nervous and apprehensive, with fears exacerbated if they felt unprepared for treatment. Healthcare staff may not receive appropriate training for working with children with IDs (Ong et al., 2017), potentially escalating challenges. Scott, Wharton and Hames' (2005) UK research into the hospital experiences of 14 young people with IDs highlighted limited communication between themselves and staff, staff dealing directly with parents and not them, feelings of fear and uncertainty, alongside boring waiting rooms and ward environments. Oulton, Sell and Gibson's (2018) UK ethnographic research highlighted what was important to nine children and young people with IDs (and their parents) in a hospital ward and in an outpatient department. Five key themes found were; little things make a big difference; stop unnecessary waiting; avoid boredom; the importance of routine and home comforts, and never assume (p.1). Despite people with IDs often having co-occurring conditions requiring

long-term management and procedures in hospital settings, studies which include the voice of children with IDs remain scarce (Aston, Breau, & MacLeod, 2014). For example, the views of children with IDs who regularly experience needle-related procedures, are rarely considered (Pascolo et al., 2018). One example of a co-occurring condition requiring multiple treatment interventions is a cleft lip and/or palate.

A cleft lip is a split between the nose and lip and is congenital. If it features on one side, it is called a unilateral cleft and a bilateral cleft if it appears on both sides. Nearly half of cleft lips occur with a cleft palate (the roof of the mouth has not fused.). Worldwide, approximately 1 in 700 babies are born with a cleft (Mossey & Castillia, 2001). It either occurs as a single impairment or is part of a syndrome (Lees, 2001).

The UK cleft care pathway comprises a series of operations and treatment, beginning with a cleft lip repair at 3-4 months and palatal closure at 6-9 months (Paliobei, Psifidis, & Anagnostopoulos, 2005). Although treatment varies depending upon cleft type and severity, there are planned common clinical events. Figure 1 is a UK cleft care example pathway.

Figure 1 here

International research suggests 7-18% of those with clefts have IDs (e.g. Chetpakdeechit, Mohlin, Persson, & Hagberg, 2010; Mueller, Sader, Honigmann, Zeilhofer, & Schwener-Zimmerer, 2007). However, just one study (from the US) has elicited the views of children and young people with IDs and clefts (aged 4-19) on treatment outcomes (appearance and speech) and self-ratings of social and cognitive skills (Strauss & Border 1993). Experiences of medical services and decision-making were not considered.

A key component of effective healthcare is shared decision-making whereby patients, their families and HCPs communicate to explore options, and decide the best treatment based on available information, evidence and preferences (Lipstein, Lindly, Anixt, Britto, & Zuckerman, 2016). Legally, children under 16 in the UK are presumed competent to make treatment decisions if they sufficiently understand and are mature enough to fully comprehend what is being suggested (Department of Health (DoH), 2001). However, even without legal competence, children's voices should form part of decision-making to facilitate trust, co-operation and enable future decision-making (Mouradian, 1999). Appropriate information and time can assist competency development (DoH, 2001).

For those with IDs, it is best practice to promote self-determination (Wehmeyer & Shogren, 2016) and decision-making rights (Blanck & Martinis, 2015). However, the voices of people with IDs are not always heard even in situations which have profound impacts on their day-to-day lives (Smyth & Bell, 2006).

The current study aimed to qualitatively explore how children with clefts and IDs, their parents and HCPs perceived their specialist cleft service e.g. accessibility, treatment, and decision-making input.

Challenges when engaging children with IDs in research include negotiating access via their parent/guardian and ensuring they understand what participating in research means (Cameron & Murphy, 2006). The consent/assent process may be challenged by attention and memory problems and impaired communication (Cameron & Murphy, 2006). People with IDs may acquiesce; offer responses which they think the researcher wants to hear rather than revealing their true opinion (D'Eath et al., 2005). Although research has engaged

children with IDs, parent and professional perspectives are more prevalent. Given the paucity of research with children with IDs, Walmsley and Johnson (2003) advocated for further studies that uncover this populations' experiences with a view to improving their lives (Walmsley & Johnson, 2003). Eliciting children's views about their hospital care is therefore paramount. As Oulton et al. (2018) stressed, "the views of children and young people with intellectual disabilities about being in hospital are rarely sought" (p.2), additionally citing a "major gap in the evidence base" (p.4). Cleft research has also traditionally excluded those with cognitive impairments. This study attempts to fill this gap, demonstrating that children with IDs can participate in cleft research.

Methods

Given the new research area, an exploratory qualitative design and methodology was appropriate to gain families' and HCPs' views about their experiences of specialist hospital treatment (Flick, 2006). Gaining parents' and HCPs' views in addition to children's was useful to capture multiple perspectives owing to the dearth of research.

This study was underpinned by contextualism which acknowledges that people can convey their personal realities, but that economic, social and cultural factors influence that reality (Willig, 1999). Therefore, participants communicate their experiences which are valid in their own terms (Bhaskar, 1978), and are partially driven by their social context (Braun & Clarke, 2006). To meaningfully draw out people's experiences, interviews were the chosen method for this study.

Semi-structured interview questions were based on previous ID and cleft research. Child and parent interview topics included outpatient clinic experiences and cleft treatment, including

Intellectual disabilities cleft hospital

decision-making. HCP interview topics included information needs, treatment and decision-making.

A favourable ethical opinion was gained from the UK (Central London) National Research Ethics Committee and approval given (reference number: 11/LO/1778).

Sample and recruitment

Children aged 10-16 with IDs and a cleft were eligible to participate, as were their parents and HCPs working in cleft care. This age bracket deliberately coincided with various treatment (e.g. surgical/orthodontic) on the UK cleft care pathway (see figure 1). Cleft service records did not detail diagnoses of IDs and staff reported this was because they operated within a clinical service, rather than defining people with other needs. It was therefore pragmatic to use criteria regarding support for IDs instead. Such support meant being in contact with a Community Learning (Intellectual) Disability Team, receiving respite care, or having school support for IDs. Children might attend a special educational needs school and/or have an Education, Health and Care (EHC) plan (such plans establish children's educational, health and social needs and detail the extra support required to meet those needs (Boesley & Crane, 2018)). The researcher aimed to interview children who had mild-moderate LDs, allowing for verbal contributions.

The total number of participants was 23, comprising five children with IDs, their parents (n=9 - three interviews were joint with both Mother and Father present), and nine HCPs. All were White British bar one child. All participants were recruited from a regional cleft care unit in the South West of England between 2012 and 2015. Attending a particular cleft care unit means that patients can potentially be treated at various regional hospitals. The precise

extent of hospital experiences of the five families is unknown, but figure 1 shows a UK cleft care pathway example. Table 1 describes child participant characteristics.

Table 1 here.

Cleft team staff initially made contact with eligible families who were then sent written details about the research. The researcher then telephoned families to answer questions and to discuss whether parents were happy for themselves and their child to participate. Their child's communication preferences were discussed and parents confirmed their child could verbally participate in the interview. Children were offered one month to process and understand the information, which was enhanced by pictures and short simple sentences, as recommended by Cameron and Murphy (2006).

Written consent forms were completed by parents for their child's participation. Children gave written assent. Upon initially meeting the children, the researcher repeated the information. As per Perry's (2004) recommendations, this was done with their parent present. Subsequently, if and when children verbally agreed to participate, parents could help complete the assent form. All children agreed their interview could be audio-recorded. Confidentiality and anonymity were stressed (unless a child protection issue was raised). Children were aware they could withdraw their data and all interviews took place in the family home so was familiar, hopefully aiding their comfort. To help children feel more comfortable before the interview, the researcher established rapport by chatting with them about television programmes and school holiday activities as per Prosser and Bromley's (1998) guidance. The researcher stressed there were no right or wrong answers as it was their views and experiences which were of interest. Breaks were offered as were further conversations at a later date. Children could choose whether or not their parents were

Intellectual disabilities cleft hospital

present for the interview, and all chose to do this. This was sometimes helpful as parents could rephrase questions to facilitate their child's understanding. It could be seen however as over-protection (Christensen & Prout, 2002) and some children may talk about things differently if they know others can hear (Gardner & Randall, 2012). Child interviews lasted between 16 and 27 minutes. Children received a thank you certificate at the end of the research.

Parents were also interviewed in their familiar home setting. Written consent to participate and an agreement for the interview to be audio-recorded was received. Confidentiality (notwithstanding child protection issues) was assured, as was the option to withdraw their data. Parent interviews lasted between 21 minutes and 2 hours 40 minutes. Table 2 details parent participant characteristics.

Table 2 here.

Parents and children received an accessible summary of the research findings.

Nine HCPs working in cleft care were interviewed. Written consent was given alongside guarantees of anonymity and confidentiality (not withstanding child protection issues). HCPs were reminded they could withdraw their data before the research was written up. HCPs from across disciplines participated (speech and language therapy (n=2), psychology (n=2), clinical nurse specialists (n=2) and surgical/orthodontic consultants (n=3)) (see Table 3).

Table 3 here.

HCP interviews took place in a private hospital room, were audio-recorded with consent, and lasted between 16 and 64 minutes. A presentation on the findings was given to HCPs after the study.

Analysis

Thematic Analysis (TA) was used to analyse the interview transcriptions to identify patterns, similarities and differences which was appropriate for an un-researched topic (Vaismoradi, Turunen, & Bondas, 2013). Braun and Clarke's (2006) stages of TA guided the analysis from the (inductive) generation of codes to defining and naming themes. To enhance the quality and validity of the analysis, the authors discussed and agreed the coding and theoretical framework. Such discussions continued until saturation point. Themes were agreed using various criteria. In some instances, the number of participants who expressed a certain theme were noted (if there were repeated references to a particular phenomenon), but this did not primarily shape analysis. Other criterion were used, such participants' strength of feeling or if they were spontaneous, unsolicited accounts.

Three key themes; struggles, tensions, and good practice in hospital/clinic. These will now be described, using illustrative quotes from interviews across participant groups.

Results

Theme 1: Struggles

This theme pertained to hospital-related struggles felt by children and parents, as echoed by HCP accounts and had two subthemes; stress and distress and power imbalance. All children interviewed were anxious about hospital and reported difficult experiences. Two children, Chloe and Emily, repeatedly and in Chloe's case, spontaneously, referenced the type of needle used for general anaesthetic (GA) administration:

Participant (P): I hate having a needle in my hand.

P: I hate having a needle in my hand.

Intellectual disabilities cleft hospital

P: I hate having a needle in my hand.

Chloe (aged 14)

Interviewer (I): So...what happens at clinic?

P: Cannula.

I: Do you talk about the operation and whether it's going to hurt or not, things like that?

P: Cannula.

Emily (aged 15)

Given Chloe and Emily's ages (14 and 15) they would have had repeated GAs as part of their ongoing treatment. Their spontaneous accounts highlighted their significance because the interviews did not specifically ask about GAs.

James referred to needles:

P: I don't like hospitals.

I: Why don't you like them?

P: 'Cause they always give me jabs.

James (aged 11)

James would have had blood tests and post-operative medications as part of his surgical care.

Parents also spoke of their child's reactions to surgery:

P (Mother): The cannula is awful, we have screaming fits. She doesn't say, 'Mum what about this operation?' It's 'Mum I'm going to try and be brave over the cannula this time.'

*P (Father): The operation takes second place really.
Mother and Father of Emily (aged 15)*

*P: She screams and they have to pin her down.
Mother of Chloe (aged 14)*

The account of a young person being pinned down for GA induction was unexpected and concerning. Parents also witnessed their child's post-operative distress:

*P: He come out the operating theatre and he was screaming...he was trying to rip at his bandage.
Father of Liam (aged 16)*

Struggles faced by families were echoed in HCP interviews who, like children and parents, referred to surgery-related difficulties:

P: ...it's making sure that we pre-empt issues partly by educating anaesthetists...and flagging up issues, putting things on the front of medical notes before surgery...this child [with IDs] is going to be very anxious about anaesthetic, is going to be given pre-med, can you consider different ways of

anaesthetising these children to reduce distress and get the play team involved so they are distracting the child at the time.

Psychology team member 1

These observations were generated by the participant's increasing involvement with the hospital ward which offered them insights into children's experiences. Post-surgical trauma was experienced by children with IDs if they were unsure of what was happening:

P: There can be traumatic psychological effects from [surgery]... [surgeon] talks about children [with IDs] who've maybe been dry at night starting to bed-wet, or having nightmares because of this thing that's happened to them, because they've not quite understood what it is, why they've gone from being well and perfectly happy, to going into hospital and this thing being done to them.

SLT team member 2

Parents articulated power imbalances felt within clinic:

P: Sometimes I felt that they were far superior than us and I felt a little bit belittled.

Mother of Emily (aged 15)

P: It's a daunting experience...you've got all this focus with all these people...they make their decisions so quickly and you

don't know what's going on...When you go there, everyone wants to see you and they're poking about...they're all talking about your child as if you weren't there.

Mother of Matthew (aged 11)

Theme 2: Tensions

The theme tensions highlighted three subthemes across participant groups; perceived levels of choice and control in decision-making, lack of HCP training in IDs and assumptions and jargon.

Children highlighted that surgical decision-making was doctor-led:

I: Who...decides what you have done?

P: Um, the doctor.

Chloe (aged 14)

I: When you have the treatment, and when you have surgery...who decides what treatment you're going to have?

P: The doctors.

Matthew (aged 11)

This stance was echoed by parents:

P: You tend to go with it, and think, well they're the experts, they know.

Mother of Emily (aged 15)

Referring to HCPs as ‘the experts’ was echoed by other parents. HCPs also questioned surgeon’s abilities to actively listen to children:

P: Surgeons are very good at asking young people what they want whether they have [an ID] or not...they’re not so good at listening to the answer.

Psychology team member 2

When facilitating conversation with children with IDs, some HCPs used the terms ‘common sense’ and ‘goodwill’, indicating a lack of training/professional guidance:

P: *The skills to enable a child to talk are not necessarily there and why would they be, they’re [staff] not trained...sometimes that’s where we fall down.*

Psychology team member 2

P: *I don’t understand how I can communicate some of the information so [children with IDs] understand it and make decisions...we are better than we were, but it’s work in progress.*

Psychology team member 1

HCPs suggested that people with IDs and their families could be involved in delivering future training about IDs. Training on disability legislation was seen as potentially helpful. A psychology team member remarked they were probably the only multidisciplinary team member who knew of the hospital’s Learning (Intellectual) Disability Liaison Nurse (LDLN).

Intellectual disabilities cleft hospital

Assumptions and jargon were referred to by HCPs which could indicate a lack of training/awareness. Staff talked candidly about assumptions that led to excluding children with IDs from appointments:

P: There is a danger sometimes with children [with IDs] that assumptions...are made...the conversation can go round the child, and is... directed to the parents, the assumption being that they don't understand anyway.

Psychology team member 1

P: The [cleft team] don't always explain things in simple language...you just lapse into jargon sometimes and it's very very bad to do that.

SLT team member 1

This account echoed parents' interview data, and evidenced that staff acknowledged the mismatch between the plain language they should use and the reality within clinic. There was an awareness of an over-reliance on written information:

P: There's too much focus on information geared towards [those] who are not struggling with learning. It's...very literacy-led so our information tends to be leaflets, some of the language is too complex...The letters we write can be very erudite and a bit too academic-ish.

Psychology team member 1

Intellectual disabilities cleft hospital

Assumptions were made by a consultant and a Clinical Nurse Specialist (CNS) who commented that children with IDs were typically given a GA for dental extractions, as they could not cope when awake:

P: I don't think we treat many [children with IDs] differently, but we do have a lower threshold for GA.

Consultant 1

P: If they [children with IDs] [need] some teeth out to help everything straighten up or some fillings and they can't cope with it in the dental chair we will...take them for GA...whereas a child without a disability who says 'No I don't want that done' we won't take them for a GA...our threshold for allowing [children with IDs] to go for GA is lower almost, but it has to be...because there is sometimes stuff...that actually does need doing...and they can't cope with it awake.

CNS 1

This is noteworthy because children and parent data emphasised acute distress at GA induction, so there is a tension there.

Theme 3: Good practice

This theme contained two subthemes, appropriate communication and information, and tailored treatment. Parents explained the help and respect shown to their child by HCPs in the multidisciplinary cleft team:

P: A couple of appointments ago, James had a funny five minutes...He went in there and as soon as the doctors started to talk to him, he turned round and said 'I ain't talking to you' and walked out. But the doctor was very understanding and he did give me a bit of time to go and calm him back down and get him to come back in, rather than say, 'well sorry mate, your time's up, I can't deal with him now, you have to make another appointment'. They didn't. They did just wait and they did give me time to settle him...we went on in, and they was alright about it.

Mother of James (aged 11)

P: [Surgeon] also asks permission... 'is it alright if I touch your lip?' or 'alright if I look in your mouth?' 'Are you okay with me doing that?'...I think from Liam's point-of-view that's made him a lot easier instead of someone going at him and just, 'right, come here, you're here at an appointment. I'm gonna look in your mouth, this is what we're here for, now don't mess me around'.

Mother of Liam (aged 16)

The importance of HCPs giving their son enough time without rushing him was noted:

P: The length of time [the orthodontic team] need to spend with him obviously is a lot longer than for a normal child. They gotta

have the patience of a saint...I think they know that now, they can't rush him.

Mother of Liam (aged 16)

Liam's Mother explained that he was given ample time to choose a particular coloured band for his dental brace as he found the decision very difficult. Using visuals and simple terms to enhance understanding was apparent:

P: There was a booklet with pictures in, what to expect [after surgery], what they can eat, which was brilliant because Emily's a big sweet eater...we had to cut those out for six weeks, she could see it written, so that was important for her to see.

Mother of Emily (aged 15)

P: They realise she has to have it explained in simple terms.

Mother of Emily (aged 15)

Parents spoke of their appreciation towards HCPs who demonstrated certain activities to their children to encourage self-management (e.g. specific teeth cleaning techniques). HCPs from across disciplines talked about 'tell-show-do' activities (regarded as the "cornerstone of behaviour guidance" (Dean, Avery, & McDonald, 2010, p.299)):

P: What we try to do is a show and tell type activity where if we're going to do something, we try to show them what...we're going to do first, so...if you're going to use a drill, then we will

show it to them, look what the noise it makes and even sometimes use their nail to run it over.

Consultant 2

P: Often what I'll do is take them into the x-ray room and sit...them in the chair and put the apron on them. This is at a speech therapy appointment, so there's nobody else, nothing's going to happen that day, so if they've looked quite nervous, we've gone in and played some games in there.

SLT team member 2

It is useful to compare these innovative and child-centred approaches with the previous HCP asserted preference to give children with IDs GAs, which is arguably easier as it serves HCPs' needs more than the child's.

HCPs, like parents, referred to using visual images to facilitate understanding:

P: Pictures work really well with them [children with IDs] and some children sign a little bit...I can't sign very much but I can do bits and bobs.

Consultant 1

P: We tend to individualise stuff for a child [with IDs]...there was a child coming to a speech investigation clinic and I got our IT guy to take some photos, and sent [them] to the family beforehand

so the Mum could show the child photos of what it would look like when they came.

Psychology team member 2

Concrete examples of individual approaches to treatment are encouraging, although some parents realised that their child may still not understand:

P: [Surgeon] talks to Liam and draws diagrams...not that he totally understands what's going on.

Mother of Liam (aged 16)

Therefore, although there were positives with regard to communication, there was also room for improvement.

The clinic environment was seen as child-friendly (e.g. toys were available) and parents particularly appreciated a family room for overnight hospital stays. It assured privacy, was less stressful and highly preferable to being on an open ward with other families.

Good practice was evident when consultants considered the impact of timing and pace of treatment for children with IDs, depending upon individual needs:

P: Recognising the pace that they're [children with IDs] happy with takes a few appointments sometimes to figure out...sometimes they respond better to...let's get in there and do it very quickly and precisely and then out, so in the minimum amount of time, and then others...prefer a more languid

approach where it's very softly softly and we're doing a little bit here and a little bit there.

Consultant 2

This echoed previous comments about individualised approaches to orthodontic work, including extra time. Deliberately delaying treatment until children with IDs had a better understanding of the treatment plan and risks/benefits was apparent:

P: [Surgery] can happen at a later age...potentially [children with IDs are] going to have the trauma without the understanding of why it's in their interests or good for them...Which is why occasionally things are delayed until it's felt that the child is more a partner in it, rather than this thing being done to them.

SLT team member 2

This quote however contradicted other HCPs' suggestions that surgery sometimes took place without children really understanding what was happening (see theme 'struggles').

Discussion

Struggles, as asserted by children and parents, centred upon stress and distress (specifically GA induction and needles) and the perceived power imbalance at clinic. Stress caused by GA induction was spontaneously reported across participant groups. It is unsurprising therefore that Pilling and Rostron (2014) reported the lack of evidence on best practice in surgery planning for people with IDs.

Children with developmental delay can be anxious and even combative in GA inductions (particularly if the HCP administering the GA is unfamiliar), so an appropriate intervention might be sedation (Tan & Meakin, 2010). However, sedation before GA is not always tolerated by children (McCann & Kain, 2001), so alternatives are welcomed. Changes to routine and hospital settings alongside the challenge of fasting may well increase anxiety (Short & Owen, 2012).

Using physical restraint to anaesthetise older children with IDs was alarming and contrasts with the apparent non-holding approach taken with their neurotypical peers. Page (2015) questioned whether restraining older children is appropriate or even safe to use. Available literature suggests restraint is reserved for young children and that occurrence largely decreases with age (Bray, Snodin, & Carter, 2015). Restraining children has been found internationally (UK, Australia, New Zealand) (Bray et al., 2018) and occurrence was influenced by profession, country, training and availability of guidance (Bray et al., 2018).

Moral and ethical issues of physically restraining children for medical treatment cannot be overlooked. This potential rights violation could be regarded as abuse (Bray et al., 2015). Possible psychological trauma following restraint could include emotional distress, phobias, lack of coping strategies and problematic relationships with HCPs (Brenner, Parahoo & Taggart, 2007). The British Medical Association, the Royal College of Nursing (UK) and the Royal Australasian College of Physicians cite gaining and recording permission to restrain as important. It is unclear whether this happened in the current study.

Some children in this study were very distressed by needles. Negative cycles of fear and needle-related pain can develop in childhood and can spiral (Noel, Chambers, & Petter,

2012), perhaps resulting in heightened pain and anxiety when approached by HCPs, and fainting (McMurtry et al., 2015). Sleeping and eating problems can also occur (Kain et al., 2004).

Managing such difficulties is paramount; fear intensity can lead to children wetting themselves and attempting to escape from HCPs (Kain, Mayes, & Caramico, 1996). Only one HCP in this study (who had worked on hospital wards) referred to alternative techniques to managing GAs for children with IDs (e.g. involving the play team to lessen anxiety). The UK Royal College of Surgeons Clinical Guidelines (2012) stipulate that pre-operative assessments with children, and families should systematically take place to consider GA suitability.

Rapport-building between the anaesthetist and child is important (Short & Owen, 2012). Additional ways of managing GA induction are soft lighting and distraction (Courtman & Mumby, 2008), sensory solutions (e.g. guided imagery and relaxation) (Fung, 2009) in addition to music, computer games and hypnotherapy; restraint should only be considered after exhausting other approaches (Christiansen & Chambers, 2005). These examples pre-date the current research so it is apparent that HCPs in this research were unaware of this good practice. Although not specific to GA, a relevant US study found that behaviour therapy (distraction, exposure therapy, counterconditioning and topical anaesthetic) was successful for eight children with IDs aged 4-16 undergoing needle placements (Slifer et al., 2011). A systematic review into psychological interventions for needle-related pain and distress for children and adolescents (aged 2-19) found evidence for the use of distraction, hypnosis and combined CBT and breathing techniques to reduce needle-induced pain and distress or both (Birnie, Noel, Chambers, Uman, & Parker, 2018). These examples

Intellectual disabilities cleft hospital

demonstrate the availability of different interventions to potentially counteract procedural-related pain and distress.

Input from Certified Child Life Specialists (CCLSs) (US/Canada with similar roles in New Zealand, Australia, South Africa, Japan, the Philippines, Serbia and Kuwait (Association of Child Life Professionals, 2018)) could be beneficial. CCLSs form part of interdisciplinary teams and aim to improve the psychosocial experiences of children in hospital via therapeutic play and psychological support. Such methods are employed to prepare children and families for medical procedures, facilitate coping and pain management techniques, support children to reflect upon previous or imminent experiences, educate the child and their family about health conditions as well as supporting familial involvement in healthcare (American Academy of Paediatrics, 2014). A recent and relevant US study demonstrated the benefits of a CCLS intervention when children (without IDs) underwent intravenous placement; lesser distress levels were reported following CCLS input (Diener et al., 2019). Therefore, the use and evaluation of CCLSs (and similar roles) when supporting children with IDs in hospital could be extremely significant.

There is a UK protocol for preparing children with IDs for theatre and recovery (Blair et al. (2017) which was developed in response to severe distress experienced by several patients with IDs in hospital settings. Protocol development was led by a Consultant Nurse in IDs, in consultation with surgeons, anaesthetists, nurses and healthcare assistants. The acronym 'TEACH' formed the protocol framework; T – take time to work with the child with IDs; E – change the environment (e.g. quiet areas); A – display positive and solution-focused attitudes; C – Communication – find optimum ways to communicate with the child and their family; H – Help – what support does the child and their family need and how can their

needs be met? (Blair et al., 2017). This protocol, in addition to using other resources and approaches already discussed, could make significant and positive changes to the hospital experiences of children with IDs.

“Vulnerability and inequality are nowhere greater than in the surgical setting” (Mouradian, 2006, p.131) and the power imbalance felt by parents in this study echoed previous literature. Power and status hierarchies are the persistent dynamic within healthcare settings in which doctors are perceived to be at the top with their particular knowledge of a particular condition, thus typically dictate appointment agendas (Greenhalgh, Snow, Ryan, Rees, & Salisbury, 2015). Such familial disempowerment however does not indicate successful partnership-working (Henderson, 2003) and could negatively impact autonomy and respect (Goodyear-Smith & Buetow, 2001).

Tensions, as typified by treatment choice and control, lack of HCP training in IDs and assumptions and jargon, were apparent. Children remarked that doctors made the decisions and the deferment of surgical decision-making by parents to HCPs was evident. Reasons for this could include learned passivity and a lack of HCP knowledge in how to facilitate children’s wishes and opinions. Healthcare decision-making is complex, but even if a child is not considered competent in decision-making, they have the right to be heard (Mårtenson & Fägerskiöld, 2008). Involvement facilitates treatment preparation, but an absence of control, and feelings of dependence can result in extreme stress for children (Coyne, 2006). Evidence highlights how children with IDs can express opinions using different tools such as choice cards, a smiley face scale, photos, and/or tick and cross cards (Lewis, 2001). Further, the UNCRC advocates that all children, whether disabled or not, have the right to an opinion

Intellectual disabilities cleft hospital

and that their views should be heard. Information should also be presented to children in appropriate formats (Article 12, UNCRC).

Therefore, individualised approaches to decision-making are fundamental (Bigby, Fyffe, & Ozanne, 2007). There are clear benefits to children being a partner in their care (e.g. feeling listened to and valued) and a further outcome could be satisfaction with clinical outcomes (Kapp-Simon et al., 2015). Deferred decision-making by parents to HCPs has been highlighted elsewhere (e.g. Nelson, Caress, Glenny, & Kirk, 2012). Power delegation to those seen as experts enables trust in HCPs, with decision deferment as doing the “right thing” for their child (p.796). Some parents therefore become “vulnerable to the power imbalance inherent in relationships with practitioners” (Nelson et al., 2012, p.802).

HCPs highlighted information and training gaps in effective communication with children with IDs which was unsurprising as medical training offers scant attention to IDs (Salvador-Carulla & Saxena, 2009). Training however can be hugely beneficial. For example, 100+ medical students who participated in a 3-hour communication skills training session by people with IDs reported increased levels of understanding and ease in communicating with people with IDs (Tracy & Iacono, 2008).

Just one HCP in this research referred to the existence of the hospital Learning (Intellectual) Disability Liaison Nurse. This is a significant untapped resource for the multidisciplinary team who could potentially learn skills and strategies to support their work. LDLN underutilization has been recognised elsewhere (e.g. Barriball, Hicks, Cohen, & Lewry, 2008). Brown et al. (2012) have highlighted positive impacts by LDLNs on education and practice development, as well as being role models and ambassadors for people with IDs.

Intellectual disabilities cleft hospital

LDLNs could provide familial support for self-advocacy or act as advocates for children with IDs (Jenkins & Northway, 2002).

Jargon and assumptions about people with IDs were evident in HCP interviews. In their investigation into stigma in healthcare settings, Aston, Bureau and MacLeod (2014) found that one of three main stereotypes was children with IDs did not understand and/or could not communicate. However, this is untrue; many children with IDs can communicate/ understand if information is accessible (e.g. sign language/photographs). This lack of direct communication could be disempowering and professionals can exclude children to affirm the parent-professional partnership (Dale, 1996). Some HCPs asserted that children with IDs were unable to cope with dental procedures when awake hence GAs were given. Given the trauma reported in this study surrounding GA induction, this assumption should be questioned. International guidelines indicate that GA may be suitable for children with IDs (and other conditions) as they may be unable to tolerate treatment when awake (e.g. Adewale, Morton, & Blayney, 2011; American Academy on Paediatric Dentistry Ad Hoc Committee on Sedation and Anaesthesia, 2008; Forsyth, Seminario, Scott, Ivanova, & Lee, 2012; Sari, Ozmen, Koyuturk, & Tokay, 2014). However there are issues to note; alternatives to GAs should be considered first and guidance emphasises the need for practical alternatives to GA (Royal College of Surgeons, England, 2012). They are not desirable in view of patient burden and costs and there are additional safety aspects and complications such as a swollen tongue/lips and nasal bleeding (Eshghi, Samani, Najafi, & Hajiahmadi, 2012). Therefore it is imperative that GA substitutes are carefully considered in partnership with children and families.

The final theme of good practice denoted two subthemes, tailored treatment and appropriate information/communication. Parents appreciated the respect and help shown by HCPs to their child, and a range of tailored treatment was discussed, for example, offering a young person with ID extra time to choose a coloured band for his orthodontic brace. This finding resonated with Oulton's et al. (2015) research which emphasised the importance of "the little things" (p.78) as fundamental to improving hospital experiences for children with IDs.

Using visuals and 'tell-show-do' activities as described by parents and HCPs were valued and enhanced understanding. Such approaches were useful but HCPs suggested their techniques were not based on policy, training or information but based on 'common sense' and 'goodwill'. Encouraging staff to identify their training needs for working with people with IDs is important and should be supported by managers (Sowney & Barr, 2004). Chew, Iacono and Tracy (2009) posited recommendations for HCPs working with people with IDs such as communicating directly, checking understanding and offering optimum time for appointments. Sowney and Barr (2004) suggested that alternative communication formats (e.g. Makaton) should be learnt by HCPs.

An awareness of individualising treatment, pace and timing for children with IDs undergoing orthodontic treatment was another good practice example in this study highlighted by HCPs and parents. How orthodontists adapt their approach to the needs of children with IDs is rarely featured in research. Musich (2006) referred to technological improvements which could benefit people with IDs such as quick-setting materials and improved flavours for dental impressions, alongside types of brace-wires which can reduce the amount of appointments needed. Hobson, Nunn and Cozma (2005) emphasised that dental treatment

was feasible for disabled children, including those with IDs, but careful planning, and ongoing assessment/evaluation with the child was fundamental. Again, this approach contrasted with the reference to GAs by some HCPs in this study.

Given the current findings, ID practitioners in the field have a key role in supporting children with IDs receiving healthcare (and by extension, adults receiving treatment). They could act as advocates for people with IDs prior to surgery to facilitate good practice. Working in partnership with parents/carers to request that certain treatment/surgical approaches are utilised as opposed to potentially burdensome approaches as highlighted here, may yield meaningful changes to practice. Practitioners are well-placed to alert HCPs to the existence and role of LDLNs (UK) and Child Life Specialists (UK/Canada) and Child Life Therapists (Australia) and the 'TEACH' protocol (Blair et al., 2017). In conjunction with families, they could assist in providing accessible information and much-needed training about IDs, and accessibility in healthcare.

Limitations

The research was cross-sectional due to PhD time constraints and limited resources. Longitudinal research to elicit changes with age could prove fruitful. The study focused on children with mild-moderate IDs who could verbally contribute and therefore excluded children with severe IDs which can be criticised for resulting in a skewed sample (Cambridge & Forrester-Jones, 2003). It is emphasised however that this qualitative exploration is a precursor to further research in which the intention is to include those with severe IDs. The study sample was drawn from one UK cleft centre so caution is needed in the application of findings, but again, this research is a starting point. Cleft team staff acted as gatekeepers by making contact with eligible families; some families who may have wanted to participate did

not have that opportunity. Participant self-selection is a limitation given previous research which suggests that participants often volunteer having had very good or very poor experiences (Peel, Parry, Douglas, & Lawton 2006). HCPs also self-selected so were perhaps more likely to already be demonstrating good practice. Only one child participant was non-British whilst all other participants were White British; it is unknown if and what different responses might be given by those with different ethnic backgrounds. Study findings must be considered within these parameters.

Conclusion

The current research demonstrates that although there is good practice within a specialist hospital clinic, it is seemingly ad-hoc and much more needs to be done to work with children with IDs and their families to individualise treatment and communication.

Ascertaining children's views on treatment using accessible formats is fundamental. Finally, findings regarding the trauma around GA induction and the use of needles for children with IDs were alarming and unsolicited, therefore warrant further research and understanding at the earliest opportunity.

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Fig. 1: Example UK care pathway for children with clefts (Guy's and St. Thomas' NHS Foundation Trust, 2016)

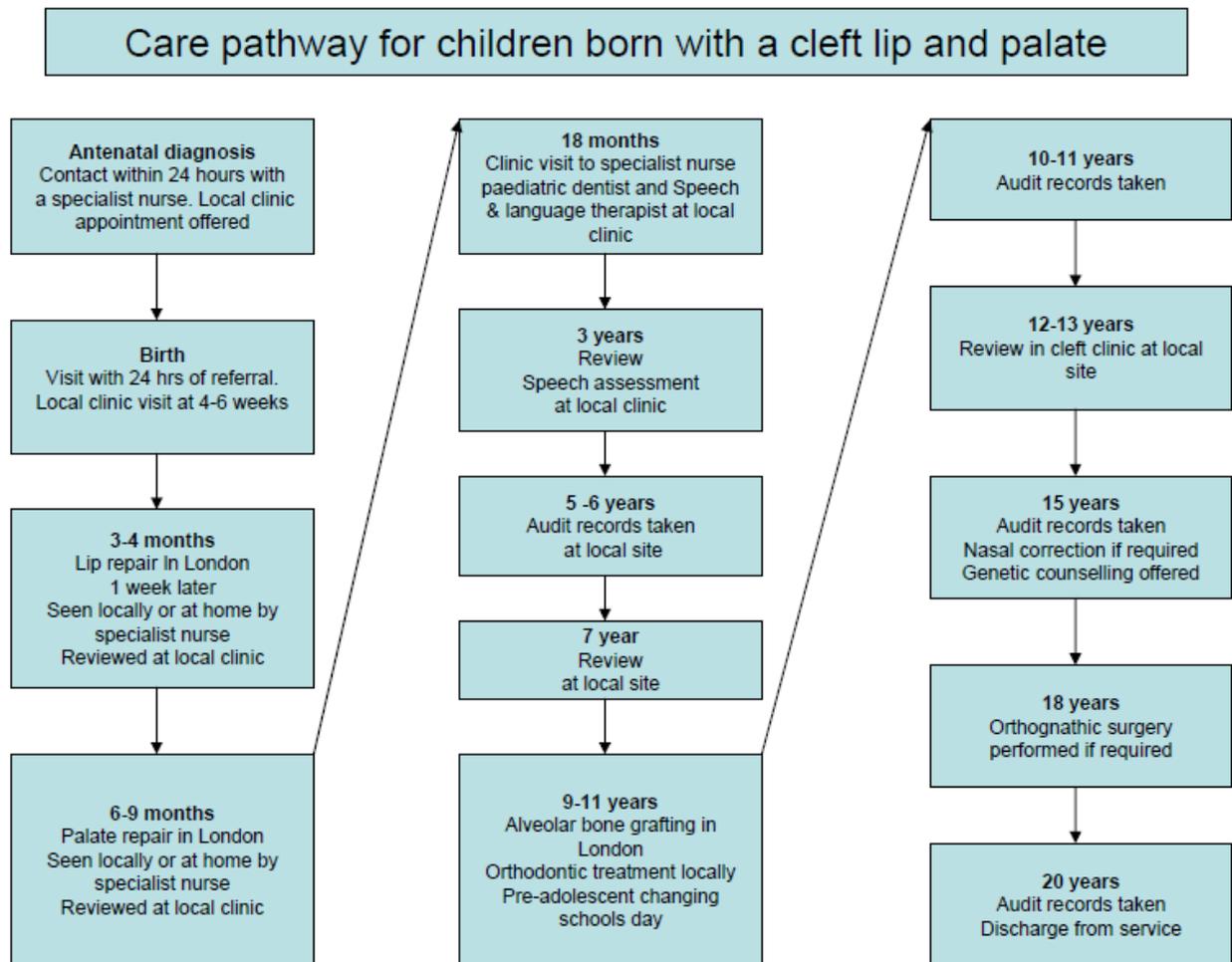


Table 1: Participant characteristics of children

| Participant number | Male/ female (M/F) | Age | Mainstream/ SEND school (M/S) | EHCP (Y/N) |
|---------------------------|-----------------------------------|------------|--|--|
| 1 | M | 11 | S | Y |
| 2 | M | 11 | S | Y |
| 3 | F | 14 | M | Y |
| 4 | F | 15 | M | Pending at the time of interview |
| 5 | M | 16 | S | Y |

Table 2: Participant characteristics of parents

| Parent of child with: | Mother | Father | Joint (Mother & Father) | Total |
|----------------------------------|---------------|---------------|--|--------------|
| LDs | 3 | - | 3 | 6 |
| Non-LD | 3 | 1 | - | 4 |
| ALNs | 5 | - | - | 5 |
| Total | 11 | 1 | 3 | 15 |

Table 3: Healthcare professional participant characteristics

| Specialism | Total |
|------------------------------------|--------------|
| Consultant (surgical/orthodontics) | 3 |
| Psychology | 2 |
| Clinical nurse specialist | 2 |
| Speech and language therapist | 2 |
| Total | 9 |