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Parental Judgement of Hearing Loss in Infants With Cleft Palate

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Abstract

Objective: To investigate whether reported parental concern is supported by hearing assessment findings in children with cleft palate. To describe this population by examining the relationship between cleft type, middle ear status, and hearing loss.

Design: Retrospective consecutive case note review.

Setting: Tertiary institutional regional cleft center.

Patients: Consecutive cases of 194 babies born with cleft palate and referred to the specialist center from January 2009 and December 2013. Following exclusions, data from 155 infants were included for analysis.

Interventions: Documented parental concern in ear, nose and throat (ENT) and speech and language therapy case notes were compared to hearing assessment findings. Findings from otoscopic examination, tympanometry, and hearing assessment were analyzed with respect to cleft type.

Results: Parental concern is not always accurately reflected by objective assessment particularly when no concern is reported. Analysis of the cohort examined suggests that cleft type is not related to middle ear findings or hearing.

Conclusions: It is helpful to be aware of parental concern and clinicians should consider that parental reports may not be accurately reflected by test results. As cleft type was not found to substantially influence middle ear status or hearing it is not recommended to adapt speech and language advice offered to families according to cleft type. Follow-up studies to increase participant numbers would support a statistical analysis.

Keywords

cleft palate, cleft type, hearing loss, parental perception, pediatrics, anatomy, otitis media with effusion, middle ear, parental concern

Introduction

Otitis media with effusion (OME) is an accumulation of fluid within the middle ear and is a frequent presentation in infants with cleft palate (Flynn et al., 2009). The high incidence of OME in this population is explained by Eustachian tube dysfunction secondary to cleft palate (Doyle et al., 1980). It presents either unilaterally or bilaterally and may not be present at the neonatal hearing screen (Szabo et al., 2010), but its onset is progressive and most common in the first year of life (Li et al., 2007). Otitis media with effusion frequently results in a persistent fluctuating conductive hearing loss (Viswanathan et al., 2008; Szabo et al., 2010). This hearing loss has been demonstrated to negatively influence speech and language outcomes in children with cleft palate (Lohmander et al., 2011; Hall et al., 2017). As the child grows, Eustachian tube function usually improves around age 6 years due to a combination of factors including growth (Moller, 1981, Alper et al., 2016). However, in the presence of cleft palate, OME can persist with evidence of negative impact in terms of psychological well-being, educational experience, and social interaction (Tierney et al., 2015; Chando et al., 2016). Early identification and management is therefore important to minimize these potential consequences.

The question as to whether parents are accurate in their judgment of hearing loss is debated. Evidence relating to the cleft palate population is sparse but largely suggests that parents are not accurate in detecting hearing loss. D'Mello and

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Kumar (2007) examined audiological outcomes of patients with a cleft attending a speech camp in India and compared these to parental/carer awareness of identified hearing difficulty. Of 43 children, 38 were identified with hearing loss. Although no quantitative data were offered, it was reported that "most of the parents/caregivers" were unaware of any hearing difficulty.

The view that mild–moderate hearing loss often goes unnoticed by parents is also supported by Luthra et al. (2009) who investigated 55 children with cleft lip and palate (CLP) where no complaints of hearing loss had been made. They found 25 children with unilateral loss and 16 children with bilateral loss. The authors noted that hearing loss is frequently overlooked unless the problem is acute.

Sheahan et al. (2003) and Engel et al. (2000) used questionnaires to examine middle ear problems in cleft palate and identify parental perception of hearing. Both authors noted low parental concern relative to the level of OME identified.

Examining the wider literature to include children without cleft palate, Lo et al. (2006) looked prospectively at 276 children selected from a population-based OME screening survey of almost 6000 six- to seven-year-olds in Hong Kong. They identified no association between parental perception and pure-tone audiometry findings. A further study by Lau et al. (2015) explored the agreement between hearing loss, measured by auditory brain stem responses, and clinical questioning in detecting hearing loss in children with Down syndrome. Overall, they found poor strength of agreement between objective testing and parental reporting of hearing loss suggesting poor parental awareness of hearing impairment.

There is limited opposing evidence that parents are accurate in their assessment of hearing loss. The evidence arises from the wider literature on children with OME not associated with cleft palate and is mostly dated. Thompson and Thompson (1991) urged that we should listen to parents following an evaluation of 49 returned questionnaires from parents of children with identified hearing loss. Parving and Christensen (1992) similarly found that parents were more accurate in their assessment of hearing loss than professionals.

A more recent retrospective review supports the view that parents are best at identifying hearing loss (Dedhia et al., 2013). A total of 78 of 923 children were identified with sensorineural hearing loss (SNHL) after passing the neonatal hearing screen. The identification of hearing loss was most often by parents (36%). However, this study covers a much broader spectrum of severity of loss than the mild conductive loss classically associated with OME.

When considering whether parents of infants with cleft palate are accurate in their perception of hearing, it is important to examine this cohort of infants and ask whether this a homogeneous population. The nomenclature of "isolated cleft palate" (CP), "soft palate cleft," "unilateral cleft lip and palate" (UCLP), "bilateral cleft lip and palate" (BCLP), and "submucous cleft palate" (SMCP) is well recognized and differentiates the broad categories of clefting. Such categories refer to anatomical differences that broadly relate to severity. For example, an SMCP may be considered the least extensive anatomical defect affecting the muscles in the soft palate, and a BCLP the most extensive, affecting the lip, nose, alveolus, and hard and soft palate. It is plausible therefore to suggest that the more extensive the cleft, the greater the potential for middle ear dysfunction and consequential hearing loss.

Some studies have examined anatomical and physiological differences according to cleft type. Harris et al. (2013) categorized 2737 children into 2 groups: CLP and CP. They found that the CLP group were significantly more at risk of developing cholesteatoma than the CP group. They postulated that the more extensive movement of tissue required to repair the CLP group may have impacted on Eustachian tube function, which led to the increased risk of cholesteatoma in this group.

Lehtonen et al. (2016) discovered significant increases by cleft type in the detection of mucous secretions suggesting OME with prevalence greatest in the CLP group. In their cleft lip group, the rates reported approximate worldwide prevalence of OME in healthy children.

Other studies report that cleft type is an important factor in relation to hearing loss. For example, Sundman et al. (2016) investigated auditory brainstem response thresholds and found that infants with CP and OME had significantly higher hearing thresholds than infants with UCLP and OME.

It has also been found that the more extensive the cleft, the more likely the affected child is to receive grommets. In the Lehtonen et al. (2016) study, significant variation was reported in number of grommet insertions by cleft group. Ahn et al. (2012) reported similar findings.

A further retrospective review revealed a significant relationship between middle ear function, grommet insertion, and cleft type (Çağlar et al., 2013). From a 175 patient cohort, abnormal Eustachian tube function was found in 68.5% of the CLP group compared with 26% of the CP group. In addition, the CLP group more often required 2 grommet insertions.

In contrast to the above reports which suggest an influence of cleft type on middle ear status and hearing, other studies have found the converse. Zingade and Sanji (2009) found no significant relationship between cleft type and presence of OME. More recently, Skuladottir et al. (2015) compared longitudinal hearing outcomes in patients with CLP and those with CP and found no significant differences in hearing levels. Lithovius et al. (2015) also found that cleft severity was not a significant factor in relation to hearing loss in their cohort of 90 patients aged 3 to 9 years. Similarly, Imbery et al. (2017) reported no significant differences between cleft types in either air conduction pure-tone average or bone conduction pure-tone average in 564 patients.

It is problematic to compare studies in relation to cleft type and middle ear/hearing status, as most either do not distinguish between different cleft types (eg, Chen et al., 2012; Szabo et al., 2010) or examine a single type (eg, Flynn et al., 2014; Lohmander et al., 2011). Those studies that do distinguish between cleft types have differing definitions, and participants can differ according to whether the palate is repaired or unrepaired. Further, participant ages vary considerably making

comparisons within and across studies challenging. Low participant numbers and lack of recent research are additional limiting factors, and there is a dearth of reports examining infants at around 1 year of age when incidence of OME is reported to be high.

To summarize, the literature generally supports the view that parents do not consistently identify hearing loss associated with OME; although there is a lack of recent reports and few studies have examined the infant cleft population specifically. It is also evident that most studies investigating hearing loss in cleft palate do not distinguish between cleft type, and there is limited evidence regarding whether cleft type influences middle ear status and hearing findings.

Study Aims

To address the issues identified above, by examining 2 questions:

1. Is parental concern about their infants' hearing at (or just prior to) palate repair supported by hearing test findings?
2. Is the degree of hearing loss and middle ear findings (the presence of OME and tympanometry findings) at time of palate repair related to cleft type?

The findings will contribute to the evidence base and may assist in the development of more targeted intervention, for example, by identifying a subgroup of children with cleft palate who could benefit from more or less frequent routine audiological/ENT review or differential speech and language therapy advice.

Methods

Setting and Design

The study was undertaken at a hospital within a city-based regional Cleft Center, 1 of 11 specialist cleft centers in the United Kingdom. With ethical approval, the researcher conducted a retrospective review of patient case notes.

Participants

All babies born with CP $\pm$ lip within the designated region of the Cleft Center are referred to the hospital where surgical repair takes place. Consecutive cases of children born with CP $\pm$ lip who underwent surgical repair at the Center were identified over a 5-year period from January 2009 to December 2013. This was a convenience sample, representative of the population studied with 5 years of data included to increase participant numbers. A total of 194 infants were identified. Twenty were excluded due to a medically diagnosed syndrome or were undergoing genetic testing, and 19 infants were excluded as ENT records were unavailable for analysis. In total, 155 infants were included for analysis (80 male, 75 female). The 155 infants were categorized into the following diagnosed cleft types: cleft of soft palate (soft) n = 36; cleft of soft and hard palate (soft + hard), n = 42; UCLP, n = 51; BCLP, n = 18; or overt SMCP, n = 8.

Method

Question 1: Is Parental Concern About Their Infants' Hearing at Palate Repair Supported by Hearing Test Findings?

A question routinely posed by the ENT consultant at the presurgery hearing test regard any concern that the family have about their child's hearing. There is a single ENT consultant within the cleft center and so the method of inquiry is likely to be similar with each family. The response is documented in the case notes. Where given, these comments were categorized into (a) no concern, (b) concern, or (c) unsure, with one category allocated per infant. Most frequently, the recorded comment was directly regarding concern (eg, "no parental concern re hearing"), but sometimes a degree of interpretation was required. For example, "happy with hearing" was interpreted as "no concern" or "parents think hearing is down" was interpreted as "concern."

The speech and language therapist (SLT) also meets with the infant and family for a separate presurgery advice session, usually carried out at around 6 months of age. The SLT provides advice including the potential impact of hearing loss on speech development. Parents are routinely asked if they have any concern regarding hearing and responses are documented. All speech case notes were analyzed for this information, and as before, the level of concern where reported was categorized into (a) no concern, (b) concern, or (c) unsure.

Audiological records are held separately following hearing assessment. It is not known whether the audiologist routinely asks about parental concern and this was not a focus of this study.

The parental concerns reported independently to ENT and SLT were examined with respect to assessment findings to investigate whether parents are accurate in their perception of hearing loss.

Question 2: Is Cleft Type Related to Middle Ear Findings (Presence of OME and Tympanometry Findings) and Degree of Hearing Loss at Palate Repair?

A hearing assessment and ENT consultation is routinely offered 1 week prior to palatal surgery, between 5 and 12 months of age. The ENT/audiology case notes of the 155 infants were examined for otoscopic and tympanometry findings. These findings indicate whether the middle ear is functioning normally. Otoscopic findings were categorized into: no OME (ie, normally ventilated ears), unilateral OME, bilateral OME, or obscured view. Tympanometry findings, indicating the presence or absence of OME, were classified as either bilaterally normal or abnormal, the latter comprising both flat and negative pressure responses. In some cases, only 1

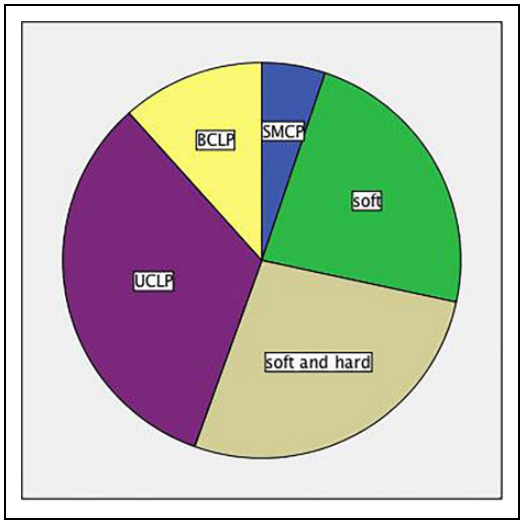

Figure 1. Distribution of cleft type.

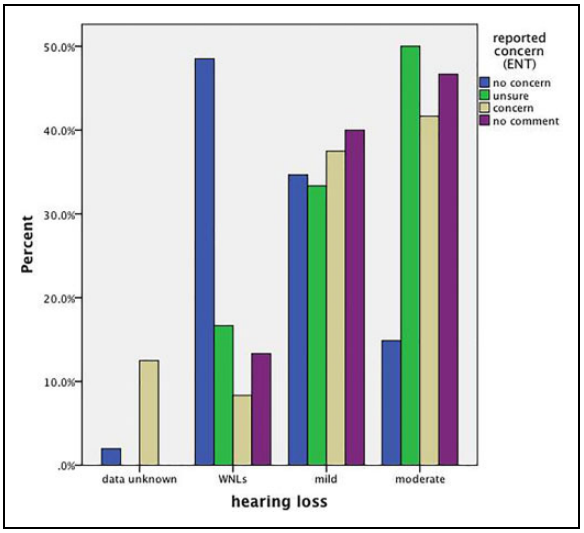

Figure 2. Percentage of parent concern reported to ear, nose and throat (ENT) consultant compared to hearing test findings.

ear gave an abnormal result, which was classified as unilaterally abnormal. These findings were examined in respect to cleft type to investigate whether any relationship exists. Due to low numbers in the SMCP and BCLP subcategories, descriptive statistics were used.

At the presurgery assessment, a hearing test is attempted using age-appropriate sound field audiometry (Visual Reinforcement Audiometry or Play Audiometry) assessing the better hearing ear. All available audiograms of the 155 infants were analyzed. Guidance was taken from senior audiologists who advised that sound field thresholds of 30 dBHL across the frequency range tested should be accepted as hearing within normal limits (WNL) which is in line with other UK protocols. Responses from 31 to 40 dBHL were interpreted as a mild loss, from 41 to 70 dBHL, a moderate loss, and from 71 to 95 dBHL a severe loss. If the infant responded at different intensity levels for different frequencies, an average of intensity levels at 500, 1000, 2000, and 4000 Hz was calculated. Only audiograms with 2 or more frequency responses were classified with each infant categorized as having a mild, moderate, or severe hearing loss or hearing WNL. The presence of a known SNHL was also noted. Once again, hearing test findings for each infant were analyzed with respect to cleft type to examine the relationship between them.

Results

Cleft Type

Figure 1 shows the distribution of cleft type in the 155 infants. This distribution can be viewed as representative of the wider cleft population (Tanaka et al., 2012, 2013).

Age at Presurgery Hearing Assessment

The mean age at hearing assessment of the 155 infants was 10 months (median 13 months, range 5-64 months) with a mode of 7 months. Only a few participants were older than 12 months; these were from the SMCP group due to late diagnosis.

Parental Concern About Infant Hearing at Palate Repair Compared to Hearing Test Findings

Documentation regarding concern (including concern, no concern, or unsure) was found in 131 (84.5%) of the 155 infants' ENT case notes. Of these, 101 (77.1%) parents reported no concern regarding their child's hearing at the presurgery hearing test, 24 (18.3%) parents reported concern, and 6 (4.6%) were unsure. Figure 2 shows the level of concern reported to ENT compared to actual hearing test findings.

Of the 57 infants with hearing WNL, no concern was reported by 49 (86.0%) parents. However, 35 (66.0%) of 53 parents reported no concern when assessment identified a mild hearing loss, and a further 15 (39.5%) of 38 parents reported no concern when a moderate loss was identified. Of the 24 (18.3%) of 131 parents reporting concern, the majority of infants were found to have either a moderate loss or a mild loss. Only 2 (8.3%) of 24 parents reported concern when hearing was found to be WNL.

When parents were unsure about their infant's hearing, the status of the infants was varied; from 6 cases where parents were unsure, 1 infant presented with hearing WNL, 2 were identified with a mild loss, and 3 with a moderate loss.

Documentation regarding parental concern (including concern, no concern, unsure) reported to SLT was found in 80 (51.6%) of 155 case notes. Where documentation was available, 63 (78.8%) of 80 parents said that they were not concerned about their infant's hearing. Concern about hearing was reported by 15 (18.8%) of 80 and 2 (2.5%) of 80 were unsure. These findings should be viewed in the context of unknown/unavailable data to SLT (75/155; 48.4%).

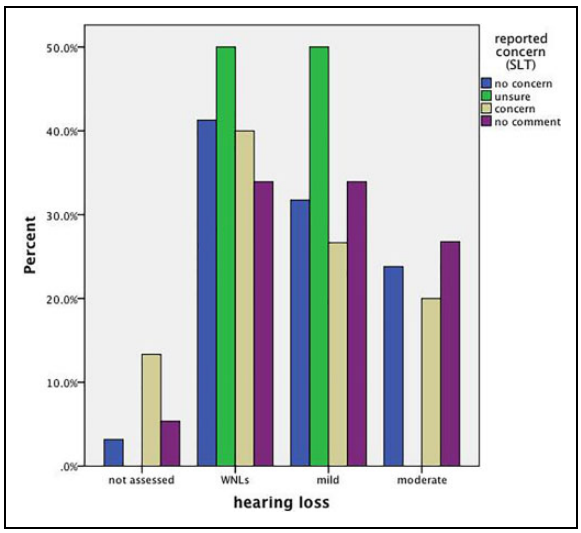

Figure 3. Percentage of parent concern reported to speech and language therapist (SLT) in relation to hearing test findings.

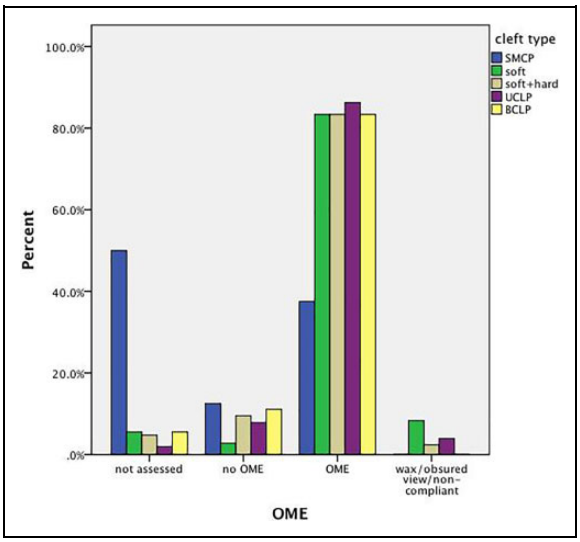

Figure 4. Percentage of otitis media with effusion (OME) found by cleft type.

Of the parents who reported no concern to SLT, 26 (41.3%) of 63 of infants had hearing WNL suggesting accurate parental judgment (Figure 3). However, almost a third of parents who reported no concern were found to have infants with a mild loss (20/63; 31.7%) and almost a quarter had infants with a moderate loss (15/63; 23.8%).

When concern was reported to SLT, the following presurgery hearing test reflected this concern in identifying a mild loss in 4 (26.7%) of 15 infants and a moderate loss in 3 (20%) of 15 infants. However, in 6 (40%) of 15 hearing was found to be WNL.

Consistency of Reporting Concern to ENT and SLT

Of the 24 (18.3%) of 131 parents who were concerned at the ENT consultation and the 15 (18.8%) of 80 parents reporting concern to the SLT, only 7 reported concern at both consultations. Similarly, of the 101 (77.1%) of 131 parents who reported no concern at the ENT consultation and of the 63 (78.8%) of 80 parents reporting no concern to the SLT, only 45 of these reported no concern to both. These findings should be viewed in the context of the percentage of documented concern reported to SLT (51.6%).

Otoscopy and Tympanometry Findings

Otoscopy. Of the 155 infants, 145 (93.5%) were assessed by otoscopic examination to establish the presence/absence of OME. Otitis media with effusion, in one or both ears, was found in 127 (81.9%) infants and only 12 (7.7%) had no signs of OME. Of the remaining 6 (3.9%) infants, the view was obscured either by wax or narrow ear canals or there was noncompliance with the examination.

Figure 4 shows the occurrence of OME by cleft type. Of the 127 (81.9%) infants with OME, there were no substantial differences with respect to cleft type with the exception of the SMCP group, all presented with the same degree of OME either unilaterally or bilaterally: soft (30/36; 83.3%), soft + hard (35/42; 83.3%), and BCLP (15/18; 83.3%), respectively. The UCLP group had marginally higher OME (44/51; 86.3%). Only 3 (37.5%) of 8 from the SMCP group presented with OME but 4 (50%) of 8 from the group were not assessed. Where OME was found, it presented bilaterally in most cases and unilaterally in only 17 (13.4%) of 127.

Tympanometry. Due to age at presurgery assessment, no infant had already received grommets and no eardrum perforations were recorded. Therefore abnormal tympanograms can be assumed to indicate the presence of OME. Overall, 131 (84.5%) infants had bilaterally abnormal tympanograms. One normal and one abnormal tympanogram were found in 13 (8.4%) of the infants and bilaterally normal tympanograms were found in 6 (3.9%). The data were not available for 5 (3.2%) infants. All cleft types, except the SMCP group, were similar in the proportions of bilateral abnormality ranging from 80.6% (29/36) in the soft group to 85.7% (36/42) in the soft + hard group (Figure 5).

Cleft type and degree of hearing loss. Compliance with sound field audiometry testing was high (148/155; 95.5%). Hearing WNL was found in 57 (38.5%) of 148 infants. Mild hearing loss was found in 53 (35.8%) of 148 infants and a moderate loss in 38 (25.7%) of 148. No infant had a severe loss and only one was identified with SNHL which was a moderate loss (from BCLP group).

Figure 6 shows the percentage of infants by hearing loss and cleft type. Of those with hearing WNL, the soft cleft group had slightly more hearing WNL compared with the soft + hard group. Similarly, the UCLP group had slightly more hearing WNL compared with the BCLP group.

There are more infants with a mild loss in the soft + hard group than those in the soft only group. Similarly, there are

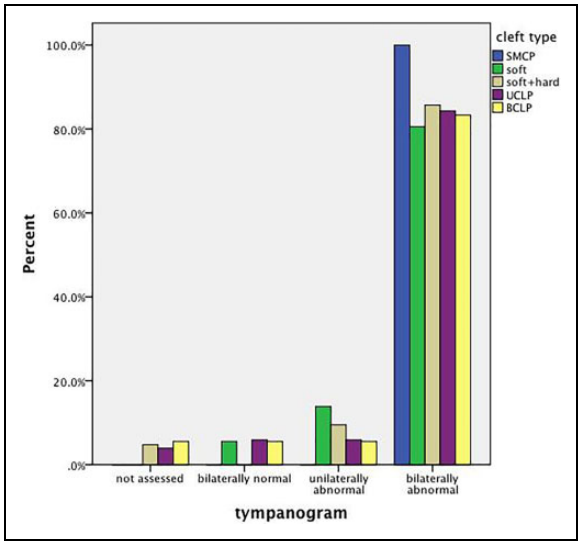

Figure 5. Percentage of infants with normal, and unilaterally and bilaterally abnormal tympanograms by cleft type.

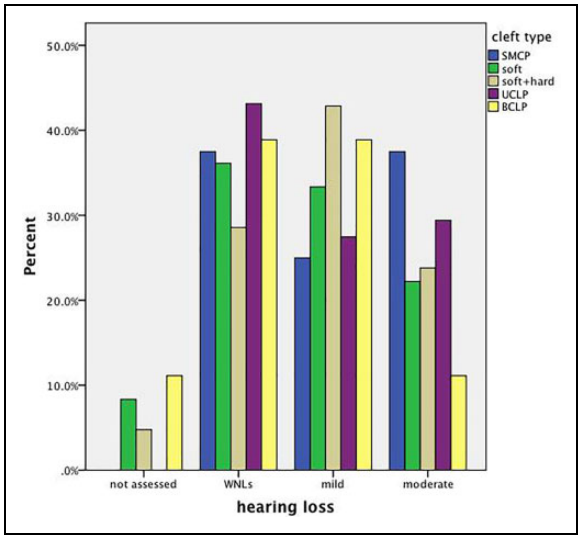

Figure 6. Percentage of hearing loss of infants by cleft type.

more infants with a mild loss in the BCLP group than in the UCLP group. Only 2 (25%) of 8 infants with SMCP had a mild loss.

The proportion of infants who presented with a moderate loss in the soft and soft + hard groups was similar. The SMCP group included the largest proportion of infants with a moderate hearing loss, but the group size is relatively small. The BCLP group contained the lowest proportion of infants with a moderate loss.

Discussion

Is Parental Concern About Their Infants' Hearing at Palate Repair Supported by Hearing Test Findings?

Documentation regarding concern (concern, no concern, or unsure) was found in the majority of ENT case notes (84.5%). This indicates that the ENT consultant consistently seeks parental opinion when considering management of possible hearing loss. Of the parents reporting no concern, approximately half (48.5%) did so when hearing was identified as WNL suggesting that these parents were accurate in their perception. Similarly, of the parents reporting concern, 79.2% of these infants presented with either a mild or a moderate loss. Only 2 infants whose parents reported concern had hearing WNL. These data indicate that when parents are concerned, there is usually a hearing loss present. The degree of loss needed to raise concern in this cohort is not crucial as almost as many concerned parents had infants with a mild loss as those with infants with a moderate loss. These findings support the view of Thompson and Thompson (1991) and Parving and Christensen (1992) who both found parents to be good at identifying hearing loss.

Overall, however, the concern reported to the ENT consultant at the time of hearing assessment is not consistently reflected by hearing assessment findings. The results suggest that over a third of parents were not aware of an identified loss in their infant. Further, of the 6 parents who were unsure about their infants hearing, 5 of these infants had a hearing loss: 2 with mild loss and 3 with moderate loss. Thus, severity of loss does not appear to increase parent's certainty regarding concern of infant hearing. The above findings are supported by the findings of Luthra et al. (2009) and D'Mello and Kumar (2007) who both reported a lack of parental awareness of hearing impairment in cleft palate.

Documentation regarding parental concern reported to SLT was found less frequently in SLT case notes compared with documentation found in ENT case notes. This may be because parental concern is a key factor in determining any potential intervention following the ENT consultation whereas hearing is just one aspect of the information covered during the SLT presurgery advice session.

Documentation found in SLT notes suggests that a substantial number of parents did not show an awareness of a loss that was subsequently identified, even when it was of moderate degree; when no parental concern was reported, almost one-third of infants presented with a mild loss and almost a quarter of infants presented with a moderate loss. In total, over half of infants whose parents explicitly report no concern were found to have a hearing loss. When concern was reported, 40% infants presented with hearing WNL suggesting that these parents were unduly concerned. This is in contrast to parents reporting concern to ENT where the majority of infants were found to have a loss to reflect this concern.

It is also evident from SLT case notes that parent perception can be accurately reflected in assessment findings. Of the parents reporting no concern, many parents (41.3%) had infants with hearing WNL. Similarly, when concern was reported a mild loss was identified in 26.7% infants and a moderate loss identified in 20% infants.

The results regarding consistency of reporting concern to both ENT and SLT revealed that only 7 of these parents reported concern at both appointments. This suggests that some

parents alter their perception of their infant's hearing. This may be due to the fluctuating nature of the infant's hearing loss or it may be a reflection of to whom the parents are reporting. It is also possible that the audiologist has separate documentation relating to parental concern about hearing.

Is Cleft Type Related to Middle Ear Findings and Degree of Hearing Loss at Time of Palate Repair?

Examination of this cohort reveals an overall incidence of OME of 81.9%. This high incidence is consistent with other studies, for example, Kwan et al. (2011) reported a 76.1% incidence of OME in cleft palate in the first 2 years of life. Varying incidence reports of OME are often explained by methodological differences. For example, due to resolution of OME with increasing age, participant age will influence reported incidence. Chu and McPherson (2005) found a lower rate of 10% but included a wide age range in their study (1-35 years). The present study assessed middle ear and hearing findings within a narrow age range using data from assessments most frequently occurring at 7 months of age (mean age: 10 months). Due to delayed diagnosis of some infants, particularly in the SMCP group, age at assessment ranged from 5 to 64 months. The overall incidence of OME should therefore be considered in the context of mean age and age range of each cleft type group. Incidence of OME may also be influenced by ethnic differences. Chen et al. (2012) reported 71.9% incidence in an Asian cleft population and concluded this comparatively lower rate could be due to ethnic variations between Eastern and Western populations. The cohort in the present study was primarily a European Caucasian group.

Incidence rates may also be influenced by diagnostic criteria and assessment method. For example, Chen et al. (2012) used tympanometry and myringotomy. The present study examined otoscopy, tympanometry, and hearing assessment findings secondary to OME. Otitis media with effusion was determined as present or absent, but in 6 cases, the view was either obscured by wax/narrow ear canals or the infant was noncompliant. These infants may have had OME and the use of myringotomy would have enabled assessment of this.

The SMCP group aside, the present study found similar rates of OME across cleft types. The comparatively low incidence of 3 (37.5%) of 8 OME in the SMCP group suggests that the least extensive cleft type is associated with a lower frequency of symptoms. However, robust conclusions regarding the SMCP group are not possible due to both small cohort size and increased mean age, reflecting later diagnosis in many cases. When OME was identified, it was found bilaterally in most cases (110/127, 86%), consistent with other reports (Kwan et al., 2011).

The tympanometry findings reveal similar results to otoscopic findings; overall, abnormal tympanograms were found in 84.5% infants. Khan et al. (2006) reported a lower incidence of 76.8% of abnormal tympanograms, but this can be explained by the wider age range investigated (1-20 years). Unlike Annigeri et al. (2012) and Handzic-Cuk et al. (2001), the present study did not identify the BCLP group as having a higher rate of abnormal tympanograms in comparison to other cleft types. Slightly more infants presented with abnormal tympanograms in the soft + hard group compared to the soft group. This particular finding supports the view that the more extensive the cleft, the more frequent the middle ear dysfunction. The fact that all 8 of the SMCP group presented with abnormal tympanograms was an unexpected finding in light of the mean age of this group and given the natural resolution of OME with increasing age. These findings contradict those of Zheng et al. (2009) who reported significantly lower abnormal tympanometry findings in an SMCP group in comparison to their other 4 cleft types. In the present study, as the submucous cleft was not yet repaired, it could be argued that middle ear dysfunction may persist until surgery attempts to restore normal anatomy and function.

The 155 infants analyzed were almost equally divided between those with hearing WNL and those with a mild hearing loss. Fewer infants were found to have a moderate loss. The finding that a mild loss is more common than a moderate loss in cleft palate is consistent with the wider literature (eg, Flynn et al., 2009). A limitation of comparing hearing loss by degree (eg, mild/moderate/severe) across studies is that different studies use different assessment methods and thresholds for defining loss (Ferguson et al., 2016). Also, some studies use headphones to obtain ear-specific data. Due to the young age of the present study's participants, headphone use was not appropriate and so ear-specific data were not obtained. The methods used (age-appropriate sound field audiometry) dictated the use of increased threshold levels. Such variations in methods and definitions of hearing loss make comparisons between studies challenging.

This study did not reveal that a particular type of cleft is more predisposing to mild or moderate hearing loss than another; the infants found to have mild and moderate loss are spread across all 5 cleft groups, and there is no clear relationship apparent between cleft type and severity of loss. An unexpected finding was that the SMCP group presented with the highest rate of moderate hearing loss (3/8, 37.5%), again contradicting the study by Zheng et al. (2009). However, it is difficult to view this finding as meaningful in the context of small participant numbers.

Strengths and Limitations

This study contributes a recent evaluation of parental judgment of infant hearing and one which is specifically related to the cleft population. This report also describes infants with different types of cleft palate at a young and narrow age range, when OME is reported to be most prevalent (Flynn et al., 2009).

The study's findings should be understood in the context of its limitations. There were some confounding factors that were not possible to control for. These included the nature of fluctuating hearing loss and timing of assessment points. It is likely that many infants will have had periods of persistent OME with associated hearing loss together with times when ears were well

ventilated with corresponding "normal" hearing. Therefore, the results are a reflection of the findings at the point of assessment rather than a constant presentation. Further, ENT and speech consultations did not occur at exactly the same time and so the gap between appointment times provides a feasible explanation as to why parents were not always consistent in their reporting of concern.

The universal neonatal hearing screen results (a pass/fail assessment offered to all newborns in the United Kingdom) may also have influenced parents reported concern. It would be of interest to explore any relationship between the outcome of the screen and subsequent parental concern.

Within each of the 5 cleft categories, there are variations in degree and severity of cleft. This variation may be important as the categories may not be as distinct as suggested when comparing the different cleft "types." In addition, despite the cohort spanning 5 years, case numbers remain small for some cleft types. Future prospective studies should aim to increase participant numbers to support statistical analysis. Similarly, due to the different mean age of the SMCP group compared with other cleft types, caution should be used when drawing comparisons with this group.

When describing middle ear presentation, infants were divided into those with and without OME based on ENT documentation. It is acknowledged that this binary categorization does not take into account degrees of OME that may have been observed and in some cases documented with qualitative comments. Finally, this study addressed the relationship between parental reported concern and hearing assessment findings but did not consider the relationship between reported concern and cleft type and middle ear status. Follow-up studies to address this would be of interest.

Conclusions

In this study, between one-third and one half of parents (depending to whom they are reporting) are unaware of a hearing loss that is subsequently identified. This reflects the wider literature regarding parental accuracy in identifying hearing loss. Although there are few recent studies and they generally not related specifically to the cleft population, the evidence suggests that parents are not accurate in their perception of their child's hearing when compared with assessment findings.

In examining this cohort, a clear relationship between cleft type and middle ear or hearing findings is not identified. Some findings suggest a tendency toward higher rates of middle ear dysfunction and hearing impairment in the more extensive cleft types. A larger study is needed to investigate this further.

In view of the finding that parental concern is not consistently accurately reflected by objective assessment, clinicians and SLTs in particular should be mindful that parents, while often accurate when reporting concern, are still frequently unaware of their infant's hearing loss. As it remains unclear whether cleft type has an influence on middle ear status or hearing, it is not recommended that speech and language advice related to hearing offered to families is tailored according to cleft type.

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