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Grappling with uncertainty - experiences of parents of infants following perinatal stroke

Abstract

Background

The term perinatal stroke describes focal damage to the developing brain due to cerebrovascular disease and occurring either before or shortly after birth. Aetiology, presentation and evolution differ from stroke in adults.

Aims

We aimed to explore early parental experiences related to having a child with perinatal stroke, including how parental psychological wellbeing had been impacted, to consider how support for families could be improved.

Methods and Procedures

We undertook a qualitative research study, using in-depth interviews of parents of infants with perinatal stroke when the infants were 5-6 months corrected gestational age. Sixteen parents (11 female, 5 male) of 11 infants with perinatal stroke took part. Thematic analysis was used in data interpretation.

Outcomes and Results

Parents described distress related to the lack of information regarding likely outcome following perinatal stroke, as well as confusion around the term 'stroke'. Guilt and self-blame were expressed, with increased emotional sensitivity. Seeking information about stroke to reduce uncertainty was a useful strategy for some, but overwhelming for others.

Conclusions and Implications

The diagnosis of perinatal stroke led to psychological distress in parents. Uncertainty following diagnosis produced significant emotional difficulties. Recommendations for practice include providing timely, paced information and psychological support.

What this paper adds?

Parents hearing the term 'stroke' in relation to their infant become confused and distressed as they can only relate it to knowledge about risk factors, presentation and outcomes in the adult context.

Uncertainty around outcome after stroke is a major source of emotional distress.

Keywords

Perinatal stroke, Unilateral cerebral palsy, Psychological wellbeing, Parent, Infant, Diagnosis, Communication, Uncertainty

Data availability statement

The data that support the findings of this study are available on request from the corresponding author. The data are not publicly available due to privacy or ethical restrictions.

1. Introduction

Perinatal stroke (PS) describes focal damage to the developing brain due to cerebrovascular disease occurring either shortly after birth (usually in the first week) or before the infant is born. The term perinatal stroke encompasses a number of different entities including neonatal arterial ischaemic stroke (NAIS), neonatal cerebrovenous sinus thrombosis (NCVST), haemorrhagic periventricular venous infarction (PVI) and haemorrhagic stroke (Fluss, Dinomais, & Chabrier, 2019). PVI most frequently occurs in preterm infants, usually in conjunction with extensive (grade IV) germinal matrix haemorrhage. NAIS is more frequent in term infants.

Perinatal stroke is the most common cause of hemiparetic (unilateral) cerebral palsy: it can also lead to intellectual disability, developmental and behavioural disorders and epilepsy (Dunbar & Kirton, 2018). One of the challenges faced by families of infants with perinatal stroke is the gradual emergence of consequences of the initial lesion over time. At the time of the initial stroke, some infants may be completely asymptomatic, whereas others may present with seizures or (particularly in the case of NCVST) may show signs of poor feeding and excessive sleepiness. Yet other infants (those with PVI) may be diagnosed through routine cranial ultrasound protocols for preterm infants; often this group have multiple comorbidities related to prematurity. Those infants who were asymptomatic may then show emerging signs of movement difficulties on one side of the body from around 3-4 months of age, as voluntary reaching and grasping develop, and delayed motor milestones, whilst usually being otherwise medically well – investigations in this group then lead to discovery of an infarct or haemorrhage which was presumed to have occurred in the perinatal period, labelled as “presumed perinatal stroke” (PPS) in this group (Lee et al., 2005). Behavioural problems, intellectual disability or other issues may become apparent later still (Kirton & Deveber, 2013) and are difficult to predict. Thus, carers of infants with PS can be left in a state of uncertainty because of delays in diagnosis due to late-emerging signs (PPS); but they can also be left uncertain about the potential severity of their child’s condition even after an early diagnosis.

Uncertainty is known to be associated with an increased subjective sense of disease severity, reduced optimism and reduced perceived control in carers of children with undiagnosed conditions (Madeo, O'Brien, Bernhardt, & Biesecker, 2012). Uncertainty regarding likely outcome

even after diagnosis could have similar adverse associations, with high levels of ongoing psychological distress, as has been reported in parents of children with other long-term conditions (Carpentier, Mullins, Chaney, & Wagner, 2006).

The diagnosis of any disorder is inherently distressing to those caring for the affected person. Carers of those with neurological disability also experience reduced wellbeing and increased stress in line with what has been described as the 'burden of care' (Özkan Tuncay & Kars Fertelli, 2019). *The impact of childhood stroke (outside the neonatal period) on parents has been captured through qualitative interviews undertaken at least one year after the stroke (Soufi, Chabrier, Bertolletti, Laporte, & Darteyre, 2017). Parents described the anxiety, guilt and fear regarding outcome that they experienced at the time of diagnosis, but also more chronic challenges to family functioning and mental health. Another study showed that mothers of children with moderate to severe perinatal stroke had poorer outcomes than controls on measures of depression, marital satisfaction, quality of life, and family functioning. Fathers suffered similarly but mothers had higher levels of guilt and anxiety (Bemister, Brooks, Dyck, & Kirton, 2014, 2015). Parental wellbeing is important in its own right; but also impacts the infant, including attachment, emotions, behaviours and ultimately child development (Smith, 2004). Despite this, there has been little research into the most effective methods of providing psychological support for parents of infants with emerging cerebral palsy (Irwin, Jesmont, & Basu, 2019).*

As described above, PPS is rather a different situation clinically, diagnostically and perhaps also emotionally from acute symptomatic stroke in a neonate (e.g. NAIS), or indeed from PVI diagnosed in an (often unwell) preterm infant. Our study focusses on parents of infants who were recently symptomatic with NAIS or with complications of prematurity including PVI.

We undertook qualitative interviews of parents of infants with perinatal stroke when the infants were around 5-6 months corrected age. During the interviews we explored how parental psychological wellbeing had been impacted by the stroke, to consider how support for families could be improved *in the early weeks and months after such a diagnosis.*

2. Materials and Methods

Participants were parents of 11 infants living in the North East of England who had sustained a perinatal stroke and who took part in a feasibility trial of a parent-delivered therapy intervention (Basu et al., 2018). **Term or preterm infants who sustained a predominantly unilateral stroke (arterial ischaemic, haemorrhagic or haemorrhagic periventricular venous infarction) demonstrated on cranial imaging and identified within the first 3 months of life were eligible to take part in the trial.** The study was approved by West of Scotland Research Ethics Committee, reference number 15/WS/0129.

2.1 Recruitment and data collection

Between August 2015 and January 2017, clinical staff at participating centres and sites identified and approached parents/carers of potential infant participants, providing flyers and information sheets. These materials were developed with the involvement of a parent of a young child with UCP. With parental consent, contact details were forwarded to a member of the research team, and the infants were then screened for eligibility to participate in the trial. If eligible, written informed consent was obtained, with parents providing proxy consent for infants. Parents/carers of infants in the trial were recruited as participants, so we could explore their experiences regarding the process. We invited mothers, fathers, and grandparents to take part if actively involved in the infant's care on a regular basis and allowed more than one such carer to participate per infant. The trial included in-depth interviewing of parents in the last month of the intervention period, when the infants were 5-6 months corrected gestational age.

The interview topic guide included a focus on parental psychological wellbeing and potential need for psychological support, as this had been raised as an area of high importance for parents in our participatory design work leading up to the feasibility trial (Basu, Pearse, Baggaley, Watson, & Rapley, 2017). Parents were interviewed at their homes, at a time convenient for them. Interviews lasted between 1-2 hours each.

2.2 Data analysis

Standard procedures of rigorous qualitative analysis were followed (Rapley, 2010). We used procedures from first-generation grounded theory (coding, constant comparison, memoing) (Glaser, 1965), from analytic induction (deviant case analysis)(Seale, 1999) and from constructionist grounded theory (mapping)(Charmaz, 2006).

Interviews were audio recorded, transcribed verbatim then anonymized. The anonymized transcripts formed the dataset for this analysis. This firstly involved becoming familiarised with the data by reading through transcripts and annotating relevant material. Using the software NVIVO, the relevant extracts were then organised systematically, to generate initial codes. The use of thematic maps then allowed for the initial development of preliminary themes, from these codes, that capture significant meaning within the data. Preliminary themes subsequently went through a thorough process of reviewing, modifying and developing, to ensure that identified themes were separate in their representation of a specific meaning, and were well supported by their associated data extracts. This produced the final main themes (with sub-themes) and defined to form the basis for analysis. Independent cross-checking was undertaken, and findings analysed and discussed as a team to ensure that identified themes were clear and well-supported by the underlying data.

3. Results

Eleven families with an infant with perinatal stroke took part in in-depth interviews (six with only the mother; five with both parents; sixteen interviewees in total). Table 1 summarises the demographic information regarding participants. **One infant, with symptom onset at 5 weeks, had AIS secondary to vasospasm following an intracerebral haemorrhage (ICH) with subarachnoid haemorrhage (SAH); all other infants had either arterial ischaemic stroke (AIS) or periventricular venous infarction (PVI). All the infants with PVI were premature. There were no infants with cerebral venous sinus thrombosis in the study. No infants in our study fell within the presumed perinatal stroke category, as all were recruited to a study of early therapy intervention which required a radiological diagnosis within the first three months of life.**

The diagnostic setting was the neonatal intensive care unit (NICU) in all but two cases. The infant with AIS secondary to ICH and SAH presented acutely through the Emergency department

and was initially reviewed by the paediatric and neurosurgical teams, then promptly referred to paediatric neurology. The infant with seizures following decannulation after extracorporeal membrane oxygenation (ECMO) was diagnosed with AIS in the paediatric intensive care unit and discussed with a paediatric neurologist.

All families discussed stressors and situations they had found difficult to deal with because of their infant's stroke. As we explored the data, we identified uncertainty as a major factor underlying adverse experiences for this group. Our analysis first covers the experience of uncertainty, then the impact of uncertainty on the family, and finally strategies used to manage uncertainty. The thematic coding structure is summarised in Figure 1.

3.1 Experience of Uncertainty

Parents described a lack of clarity in discussions with healthcare professionals regarding the likely severity of any future impairment which their child might have because of the stroke which they had suffered. They had a range of **unanswered** questions: ~~that could not be answered~~.

'cause I says, like, "Will he walk? Like, will he do this?" Like, I want him to do these things. It's like this tiny little baby, like, lying there like his future, like that's what's in the balance.
(Sheila, mother of James)

They became aware they would have to wait for an unspecified length of time for symptoms to become apparent and **that the likely** outcome was **not yet** clear:

He (health professional) said, "It's not 100% sure, what she'd be able to do." Erm, and then he said, 'But, she might be perfectly fine, we just don't know'.
(Larry, father of Grace)

This uncertainty was compounded by difficulties in taking in and retaining information around the time of diagnosis. The enormity of the situation for parents, that they 'couldn't believe'

what they were being told often meant that they could only manage so much information at one time. The information breached all the expected norms of becoming a parent. The pace of events with which they were required to keep up to date added an additional layer of challenge:

I mean, to be honest, like, they were coming at us with stuff like, always coming onto the ward and telling us stuff and like were like, really like black and white about it, like they didn't like sugar coat things up to me, they were sort of like, "Basically your baby could die in 72 hours." And I was like, "Right."

(Quinn, mother of Ian)

The use of ~~some~~ medical terminology **was challenging for parents, who whilst they wanted to understand their child's condition, would feel overwhelmed by jargon and sometimes fear the worst when it was used:**

Science, science scares me, it always has. Erm, as much as I would, you see, I'm one of these people, I, I would really like to know why it happened. But then, if somebody came and went, "Well, because of this and that and the other," and threw big, long, scientific words at us, I'd be, like, "So you're telling us that my child's dying."

(Imogen, mother of Evan)

The word "stroke" was not only frightening, with one father describing it as 'the worst word I ever heard' – but also particularly confusing. A father outlined the tension, (Barry, father of Bobby) – 'Stroke to me depicts total loss of movement and yeah, I think it emphasises the disability'. Parents had some familiarity with stroke in adults but struggled to relate this term to an event that could affect their newborn infant and the information they were being given about the future. Many were previously unaware - 'Nobody...knew that babies could have strokes' (Una, mother of Kelly). The lack of public awareness of neonatal stroke and the differences in presentation from adult stroke are likely to have contributed to this problem.

3.2 Impact

Parents, especially mothers, expressed feelings of guilt and personal blame, even when they were aware the stroke was not their fault:

You think about, like, and I mean you do beat yourself up emotionally, and I was like, 'Is it your [my] fault that this happened?

(Sheila, mother of James)

Given the uncertainty about the exact aetiology of the stroke in many cases, there remains a void to be filled with an explanation, to the extent that parents cause themselves to believe that they are responsible for the event. Parents reported ruminating on disparities in apparent or potential abilities of their child, in comparison to both typically developing infants and other infants with stroke, whom they had usually come across in hospital settings:

"Is this why he's like this?" There was a lot of that and wondering, you know, "Is my baby acting like any, you know, other child, er, you know, of his age or his corrected age?"

(Claire, mother of Bobby)

Failure to reach certain developmental milestones by infants caused parents significant anxiety. It was particularly confusing for parents of preterm infants as they tried to understand whether any apparent delay in development was outwith that expected due to prematurity alone, or if any unusual behaviours could be attributed to the stroke. Hypervigilance regarding the infant's behaviours and development, and constant worry about the outcome, sometimes led to reduced enjoyment of interactions with the infant by the parent:

I was like getting up, and I was like, “I cannot enjoy him when he’s had all that bad time in the neonatal, and I feel like I’m watching his every movement” ... cause I, I was waiting for something to, to not work properly or something bad to happen”.

(Sheila, mother of James)

This was in addition to descriptions of limited time for play activities because of the burden of hospital appointments and care for the infant. The stress of the situation led to parents behaving out of character. Here, a parent reports on a conversation she had with her partner:

He says, “you were just a different person,.....~~and everything, everything, kind of, came into focus for you, and you just started ... Everything, kind of, you just came back to being (wife).~~” And, you know, “you did, you did certain things,” he says, “and you would shout at us for no reason,” ~~and, you know, all, all the little things came back. And he says, “I knew that we were all going to be all right.”~~

(Imogen, mother of Evan)

This clearly impacted on the family dynamic and parents reported ongoing emotional sensitivity, with one father describing his partner getting emotional, ‘more hourly than weekly’.

3.3 Managing uncertainty

Some parents actively and independently sought out information about their child’s condition to address their concerns and to help them prepare for the future. This could enable them to feel more in control of the situation through acquisition of knowledge. However, this was not always the case:

Totally frightened myself. I read the NICE guidelines, knowledge is power but in a lot of the times now the mummy knowledge isn’t power and sometimes, sometimes it’s nice to be a novice and not understand things.

(Claire, mother of Bobby)

Thus, accessing the information could also lead to a sense of being overwhelmed and distressed, or guilt that parents were not doing enough to help the situation. Other parents were initially unwilling to explore the implications of the diagnosis in detail but felt more empowered to seek information (including information from professionals) over time.

Until now I never wanted to ask, "What does this mean?" because I probably haven't been in the right place, whereas now I think when we go to his next consultant appointment with (doctor) I will feel comfortable saying, "Will you just explain what this means to me?"
(Emma, mother of Cameron)

Differences between individual preferences for information and changes in these preferences over time were particularly evident in relation to the provision of 'Real Life Stories' to parents. Provided within the therapy manual supplied to parents in the feasibility trial, these aimed to alleviate distress and uncertainty by sharing stories from affected children and their families to show active participation despite physical disability. Some parents were reassured and comforted by this information. It enabled them to become aware that 'it's not just me that's going through it. It's other, other people. It's – I'm not alone in the world' (Alana, mother of Adam). Some sought out other parents with children affected by the same condition e.g. through social media, outlining that 'it's been quite good just to read other people's stories and to find out how they're coping' (Geraldine, mother of David). However, others were not ready to envisage potential future outcomes in such detail.

Parents expressed the relief they experienced when professionals pointed out signs of progress in their infants, and the sense of comfort where they had ready access to a healthcare professional whom they trusted to 'keep an eye on' their infant. There was also a level of acceptance of uncertainty.

I mean I'd love someone to tell us what she's gonna be like in five years' time, but nobody can tell you what your child's gonna be like in five years' time, regardless of what they're like, so...

(Marion, mother of Grace).

There was a real sense of resilience and determination to deal with whatever the future would hold, that 'obviously you've just got to be strong for them, really' (Olivia, mother of Henriette).

4 Discussion

In this analysis we aimed to explore how parental psychological wellbeing had been impacted by having an infant with perinatal stroke. We were struck by how much of the difficulties faced arose from uncertainty surrounding the diagnosis and its outcomes. The impact of uncertainty on those with illness and their carers has been studied in other settings (Mishel & Braden, 1988). By mapping perinatal stroke on to Mishel's model (Figure 2), it becomes clear that there are specific features of this condition that drive high levels of uncertainty. Unfamiliarity with the condition (on the part of parents but, notably, also some healthcare professionals), combined with a high level of variability in presentation and gradual evolution of signs of the condition over many months, make it difficult for healthcare professionals to give a clear statement to parents regarding what will happen. Lack of clear information and high levels of uncertainty contribute to an appraisal of the situation as dangerous and a threat to the infant's future. There may be maladaptive coping responses with a strong sense of guilt (driven by lack of understanding of why the stroke has occurred and perhaps worsened by knowledge of modifiable risk factors for stroke in adults), and negative impact on family function. Parents may mobilise strategies such as information seeking, professional support and comparison to others, and gradually adjust to the situation over time as with other conditions. Thus, it is particularly in the early months that uncertainty in perinatal stroke is a significant factor. The process of mapping out these factors

assists in identifying approaches to reduce uncertainty and support parental coping strategies. These are discussed below.

4.1 Information management

Some of the parents interviewed felt that healthcare professionals left them feeling uncertain about their infant's prognosis, through guarded information provision. Furthermore, they were confused and sometimes distressed by some of the medical terminology used, including the word 'stroke', which they associated with the condition in adults. Yet other parents felt overwhelmed, anxious and not ready for information provided in lay terms from parents of children with perinatal stroke aimed at providing some guidance about what the future may hold. Parents who experience greater uncertainty feel in less control of their child's medical condition, which may lead to less effective coping and poorer adaptation (Madeo et al., 2012). However, providing information without the opportunity to actively discuss concerns and management strategies can lead to ongoing unhelpful rumination. Whilst some parents were proactive in seeking information from healthcare professionals and other sources, other parents may have been less experienced in information seeking and interpretation and might have valued more guidance in this process. Recent literature acknowledges the challenge for parents in identifying and asking important questions: one approach is to use a checklist as a framework to prompt questions which might otherwise remain unaddressed (Lemmon et al., 2019).

The literature describes information management as superior to information provision in health contexts: information management factors in the state of readiness and willingness of the patient and family to receive each piece of information, and their goals and expectations (Brashers, Goldsmith, & Hsieh, 2002). Given the propensity for the information around diagnosis to cause distress, it is surprising that psychological support for parents is not made more routinely available.

4.2 Uncertainty management

A critical issue regarding information provision around outcome after perinatal stroke, and a major difference from stroke in adults, is the lack of specific knowledge by healthcare

professionals. Only a proportion of infants develop cerebral palsy following perinatal stroke, and many healthcare professionals adopt a 'watch and wait approach', making a diagnosis of cerebral palsy once clear signs of motor features of this condition have appeared. This approach prolongs the period of uncertainty regarding motor outcome unnecessarily for some families, when in fact early magnetic resonance imaging of the brain can accurately predict which infants will develop cerebral palsy (Boardman et al., 2005; de Vries, van Haastert, Benders, & Groenendaal, 2011), though the severity of motor impairment can not be accurately specified from this imaging. However, longitudinal studies show that the vast majority of children will be able to walk and talk following neonatal AIS (100% in a 7 year follow up study of 100 term newborns)(Chabrier et al., 2016). Parents should not be left wondering whether their child will be able to walk, yet this was a question raised by one of the parents we interviewed and is recognized as a question to which many parents want an answer. It should be acknowledged that uncertainty often remains regarding the severity of cerebral palsy and of comorbidities such as epilepsy, autism, cognitive and behavioural difficulties which may occur (Chabrier et al., 2016; Lõo et al., 2018). Healthcare professionals have a duty to minimize their own uncertainty through acquiring knowledge, and to be comfortable with handling and communicating the remaining uncertainty. The field of research into early diagnosis and early intervention is expanding rapidly, with a growing evidence base supporting certain targeted approaches to improve motor outcomes after early acquired brain injury such as perinatal stroke (Basu, 2014; Basu & Clowry, 2015; Hadders-Algra, 2021; Novak, Morgan, & Adde, 2017). Where a healthcare professional may be less familiar with a particular condition such as stroke, it is appropriate to refer to another specialist such as a neurologist or even neonatal neurologist who can provide more information to families, thus reducing unnecessary uncertainty.

Research into other chronic illness diagnoses suggest patient uncertainty is not addressed or communicated well by healthcare teams. In another qualitative study, participants reported doctors would often give leaflets with information about the illness, but not address any uncertainties or go over any management strategies. Where there was involvement of a psychologist in the team, these difficulties were lessened. Patients wanted active communication with healthcare providers

in managing their uncertainty, in terms of appraisal, providing coping strategies and monitoring symptoms of distress (Brown, 2016). However, healthcare providers felt they lacked relevant skills and resources to communicate uncertainty with patients.

A recent study explicitly explored clinician views regarding prognostic uncertainty in the setting of neonatal hypoxic-ischaemic encephalopathy (HIE)(Rasmussen, Cascio, Ferrand, Shevell, & Racine, 2019). Clinicians were acutely aware of the sources of uncertainty in this context, and refreshingly, sought ways to communicate this explicitly to families.

Management of uncertainty is an essential skill which should be included in educational programmes for healthcare professionals (O'Riordan et al., 2011). This would enable healthcare professionals to embrace the cognitive, emotional and ethical dimensions of handling uncertainty in diagnosis, prognosis and shared decision making (Alam et al., 2017).

4.3 Impact on Emotional adjustment

Increased levels of uncertainty about aspects of an illness at diagnosis (e.g. confusion due to the complexity of the situation, or lack of clarity about the likely outcome) are predictive of greater psychological distress and longer term adjustment outcomes, above and beyond the variance accounted for by child illness duration and parent psychological distress, ~~at the time of diagnosis~~ (Carpentier et al., 2006). As a specific example of this in perinatal stroke, a sense of guilt and self-blame was apparent from our study: parents (especially mothers) were left wondering whether the stroke was their fault.

Heightened guilt and self-blame have been reported previously in parents of infants with perinatal stroke, and found to be associated in the longer term with caregiver depression and worsened family functioning after controlling for child variables (Bemister et al., 2015). It is possible that families draw parallels with stroke in adults and wonder whether factors relating to the mother's lifestyle during pregnancy could have led to the stroke. Importantly, this problem is at least partly remediable by stressing to families that the stroke has not occurred through any fault of their own.

Parents also described being in a state of hypervigilance following the diagnosis, as well as experiencing resurgence of negative emotions with setbacks in overall progress. Chronic sorrow theory describes the long-term sadness experienced due to the continual losses that come with a diagnosis such as perinatal stroke (Lindgren, Burke, Hainsworth, & Eakes, 1992). Even years after the diagnosis, parents may experience intensified chronic sorrow symptoms following a triggering event such as new illness or failing to reach expected developmental milestones, highlighting the long term process of adjusting to the diagnosis (Coughlin & Sethares, 2017; Whittingham, Wee, Sanders, & Boyd, 2013). This is in line with other research showing a negative impact of the child's condition on parental health and wellbeing (Ha, Hong, Seltzer, & Greenberg, 2008).

Professional support was highly valued by parents in our study in terms of providing reassurance and advice. Whilst parents sometimes needed guiding to sources of professional support for themselves, their emotional state was greatly helped by receiving direct support for their infant's condition. The sense of vulnerability in parents and the reliance on the competence of healthcare professionals, combined with a sense of ambivalence and difficulty trusting that their child's needs will be understood and met by others, have been well summarized elsewhere (Iversen, Graue, & Råheim, 2013). Healthcare professionals need to be aware of these feelings in parents to enhance their understanding and support.

4.4 Strengths and Limitations

Our study was unique in its focus on the early months following diagnosis of perinatal stroke, a condition which still provokes confusion about the terminology, high levels of uncertainty around outcome, and in which sequelae of the stroke emerge gradually over time, in contrast to the situation with stroke in adults. At this early stage parents were still adjusting to the diagnosis **and were reflecting on very recent events. Our paper complements a recent publication (Leal Martins, Chabrier, & Fluss, 2021) which interviewed parents 2-10 years after their infant's stroke, capturing more matured reflections and demonstrating ongoing parental emotional disturbance following those early events. Our paper also differs from Leal Martins' perspective (which focused on NAIS), in having a more heterogeneous set of stroke aetiologies including preterm infants with PVI.**

One limitation of our study is that fewer fathers than mothers contributed to the interviews. Fathers also contributed less of the overall perspective, perhaps because the primary caregiving role was often through the mother for practical reasons: working fathers in this study were all working their usual hours by the time of the interviews. It is evident that fathers are not immune to the psychological demands of caring for a child with disability and it is crucial to develop a more thorough understanding of fathers' psychological outcomes, as well as the impact on other carers and other family members such as siblings.

We acknowledge that many factors other than uncertainty undoubtedly contribute to parental distress after a diagnosis of perinatal stroke – these factors include the traumatic nature of the delivery in some cases; the quality of communication about the diagnosis (Leal Martins et al., 2021), and the impact of other traumatic medical events associated with prematurity or the onset of the stroke. Given the life-threatening nature of some of these events, it is perhaps surprising that parents did not dwell more on the uncertainty regarding whether their infant would live or die. Another qualitative study of parent experiences in the neonatal unit (not specific to perinatal stroke) captured a parental viewpoint that quality of life was a greater concern than survival (Harvey et al., 2013): this however does not negate the extreme stress of being the parent of an infant with life-threatening illness.

4.5 Implications for future research and practice

Our study has implications for optimal clinical practice in managing perinatal stroke. Firstly, reduction of uncertainty regarding motor outcomes on the part of the healthcare professional could be achieved through education. Secondly, education of healthcare professionals in communicating and handling uncertainty would be of benefit. Thirdly, it is valuable for professionals to understand the psychological stressors faced by parents of infants with perinatal stroke and to help reduce these where possible. Clear communication, in particular stressing that parents are not to blame for the stroke, is critical. An ethical framework for improving communication and decision-making in

partnership with parents in the context of neonatal neurological injury was published in 2016 (Racine et al., 2017). This framework identifies specific areas for personal reflection, and merits formal evaluation.

Finally, increased availability of psychological support for families of infants with perinatal stroke and other conditions with similar threats to parental wellbeing would be of great value.

Table 1: Participant details

Legend

*One infant had a sibling who died. In respect for confidentiality, we have not added this to the table but in memory and respect for the infant and family we mention this here.

Pseudonyms have been used.

Abbreviations

AIS: Arterial ischaemic stroke. APH: Antepartum haemorrhage. CDH: Congenital diaphragmatic hernia. CPR: Cardiopulmonary resuscitation. CRUSS: Cranial ultrasound. CTG: Cardiotocogram. EmCS: Emergency Caesarian section. HDN: Haemorrhagic disease of the newborn. HFOV: High frequency oscillatory ventilation. ICH: Intracranial haemorrhage. IUGR: Intrauterine growth retardation. NICU: Neonatal intensive care unit. NVD: Normal vaginal delivery. PHI: Pregnancy-induced hypertension. PVI: Periventricular venous infarction. SAH: Subarachnoid haemorrhage.

Figure 1: Thematic coding structure

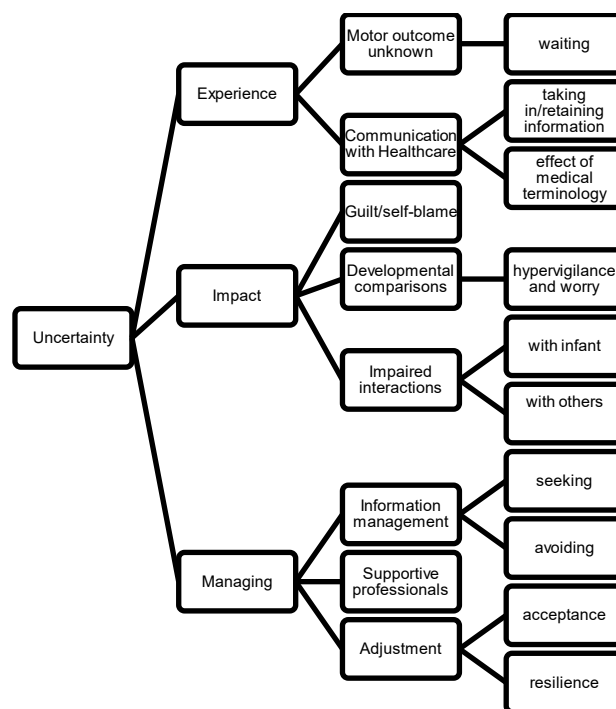
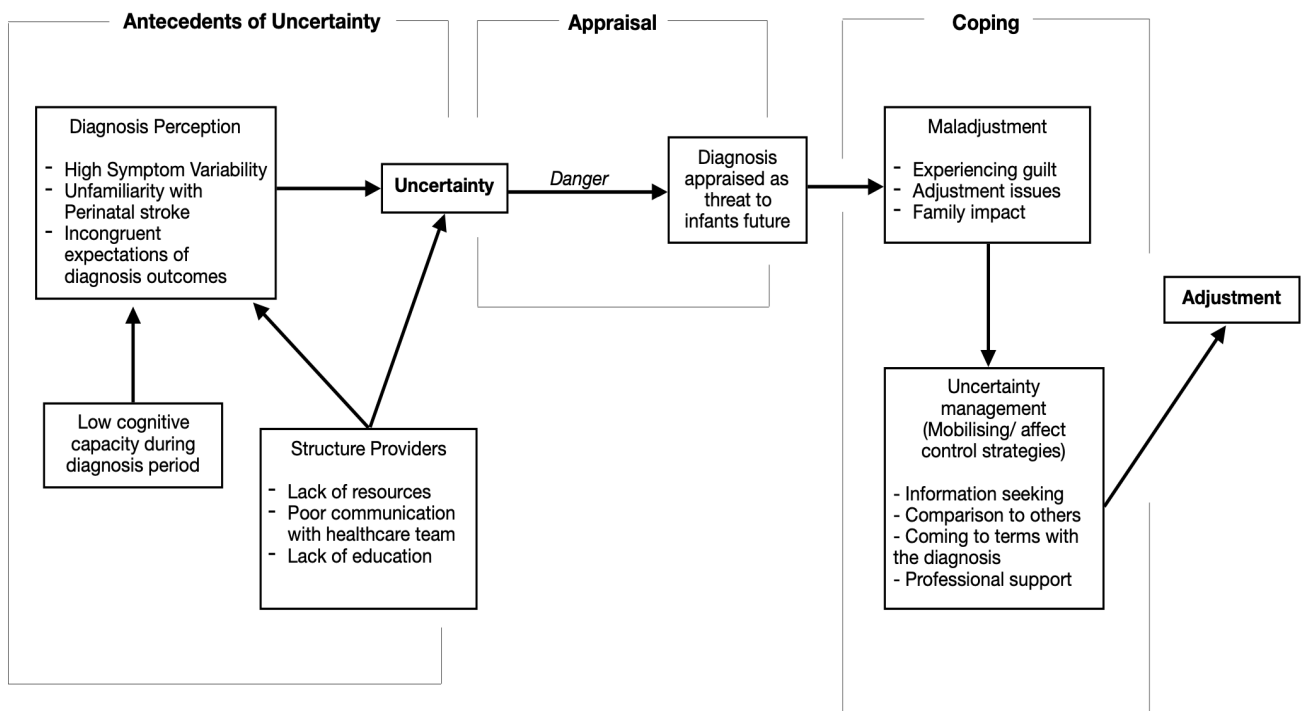


Figure 2: Mapping perinatal stroke to Mishel’s Uncertainty in Illness Model



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