

# Parental Experience of Sleep-Disordered Breathing in Infants With Cleft Palate: Comparing Parental and Clinical Priorities

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## Abstract

**Objective:** To identify outcomes relating to sleep-disordered breathing (SDB) that are relevant to parents, during the early weeks of caring for infants with cleft palate (CP), and compare these with clinical outcomes identified in a systematic search of research literature.

**Design:** A qualitative study using telephone/face-to-face interviews with parents explored their understanding of breathing and respiratory effort in infants with CP.

**Setting:** Care provided by 3 specialist cleft centers in the United Kingdom, with study conducted in parents' homes.

**Participants:** Criteria for participation were parents of infants with isolated CP aged 12 to 16 weeks. Thirty-one parents of infants with CP (over 12 weeks) were invited to participate in the interview. Interviews were completed with 27 parents; 4 parents could not be contacted after completing the sleep monitoring.

**Results:** Parents' description of infants' sleep suggests that breathing is not considered as a separate priority from their principal concerns of feeding and sleeping. They observe indicators of infants' breathing, but these are not perceived as signs of SDB. Parents' decision to use lateral or supine sleep positioning reflects their response to advice from specialists, observation of their infants' comfort, ease of breathing, and personal experience. Outcomes, identified in published research of SDB, coincide with parents' concerns but are expressed in medical language and fit into distinct domains of "snoring," "sleep," "gas exchange," and "apnea."

**Conclusions:** Parents' description of sleeping and respiration in infants with CP reflect their everyday experience, offering insight into their understanding, priorities, and language used to describe respiration. Understanding parents' individual priorities and how these are expressed could be fundamental to selecting meaningful outcomes for future studies of airway interventions.

## Keywords

infants, cleft palate, sleep-disordered breathing, outcomes

## Introduction

This article presents parents' perspectives of airway issues and sleep-disordered breathing (SDB) in infants with cleft palate (CP). Parents' priorities are discussed in relation to clinicians' priorities for identifying and managing respiratory effort, identified from published research regarding SDB in infants with, and without, CP. When selecting outcomes for use in effectiveness studies of airway interventions in children with CP, the opinions of parents with "lived experience" is fundamental to the meaningful evaluation of benefits. However, we are not aware of any reports in the literature. Therefore, understanding the shared and individual priorities of parents and clinicians is

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important in the evaluation of interventions and to enable clinicians to support parents better.

Airway problems frequently occur in infants with craniofacial anomalies (Boston and Rutter, 2003; Steward, 2007; Shadfar et al., 2012) and are associated with sleep-related breathing difficulties, such as SDB (Tan et al., 2016). The risk of significant airway obstruction is well understood in Pierre Robin sequence (PRS) and severe isolated micrognathia, with infants frequently managed in the inpatient hospital setting (Collins et al., 2014). Interventions may include sleep positioning (lateral lying), airway adjuncts (nasopharyngeal airway), nasogastric feeding (nasogastric tube), and tracheostomy. Much less is understood regarding SDB in infants with isolated CP. These infants do not normally require inpatient airway management, as they are managed within the family context in the United Kingdom, under the supervision of the clinical nurse specialist (CNS).

Sleep-disordered breathing may present with snoring, with or without apnea, arousals from sleep, and poor sleep quality. Possible longer term consequences of untreated SDB include altered craniofacial growth, neurocognitive delay, pulmonary hypertension, and decreased educational attainment (Blunden and Beebe, 2006; Karpinski et al., 2008; Lam et al., 2010; MacLean et al., 2012). The significance of these risks in children with CP requires further research.

Parents of infants may be aware of clinical indicators of airway obstruction but may not necessarily interpret their observations as indicative of SDB. Consequently, SDB may be untreated in infants with CP, with implications for short- and long-term outcomes. Given that SDB may be managed by relatively simple interventions, such as sleep positioning, it could be important to understand parents' awareness of indicators of SDB.

There are a number of outcomes related to SDB in infants with CP used in research, but the literature reports little information about parents' understanding of SDB and their priorities during infancy. Consequently, it is uncertain how parents' priorities match or overlap with researchers' decisions about essential outcomes. In keeping with guidance on using qualitative evidence to contribute to the development of core outcome sets (Keeley et al., 2016), we investigated the views of parents looking after infants with CP, aged approximately 16 weeks, exploring parents' opinions and experience of their infants' breathing and sleeping. These are discussed using evidence from a mapping exercise of outcomes reported in the research literature related to SDB in infants with, and without, CP.

The study aims were to:

1. Identify outcomes relating to breathing and sleep that are relevant to parents, as reported in their own words during the early weeks of caring for infants with CP.
2. Undertake a systematic search of the research literature for SDB in infants with, and without, CP.
3. Compare outcomes from the systematic search of research literature and parents' primary concerns in

**Table 1.** Inclusion and Exclusion Criteria.

Inclusion criteria	1. Infants diagnosed with an isolated cleft palate
	2. Age <1 month at recruitment
Exclusion criteria	1. Infants with cleft lip with or without cleft palate
	2. Infants with an associated syndrome
	3. Infants who require an intervention to assist with breathing (nasopharyngeal airway [NPA]) at initial recruitment interview (the requirement for such an intervention as part of normal clinical care following enrollment in the study will be recorded)
	4. Infants who required an intervention to assist feeding (nasogastric tube [NGT]) at initial recruitment interview
	5. Infants born prematurely (<36 weeks' gestation)
	6. Infants with known cardiorespiratory disease
	7. Infants of families with a history of sudden infant death syndrome (SIDS)

order to develop initial suggestions toward a core outcome set for SDB in infants with CP that is meaningful to parents as well as relevant to research.

## Method

### Research Design

A qualitative study, using semi-structured interviews, explored parents' perspective of sleep and breathing in their infants with CP. Researchers interviewed parents about their views as part of an observational study monitoring infants with CP. During the full study, parents participated in 2 separate sleep monitoring sessions when their infant was below 4 to 7 weeks and second monitoring at 3 months (see SLUMBERS). One researcher interviewed parents after they had either (1) completed 1 or 2 sleep monitoring sessions or (2) declined to participate in sleep monitoring but consented to be interviewed.

### Recruitment and Sampling

We recruited parents of infants with CP participating in the observational study in 3 specialist cleft centers in England using inclusion and exclusion criteria as determined by the main study (Table 1). Twenty-seven parents of infants aged 6 to 16 weeks diagnosed with CP were interviewed (26 mothers and 1 father). A total of 31 parents were invited to participate in an interview; 4 could not be contacted following their participation in sleep monitoring and were lost to the study.

Research nurses or CNS arranged for sleep monitoring of infants to take place with parents and then invited them to participate in an interview with a qualitative researcher, usually after 2 sessions of sleep monitoring. The study received ethical approval from the Health Research Authority. Research nurses or CNS took informed written consent at the outset of the study, with confidentiality and data security assured. The qualitative researchers contacted parents by telephone, arranging face-to-

face or telephone interviews according to parents' preferences. Recruitment continued until the qualitative themes were well developed and theoretical saturation was achieved.

### Data Collection and Analysis

Data collection took place over 8 months in 2016. Parents participated in semi-structured interviews in which the researcher used a topic guide (Appendix A) to gather parents' views of (1) their infant's sleep behavior, (2) major concerns about sleeping and breathing, (3) sleep positioning, and (4) experience of participating in observational study. The topic guide was developed with an advisory group of researchers, clinicians, and parents. We recorded the interviews using audio recordings (24 parents) or field notes (3 parents). Field notes were used when parents opted to have a conversation with the researcher without arranging a time for a phone interview when it could be audio recorded. Each audio recording was transcribed verbatim prior to analysis. We managed the data using the NVivo software system (2012). NVivo enables systematic storage, organization, and qualitative analysis of data, including coding interview data and identification of recurring themes. In the first instance, each interview was coded independently by 2 researchers. Codes were cross-checked by both researchers and final coding agreed for each interview. The codes across the interview data set were then compared and contrasted to interpret the data by identifying main themes and subthemes (Braun and Clarke, 2006). The codes and themes were reviewed independently by a third researcher to confirm or question the original analysis. The themes derived from this stage of analysis were included in a matrix format of "cases by themes" to enable analysis of the relationships between cases and themes of the entire data set using a "framework analysis" approach (Ritchie and Spencer, 2002).

### Systematic Search of the Research Literature

A systematic search was undertaken to identify studies reporting on the incidence, severity, and management of SDB in children prior to repair of the CP (typically less than 1 year of age). Further studies reporting outcome measures in SDB in infants with, or without, CP were identified. Randomized and nonrandomized-controlled trials, prospective cohorts, case-control studies, and case series were included. Full details of the inclusion criteria, search strategy, and data extraction process are presented in Supplemental File. Given that the review aimed to provide a "map" of the outcomes used in the current research literature, no risk of bias assessment was undertaken. Details regarding the SDB outcomes were tabulated and categorized into broad domains.

## Results

### Findings From the Qualitative Study

Parents were given the option to complete a telephone interview or a face-to-face interview. The majority opted for a

telephone interview (25 parents). Interviews lasted between 15 and 35 minutes. Basic demographic data indicated that parents were predominantly from low to medium socio-economic status (SES) groups as indicated by the English Index of Multiple Deprivation (2015) score using the family's postcode. Fifteen infants were first babies in the family. Findings are reported using the words of parents, coded P1-P27.

### Parents' Descriptions of Infants' Breathing

Parents used a wide variety of terms to describe infants' breathing during sleep, with many explicitly describing their baby's breathing as noisy ( $n = 14$ ). The language they used ranged from commonplace terms such as "noisy breathing" (P1, P2, P6, P14), "heavy breathing" (P6, P17), "snoring" (P6, P15, P17), and "snuffly" (P3, P6, P10, P12) through to more idiosyncratic terms such as "ack-ack noise" (P13), "crackly" (P5), "grunts and moans" (P7), and observations of associated behavior, for instance, "sucks his lips" or "flails his arms." A number of parents specifically expressed concern about choking ( $n = 11$ ).

Parents' descriptions indicated that they were careful observers of their infant's behavior and monitored their infants closely during sleep:

Sometimes she can be more of a heavy breather than other times, but not actually that bad really . . . I think it's—as I say it's just she had her mouth open a bit like a snore—not a full, not like a proper snore but just that, sort of, a bit like a light snore. (P17)

Parents often referred to making judgments about whether a behavior or condition warranted concern. They made decisions based on observing a range of signs, separating what they regarded as normal for their child and what was seen as unusual or potentially problematic. For example, P1 described her infant as a "very noisy breather." Her description was detailed and referred to visual observations as well as what she could hear of her infant's breathing including breath-holding and gasping for air:

He was a very noisy breather, he'd like blow bubbles, he would kind of, sort of, apnea he would hold his breath for a couple of seconds and then he'd do a gasp. (P1)

Most parents did not use medical terminology in their descriptions, and where it was used, it tended to be in the context of more colloquial descriptions. P1 used the term "apnea," but specifically in the context of her account of a conversation with the cleft nurse. P13 referred to "oxygen sat levels" during her infant's sleep, but, as the following example illustrates, she felt more confident judging her infant's comfort than ease of breathing.

Yeah, I don't know if she was able to breathe but more . . . she was more comfortable put it that way. I don't think she was not . . . I don't think her oxygen sat levels were bad on her back because she was being monitored all the time for this infection. (P13)

The association between breathing, sleep positioning, and oxygen levels did not routinely form part of parents' conception of their baby's well-being. This parent's first observation related to assessment of everyday behavior (comfort), followed by reference to the professionals' assessment of oxygen saturation levels, as secondary information. In this example, the mother was speculating about the medical implications of sleep positioning, expressing caution or uncertainty in the way she talks about it.

Five key features relating to parents' description of the noise infants made during sleep were derived from the interviews.

1. There were indications that parents "get used to" the sound and signs of breathing and become less anxious, assuming that the noise of their infant's breathing is a feature of their infant that does not necessarily indicate breathing difficulties, as illustrated by P6:

No when he was first born he was quite like noisy, but he doesn't seem to as noisy now anyway, so no not now. I think, I don't know if we are just getting used to it as well. (P6)

2. Parents linked choking and reflux with breathing difficulties but acknowledged that they may not have thought about implications of breathing and oxygen levels or the significance of the signs they observed. P6 commented, "I don't think I even realized about the breathing to begin with." P17 distinguishes between awareness of oxygen levels and considering implications for her infant:

Not that I'd necessarily thought about it, but yes, I would be aware that it would be important for her to have a consistent amount of oxygen... I can't say it's something I have particularly thought about before. (P17)

3. Parents did not always make an association between infants' snoring and the possibility that this signaled longer term breathing difficulties. There were examples of parents finding snoring reassuring, considering it as an indicator that their infant was still breathing, as illustrated by one parent:

But then I think actually with him being noisy that kind of reassuring because I could hear him breathing. I kind of, that worked to make me feel better as well, yes. (P6)

Parents observed differences between family members' level of concern. For instance, P16 described how her husband worried if their infant's breathing was quiet:

No he just worries when she goes quiet. It's not the fact that she breathes loudly; it's just he feels more comforted when he can hear her breathing. He's quite happy with her being a heavy breather; it makes him feel better. (P16)

4. Parents did not consider gradations in difficulties with breathing. They expressed concern about the extreme risk that their infant may stop breathing altogether, as illustrated by P7, "Because I'm scared that he'll stop

breathing and die." Most parents did not indicate awareness of the possibility of chronic obstruction characteristic of SDB, indicated by noisy breathing. Concern about breathing subsided as they became used to looking after their baby. Many expressed considerable anxiety initially,

I suppose the thing I find worrying is when she's asleep, just making sure that her breathing's okay, because I do worry about that, especially when... we've got used to her more with how her breathing is and everything, but when she was first born, you know, I'd come in the room, even in the day, just for two minutes to go and do anything, I wouldn't... you know, I wouldn't leave her. I used to get my mum to come round so I could go to the toilet and like have a shower and stuff. (P26)

5. Some parents expressed uncertainty about the relationship of CP to other symptoms and features of their infant's breathing during sleep. For example, P1 believed that her infant's breathing difficulties were unrelated to the CP. Another parent believed that her child's small nostrils contributed to breathing difficulties and the potential for interfering with infant's breathing:

Just, I think it may be a bit more complicated because he has breathing difficulties anyway, separately to his cleft lip; they are on about that his tongue is falling to the back of his throat when he is breathing. (P1)

### *Parents' Concerns About Sleeping and Breathing Were Embedded in the Wider Context of Caring for Infants With CP*

To understand what parents worry about most in relation to their infant's breathing and sleep, we asked parents about their greatest concern in caring for their infant. Parents' descriptions indicated that the majority perceived infants' breathing, sleeping, and feeding as interlinked and embedded in the wider context of caring for a young infant with CP. P7, for example, demonstrated a wide range of observations that she associated with her infant's breathing during sleep, during feeds, and after feeding:

I: And do you notice, apart from the noise, is his sleep disturbed?

P: He'll like jump, or something like that, isn't it?

I: So he'll sort of jump and...?

P: Or do a loud screech.

I: Yeah. Oh gosh, that sounds really worrying. So how... any changes in color, or anything like that, you've been aware of?

P: He chokes a lot anyway, you know, when he's having his bottle.

I: So, he's choking when he feeds?

P: No, afterwards he'll be sick or if I give... he's got thrush and I give him his thrush medicine and I think it's too thick for him and he goes purple (P7).

Many parents interviewed did not have concerns relating to breathing in isolation. For example, P12 reported her concerns as “nothing to do with his breathing.” Furthermore, parents did not refer to possibilities of chronic airway obstruction characteristic of SDB. The dominant feature of parents’ discourse focused on issues relating to feeding and sleeping. The primary concern was feeding their infant successfully, given the difficulties associated with a cleft in the mouth. Parents expressed concern about nutrition and weight gain, choking during feeds, regurgitation after feeding, and disappointment about not being able to breastfeed.

When asked about difficulties in breathing, parents frequently referred to choking during and after feeds including during sleep, rather than obstructed breathing occurring during sleep. They did not routinely focus on discrete aspects of behavior, such as breathing or sleeping, but tended to conflate these elements within wider concerns about feeding. For example, P7 explained, “when he’s asleep he chokes because he’ll be sick in his sleep and then it comes out of both holes.”

A small proportion of parents explicitly linked choking to their infant’s breathing. This was expressed as a fear that their infant would stop breathing, “I was scared in case she chokes or stops breathing because she’s got that hole” (P2). Two parents described episodes of choking that were severe enough to call emergency services:

She’s got reflux, what had happened is it had pooled again and just blocked her off, and we just needed to get her and that’s when we got admitted for the evening. We had to ring the ambulance out and then they put her on ranitidine, which has helped with the reflux, which has obviously helped with her sleeping as well (P10).

Reflux was a concern for 6 parents (P4, P9, P10, P14, P26, P27) and some described using medication to manage this. P13 referred to fear of airway blockage related to PRS, a condition discussed with the CNS and researched using Google:

I was thinking has she got this Pierre Robin thing, is there a problem with her airways, is that why she’s making this weird ack, ack noise and kicking off. (P13)

P7 was advised by the CNS that her son had small nostrils as a family trait. She believed that was why he was not getting enough oxygen during his sleep.

[The cleft nurse] thinks that his nostrils are too small, you know, his holes, because his dad snores and his brother snores as well really loud. So she thinks that it could be something that runs in the family (P7).

In summary, parents expressed their greatest concern as feeding, with breathing difficulties associated with choking and regurgitation rather than underlying, chronic airway obstruction.

### Parents’ Perception of Managing Respiratory Effort

Parents’ use of lateral or supine positioning for their infants’ sleep varied but generally followed the specialist advice provided by cleft practitioners. Parents’ decision to use a lateral sleep position to improve respiratory effort tended to be based on specialist advice, their observations of infant’s comfort, and parents’ preferences and beliefs based on experience. A number of parents expressed strong views about preferring one position to another, but there was no consensus between parents about one position being superior. Rather, they stressed that the position “works” based on their close observation of their infant.

All the parents discussed sleep position with health practitioners but did not always recall the reasons why a position was recommended to them. For instance, P12 thought the lateral position was due to the infant’s regurgitation of food, but she expressed uncertainty by the time she was interviewed:

I: Did you know the reason for the side position being recommended?

P12: Oh, I can’t, do you know, I know I was told, and, you know . . . Because I know at first I thought it was due to him regurgitating all the time, and I think it was . . . wasn’t necessarily due to that, but I can’t remember now, you know.

Parents formed their own opinions as well as following advice of clinicians. For example, P2 put her infant in a lateral sleeping position when/as advised by a doctor in the baby care unit. However, she believed the infant’s breathing was better on the side.

On her side her breathing’s alright, but on her back she makes this funny noise, and like I can’t really say, like [makes funny noise] noise . . . . So that really scares me. (P2)

In this example, the parent made a clear reference to the importance of comfortable breathing. In contrast, P13 described her infant’s comfort and distress as her main considerations in choosing a sleeping position.

Because whenever she slept on her back she’d get quite distressed and she sort of made . . . like even if you were changing her nappy when she was first born she made this ack, ack, ack, ack type noise which.

And we had a couple of incidents while we were in hospital where she was being sick and she couldn’t make a noise and she was just flapping her arms and she was on her back. But as soon as you put her on her side it just drains away so it’s not an issue. (P13)

Some parents were cautious in adopting the advised sleep position and carefully monitored the infant’s responses to positioning. On occasions, the choice of sleeping position was portrayed as the infant’s choice as indicated by their comfort, rather than that of clinicians or parents.

### Important Considerations for Parents

None of the parents' accounts referred directly to "outcomes." Parents referred to significant concerns and described their anxieties and aspirations clearly. They also commented on the rapid changes in their infants as they grew and developed during the first 3 months; routines altered as parents adapted to changes in their infant. As a consequence, their major concerns changed during the first months. The principal outcome for parents tended to be immediate challenges such as feeding and managing sleep. There were examples of longer term outcomes that were not directly related to breathing, such as managing the difficulties associated with future operations (P6, P13, P22, P23, P28), speech and language development (P6, P16, P26, P28), and bonding between parent and child (P5).

Some parents found it difficult to answer the question about their primary concerns. For example, P15 had no concerns about any aspect of her infant's feeding, breathing, or sleeping, suggesting that her infant's personality contributed to her confidence:

But like she's a happy, chilled out child as well, so it makes it a lot easier, doesn't it? (P15)

### Results From the Systematic Search

A total of 32 studies were identified. Fifteen studies focused on SDB in infants with CP and 17 studies examined SDB in infants without CP. A Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) diagram showing the flow of studies in the systematic review process, along with the characteristics of all included studies and reasons for exclusions, is presented in the Supplemental File.

The outcomes reported in the identified studies were broadly grouped into the following domains:

1. Snoring: No definition was provided for the outcome or clarity about measures used to evaluate snoring.
2. Sleep: Eight studies reported on total sleep time, time to fall asleep, night sleep, day sleep, nocturnal awakening, arousal events, nights of restless sleep, arousal, periodic breathing time, stridor or stertor, time of quiet, and SDB.
3. Gas exchange: Defined as the process by which oxygen is taken in, and waste gas removed from the bloodstream; can be affected by airway obstruction during sleep.
4. Apnea: defined as episodes of breath holding while asleep.

Table 1 in the Supplemental File presents the "long list" of outcomes reported in the individual studies and their definitions (where available).

### Discussion

This article presents parents' description of their concerns regarding sleeping and breathing in their infants with CP.

Using qualitative methods provided an opportunity to understand parents' perspectives of potential SDB and explore the meaning they attach to behavior and symptoms they observe in their infants. Comparing parents' description of breathing with outcomes used in studies of SDB in CP reveals that parents' main concerns are generally similar to research priorities. All of the outcomes derived from the systematic search were mentioned by some parents, but not all the parents referred to them. However, parents tended to interpret their infants' behaviors, such as sleeping and feeding, as closely linked, rather than consider specific behaviors as separate and discrete entities. They also attached different significance to common indicators of SDB, such as snoring. Snoring was frequently mentioned by parents but rarely considered as a trigger for concern. Parents used a variety of descriptive labels, occasionally making use of medical terminology that they had heard professionals use. The differences between parental knowledge, based on intimate involvement with an individual child, and professional knowledge, founded on systematic study and wide experience of groups, has been well-documented (Callery, 1997; Carlhed et al., 2003), but implications for identifying key outcomes in infants have not been investigated.

There are 5 notable differences between the clinical outcomes used by researchers and parents' primary concerns. First, parents considered breathing together with feeding and sleeping and rarely raised breathing as a primary concern in isolation. The focus of concern, the principal outcome for parents at the outset of caring for a child with CP, are immediate challenges such as feeding and managing sleep rather than ongoing difficulties with breathing that are characteristic of SDB. Occasionally, they referred to longer term outcomes that are not directly related to breathing, such as speech and language development and managing the difficulties associated with future surgical interventions. In contrast, research examining SDB investigated breathing as a discrete behavior identified and measured in the research by snoring (Rustemeyer et al., 2008; Moraleda-Cibrián et al., 2014), sleep quality (MacLean et al., 2008), gas exchange (Silvestre et al., 2013), and observation of apnea (MacLean et al., 2012, Moraleda-Cibrián et al., 2015).

Second, those parents who expressed concern about breathing generally referred to episodes of choking or reflux, associated with both feeding and sleeping. These episodes were perceived as serious and caused great anxiety. Two parents sought emergency advice following choking episodes and their infants were hospitalized. Several parents referred to reflux medication improving their infant's sleep and breathing, although data on this were not explicitly collected. Infants with PRS are prone to increased reflux (Marques et al., 2009) but there is little evidence indicating that infants with isolated CP are also prone to increased reflux. Choking and reflux-related incidents do not routinely form part of SDB studies but perhaps should be considered as the increased work of breathing during sleep may result in increased negative intrathoracic pressure during the respiratory cycle and an increased tendency toward extra-esophageal reflux.

Third, the language parents used to describe breathing showed considerable variation and they often had difficulty describing their observations. Parents used terms such as a “light snore” or a “heavy breather,” an “ack-ack” sound, or “funny little noise.” Studies in the literature review measured length and frequency of snoring as indicators of SDB. The presence or absence of “airway noise during sleep” may be an appropriate alternative outcome description to “snoring,” to describe this consequence of upper airway obstruction. Medical terminology, such as “gas exchange,” “blood gas,” and “apnea,” were mentioned by few parents. However, some parents described changes in infants’ color, breath holding, and on one occasion, oxygen saturation.

Fourth, while parents are careful and attentive observers of their infant’s behavior, they may not identify or interpret clinical indicators in the same way as clinicians and researchers. For instance, snoring is considered an indicator of SDB for researchers but could be perceived as reassuring for parents caring for their infant or simply a unique characteristic of their child, but none of our sample described it as an indication of problems with breathing. This would suggest that work is required to educate parents regarding SDB, its symptoms, consequences, and management.

Finally, infants with CP are changing rapidly as they grow and parents described adapting their care. Parents’ referred to finding it difficult to remember the specifics of caring for their infant in the early days and how routines changed as babies developed their own preferences and families adapted to the additional needs of their infant. This highlights the importance of prospective data collection when studying SDB in children with CP or evaluating airway interventions in CP.

Parents’ accounts in this study provided their perspectives, use of language, and tendency to include wider issues from the context of caring for infants in their dialogue about primary concerns. Future research designs, including design of sleep questionnaires for this population, could benefit from incorporating the language used by parents and may want to consider the relevance of wider context on SDB. Breathing is rarely considered as a discrete element of an infants’ well-being, but considered in relation to feeding by parents.

Interpreting the findings from the qualitative study raises important issues for the potential consequences of SDB and CP upon the airway. The evidence from the systematic search suggests that infants with CP are more prone to SDB, with potential long-term consequences. Our study suggests parents fear episodes of choking relating to feeding and reflux that appear life-threatening rather than ongoing signs of SDB such as snoring. This raises a difficult ethical issue for clinicians. Should parents be made aware of the risks of SDB, signaled by indicators that are considered benign by parents, such as snoring? Raising awareness could, on one hand, prepare parents and offer interventions that alleviate symptoms, such as sleep positioning. However, on the other hand, it may raise anxieties unnecessarily and prompt concern about longer term outcomes such as neurocognitive delay, which have yet to be proven conclusively in research.

Recent attempts to include service users in a process of developing core outcomes sets (Core Outcome Measures in Effectiveness Trials, <http://www.comet-initiative.org>) for effectiveness studies have employed Delphi techniques, with panels of patients and clinicians voting to achieve consensus to select outcomes (Williamson et al., 2012; Keeley et al., 2016). The current study highlights the difficulties of using this approach in studies involving infants. Parents are at an early stage in their experience and infant characteristics are changing rapidly. They often describe their experience in idiosyncratic ways without a consistent use of language to describe their observations of their infant. The study illustrates the feasibility and benefits of using qualitative investigation to understand parents’ priority outcomes, differences in language used between parents and researchers, and variation in interpreting infants’ characteristics during the early weeks of life. The aims of the qualitative study did not intend to achieve consensus but provide a depth of understanding of parents’ perspective of outcomes, foregrounding differences between parents’ perceptions and researcher priorities in investigating SDB.

## Conclusion

This study has 3 specific implications for practice and research. First, parents may be unaware of the presence of SDB and potential risks for long-term outcomes for their infant. Parents make careful observations of their infants’ behavior but tend to conflate breathing issues with more immediate concerns such as feeding and sleeping. This raises a question for clinicians regarding parent education. It is important to balance enabling parents of infants with CP to interpret signs of SDB, while avoiding raising parental anxiety unnecessarily. Second, the evidence that parents’ descriptive language of infant breathing rarely coincides with the labels found in research literature suggests that a “language gap” between parents and clinicians may exist. While parents and clinicians use different terminology, there is potential for misunderstanding. Clinicians may need to consider modifying their use of language when communicating with parents. Encouraging parents to articulate their observations and clinicians to be aware of terms used by parents to describe the nature of their infants, breathing could be an important step in collaboratively identifying infants at risk of SDB.

Third, clinical research focuses on discrete medical outcomes, often using terminology that has limited meaning to parents. When selecting outcomes for use in effectiveness studies of airway interventions in children with CP, the opinion of parents with “lived experience” is fundamental to meaningful evaluation of benefit. Therefore, in order to evaluate interventions for SDB in infants with CP, research needs to consider the priorities of both parents and clinicians to agree appropriate outcomes for future studies.

## Appendix A

### Topic Guide for Interviews With Parents

1. Background to child's sleep
 

Ask about child's sleeping behaviors/habits (how are they sleeping, disturbance, comfort, difficulties in breathing)

  - From birth until now
  - How felt when first put child down to sleep
  - Any changes in these feelings over time?
  - Any difficulties (choking, noises when sleeping, greatest concern, greatest impact on parents)
2. Information about sleeping
 

Advice parents have had about sleeping position

  - Who provided this?
  - How often is it mentioned?
  - How felt about this information?
  - Potential clash with national advice for back sleeping
  - How easy is it to follow this advice?
3. Being part of the observational study
 

Explore what made the parent decide to take part

  - What influenced their decision?
  - What were your reasons for taking part?
4. Child's sleep during the observational study
  - Can you tell me how your child's sleeping has been on the nights when they were monitored as part of the study?
  - How far has this been normal for your child?
  - How easy was it to sleep your child on (their side/back)?
5. Experience of being in the study
  - How have you found being in the study?
  - What has been difficult? (using equipment)
  - What, if anything, could have been improved? (views on monitoring in research center)
  - What has been good?
6. Information about the study
  - How clear was the information provided to you about the study?
  - What, if anything, could have been improved?
  - Who did you talk to about taking part?
  - How easy was it to decide to take part?

### Authors' Note

P.C. designed the interviews, analyzed the data, and drafted and revised the manuscript. K.D. and Y.L. collected and analyzed the data and drafted and revised the manuscript. I.B. designed the research and drafted and revised the manuscript. A.M.G. conducted the literature review and drafted and revised the manuscript. All authors read and approved the final manuscript. Ethical approval gained from NRES Committee North West—Greater Manchester Central 15/NW/0010.

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### Supplemental Material

Supplemental material for this article is available online.

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