Closer to the truth on national fistula prevalence after unilateral complete cleft lip and palate repair? The Cleft Care UK study

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Abstract

Objectives: To 1) determine the prevalence of non-perialveolar palatal fistula up to age five following repair of unilateral cleft lip and palate (UCLP) in the United Kingdom (UK); 2) examine the association of palatoplasty techniques with fistula occurrence, and 3) describe the frequency of fistula repairs and their success.

Design: Cross-sectional study

Setting: All 11 centralized regional cleft centers in the UK.

Participants: 268 children born between 2005-2007 recruited by Cleft Care UK (CCUK), a nationwide cross-sectional study of all five-year-old children born with non-syndromic UCLP.

Main outcome measure: Non-perialveolar palatal fistula prevalence up to age five.

Results: Fistulas were found in 72 children (31.3%, 95% CI: 25.4 to 37.7%), and had no significant association with palate repair sequences. Twenty-four fistulas were repaired by age five, twelve of which had data showing 10 (83.3%) successful repairs.

Conclusion: The prevalence of non-perialveolar fistulas following primary palatoplasty of UCLP in the UK was higher than previously reported. This information should be part of the preoperative discussion with families. Prospective collection of the presence of fistulas will be necessary before we can associate the occurrence of fistulas with a surgeon, institution, surgical technique or protocol of care.

CPCJ Preset keywords: Nonsyndromic clefting, Hard palate, Soft palate, Palatoplasty, Surgical complications.

Introduction

Palatal fistula is a well-recognized complication following cleft palate repair. Small fistulas can lead to nasal regurgitation of fluids, while larger fistulas can cause problems with speech due to difficulties in articulation, audible nasal emission, and hypernasal resonance. The reported presence of fistulas after palate repair varies from 0% to 77.8% in the literature (Hardwicke et al. 2014; Bykowski et al. 2015). While the definition and functional impacts of a palatal fistula are well understood, there is no internationally standardized and validated methodology for the timing and recording of these assessments, nor is any assessment routinely and independently verified (Moar et al. 2016). Reporting bias is therefore likely to be high. Fistula prevalence is reported to be low in incomplete clefts of the soft palate, high in bilateral complete cleft palates, and so case mix is essential in comparing fistula prevalence (Hardwicke et al. 2014). Some studies also included intentional perialveolar fistulas while others did not, and some clinicians may ignore asymptomatic fistulas (Cohen et al. 1991).

Following the Clinical Standards Advisory Group (CSAG) recommendations at the turn of the century, all cleft treatments in the United Kingdom (UK) were centralized and mandatory audit standards established. The new standard requires a 5-year assessment in all cleft children (Sandy et al. 1998; Bearn et al. 2001; Sandy et al. 2001; Sell et al. 2001; Williams et al. 2001; John et al. 2006; Britton et al. 2014; Scott et al. 2014). Children with clefts access healthcare in one of the eleven designated cleft centers provided by the UK National Health Service, thus enjoy a comprehensive antenatal to adult service by multidisciplinary cleft teams.

The Cleft Care UK (CCUK) is a national cross-sectional survey of 5-year-old children with non-syndromic UCLP in the UK after service centralization (Persson et al. 2015). CCUK provides a unique opportunity to investigate fistula prevalence across all UK cleft centers. To date, this national prevalence is unknown since the CSAG study (Moar et al. 2016). Using CCUK data, our objectives were to determine the prevalence of non-perialveolar fistulas following palatoplasty

up to and at five years of age; to describe the association of palatoplasty techniques with the presence of fistulas and for those with fistulas, describe the frequency of fistula repair and its success.

Methods

Study design and sample

CCUK set out to recruit all 5-year-olds with non-syndromic UCLP born in the UK between 1 April 2005 and 31 March 2007 using comparable methods to the CSAG study (Sandy et al. 1998). Comprehensive information on the treatment and outcome of these 268 five-year-old children were collected. A full description of CCUK design, eligibility criteria, recruitment procedures, and data collection method for this study was published in detail previously (Persson et al. 2015). In brief, the study collected surgery methods (techniques, dates, complications) and routine clinical measures (speech recordings, hearing, photographs, models, oral health, psychosocial factors). Eligible children at age 5 attended research clinics in each participating center and were assessed independently by a local pediatric dentist and a local cleft surgeon. Participating dentists were calibrated by the British Association for the Study of Community Dentistry (BASCD). The dentists recorded their assessments as-is on the day, whereas the surgeons recorded based on their findings at the time and information collated from medical notes. The methodology for each clinical measure followed those of the earlier CSAG study.

Appendix I lists the CCUK data fields interrogated in this study. Dental and surgeons' forms have the same entry fields on the presence, location, and diagram drawing of any palatal fistula. The Surgeons' form also contained fields on dates and descriptions of palatoplasty and fistula surgery. Therefore data from the surgeons' forms and dental forms were utilized in different analyses as follows:

Data pooled from both surgeons' and dental forms: fistula prevalence following primary palate repair (up to age five).

Data from Surgeons' forms only: palatoplasty techniques; fistula repair surgery.

Data from dental forms only: the status of repaired fistulas at age five.

Definition of a fistula

For this study, a fistula was an unintentional connection of mucosal surfaces between the oral and nasal cavities arising after primary palatoplasty.

Fistula locations

Responses in the CCUK surgeon and dentist assessments included recordings of fistula presence, fistula locations (perialveolar region, hard palate, soft palate, uvula), and a diagram drawing (Appendix I). All of these fields were reviewed by both first and second surgeon authors to determine the presence of fistula and assign fistula location by Pittsburgh classification (Figure 1) (Smith et al. 2007). Any fistula of the hard palate recorded as post-alveolar was considered to be at the primary/secondary hard palate junction (Pittsburgh V). Further distinctions between Pittsburgh V and VI (lingual-alveolar) were made based on diagram drawings. Lingual-alveolar fistulas with extension into the hard palate involving less than 50% length of the primary palatal shelf were classed as Pittsburgh VI, as these are almost undoubtedly intentional fistulas staged for later repairs at the time of alveolar bone grafting (Cohen et al. 1991). A single long fistula may cross multiple Pittsburgh locations, and for Pittsburg classification only, it can be counted as a location more than once.

[Insert Figure 1.]

Pittsburgh I-V represented non-perialveolar fistulas. To accurately obtain their prevalence, fistulas were counted only once if they occurred anywhere in the secondary palate or incisive foramen. A few children had more than one non-perialveolar fistula, and these were noted but only counted as one fistula to calculate prevalence.

Fistula prevalence

After applying the fistula definition and Pittsburgh classification to the data, perialveolar fistulas (Pittsburgh VI and VII) were assumed to be intentional and therefore excluded from this study (Cohen et al. 1991). All mentions of palatal fistulas from here on refers to the non-perialveolar fistulas (Pittsburgh I-V) unless otherwise specified.

Dentists collected information on fistula prevalence at the audit clinic when the child was around five years of age. Surgeons entered information on fistulas present at and before the audit clinic. By pooling information from dentists and surgeons, we were able to obtain an improved estimate of fistula prevalence up to the age of five as missing or unclear fistula information in one data set could be checked and confirmed in the other. Records without information on fistula surgery were further excluded to help streamline data analysis on subsequent fistula surgery. This study defined a child as having had a fistula if either the dentist or surgeon reported the presence of a fistula at any time up to and included their CCUK assessment at age five. The finding is effectively the 'prevalence following primary palate repair,' and comparable to that defined by Bykowski et al. and Hardwicke et al. (Hardwicke et al. 2014; Bykowski et al. 2015).

Fistula surgery

Specific fields in the surgeons' forms recorded repairs made to fistulas before the age of five. When matched to data from dental examinations, which indicated the status of the repaired fistulas at age five, the proportion of successful fistula repairs was estimated.

Primary palatoplasty techniques

Due to limitation of the data collection to account for regional and individual surgeon variations on palatoplasty techniques, cleft repairs were broadly categorized by their repair stages as the surrogate measure for technique: 1) stage one: lip and hard palate repairs, followed by stage two: soft palate repair (Oslo type sequence); 2) stage one: lip repair,

followed by stage two: complete palate repair ('traditional' sequence); 3) stage one: lip and soft palate repairs, followed by stage two: hard palate repair (Schweckendiek type sequence); and 4) other palate repair sequences (Schweckendiek and Doz 1978; Fudalej et al. 2009). These were not restricted to the age schedule originally prescribed by any named sequence. Fistula occurrences were compared according to cleft repair sequences.

Surgeon and experience

The annual cohort size of new babies with cleft lip and palate for the UK cleft surgeons is approximately 35 to 55 per year (The cleft registry and audit network (CRANE) Report 2012). We compared each UK surgeon (anonymized) with the number of UCLP children they operated on and the fistula occurrence in those children.

Multiple deprivation percentile

In the UK the multiple deprivation percentile is a relative measure of social deprivation from 0 (most deprived) to 100 (least deprived). It is used to compare health outcomes against social circumstances. Its use and relevance in CCUK were previously reported by Smallridge et al. (Smallridge et al. 2017).

Statistical analysis

Participants were matched between surgeons' and dental forms by unique CCUK identifiers using FileMaker (FileMaker, Inc, Santa Clara, CA, USA). Fisher exact test (2-by-2 contingency table analysis) was used to compare the fistula prevalence of two groups, while the Fisher-Freeman-Halton tests compared between more than two groups. The results were expressed as 95% confidence intervals (CI) and *p*-values (Sterne and Davey Smith 2001). Stats Direct version 3.0 software (StatsDirect Ltd, Cheshire, United Kingdom) was employed except for logistic regression comparing between surgeons and experiences where Strata version 15.1 (StataCorp LLC, Texas, USA) was used. To estimate the variation in fistula occurrence between surgeons we used a mixed effects logistic regression model with surgeons as the random effect. The variance partition coefficient (VPC) was estimated - in our model this is equivalent to the intraclass correlation coefficient. VPCs can take values between 0 and 1. Values closer to

1 indicate more variation between surgeons. For example, a VPC of 0.2 indicates that surgeons could explain 20% of the variation in fistula prevalence.

Results

Sample description

CCUK identified 359 eligible children with UCLP, of which 85 failed to attend the assessments, and a further six declined to take part in the research. Thus 268 children were recruited to the CCUK cohort (Figure 2). Surgeons returned research forms on 251 (93.6%) children, of which 211/251 (84.0%) recorded fistula information. Dentists returned forms on 263 children (98.1%), of which 148/263 (56.3%) recorded fistula information. After pooling the surgeons' and dental data, fistula information up to age five was available for 251/268 (93.7%) children. A further 21 records without fistula surgery information were excluded, giving 230/268 (85.8%) children for fistula prevalence and fistula repair analyses.

Of the 251 children included in the pooled data, the median age was 5.5 years (IQR: 5.4-5.8), and 168 (66.9%) were male (Table 1). There was no evidence of a difference in age, sex, multiple deprivation percentile, frequency of left-sided cleft, and age at primary repair between the children included in the fistula analyses and those excluded due to missing data ($p>0.7$ for all comparisons).

[Insert Figure 2.]

[Insert Table 1.]

Fistula locations

Table 2 shows fistula locations by Pittsburgh classification. Some fistulas spanned more than one Pittsburgh location, and are counted for each location affected. With perialveolar fistulas (Pittsburgh VI/VII) excluded, fistulas most commonly involved the secondary hard palate (Pittsburgh IV), followed by the primary/secondary palate junction (Pittsburgh V)(Table 2). There was no uvular fistula.

[Insert Table 2.]

Fistula prevalence

Pittsburgh I-V were grouped to estimate the prevalence of non-perialveolar fistula up to age five, and per the methods, each child was counted once if a fistula involved any part of Pittsburgh I-V by location. In 230 children, surgeons and dentists reported a combined 72 cases with fistulas giving a prevalence of 31.3% (95% CI: 25.4 to 37.7%)(Figure 2), which is substantially higher than 17.9% (p =0.0057) found in the most recent systematic literature review (Hardwicke et al. 2014). Three (4.2%) of the 72 cases had two fistulas in the secondary palate. Perialveolar (intentional) fistula was present in 71/230 cases (30.9%). 18 of the 72 non-perialveolar fistulas had a connection to a peri-alveolar fistula (25.0%).

Fistula surgery

Repairs were attempted in 24 (33.3%) of these 72 fistulas, indicating 10.4% of the 230 children had a fistula surgery by age five. Of the 24 fistulas repaired, 12 had fistula status recorded by dental assessment at age five to show two fistulas remained open, indicating a potential 83.3% success in fistula repairs.

Primary palatoplasty techniques

One hundred sixty-three children had complete information on palatoplasty sequences and fistulas (Table 3). Of these, 69.3% repairs used the Oslo type sequence, while 21.5% repairs used the traditional repair sequence. All other sequences accounted for 9.2% of the cases.

When comparing fistula occurrences with repair sequences using the pooled data (Table 3), the Oslo type sequence achieved the lowest fistula prevalence (30.1%; 95% CI = 21.8-39.4%). The fistula prevalence was higher following the traditional and Schweckendiek sequences (37.1% and 62.5% respectively), although there was no statistical evidence of a difference in

prevalence between sequences (p = 0.25), fistula distributions by Pittsburgh types were similar between repair sequences (p = 0.73).

[Insert Table 3.]

Surgeon and experience

CCUK did not differentiate between cleft surgeons or cleft trainee surgeons who did one or more of the stages of the palate repairs. Because CCUK recorded different repair sequences (lip and hard palate first, or lip and soft palate first or lip only first), the operative surgeon for the final palate closure procedure is the only surgeon against whom the prevalence of fistula is reported. The variance partition coefficient (VPC) was 0.24, (95%CI =0.09-0.49) for the 29 surgeons who performed the final palate closure operation indicating that 24% of the variation in fistula prevalence may be attributed to the surgeon. This estimate was not adjusted for differences between surgeons such as surgical sequencing, or seniority of the surgeon. Figure 3 shows the 29 surgeons' individual predicted fistula prevalence estimated by a mixed effects logistic regression (maximum likelihood estimation, adaptive quadrature. Strata version 15.1). The result suggests only one of the 29 surgeons is an outlier for high fistula occurrence.

[Insert Figure 3.]

Discussion

To the best of our knowledge, this is the first report of a UK national fistula prevalence since the CSAG study and regional centralization of the cleft services. At 31.3% the fistula prevalence after primary palatoplasty of UCLP was substantially higher than 17.9% estimated in the most recent systematic literature review (Hardwicke et al. 2014). It was nonetheless reassuring that attempts to repair fistulas appeared mostly successful. The finding of this study is likely to be a more accurate representation of the UK fistula prevalence than from available literature due to design of the CCUK that resulted in a lower loss to follow-up rate, a double fistula screening process (surgeon and dentist), and a more inclusive fistula definition (symptomatic and non-symptomatic).

Two contemporary analyses by Bykowski et al. (a meta-analysis) and Hardwicke et al. (a systematic review) attempted to review all published studies since the year 2000 to estimate fistulas prevalence following primary palatoplasty (Hardwicke et al. 2014; Bykowski et al. 2015). In the main, much of the data was derived from retrospective studies with small numbers (Hardwicke et al. 2014). There were only two randomized and two prospective studies that were considered higher quality (Helling et al. 2006; Richard et al. 2006; Williams et al. 2011; Annigeri et al. 2012). Fistula presence following palatoplasty in UCLP children varied from 17% to 41.6% in these four studies (Level of evidence I-III), overlapping with estimates from the present study. Despite very different surgery timings, techniques and follow up protocols from the studies reviewed, Bykowski et al. estimated an overall presence of fistulas following palate repair as 4.9%. In slightly more detail, Hardwicke et al. concluded 8.6% overall and 17.9% for UCLP. Hardwicke conceded that his figures were inferred from reported data of undetermined reliability, especially as small or asymptomatic fistulas may not have been acknowledged. It is therefore unlikely that these represent accurate figures for the presence of fistulas following palate repair (Hardwicke et al. 2014).

CSAG in 1998 reported the first UK nationwide fistula prevalence on 239 five-year-old and 218 twelve-year-old children with UCLP, which were 39% and 10% respectively (Sandy et al. 1998). The present study suggests that fistula prevalence up to age five had improved since the CSAG, although CSAG did not assess a fistula to be intentional (perialveolar) or not, or if any previous fistula had been repaired (Williams et al. 2001). More recent fistula information in the UK has not been available beyond regional audit forums. Sommerlad in his UK personal series of UCLP cases reported only symptomatic fistulas repaired in 14% (11 of 80) children following Oslo staging and radical intravelar veloplasty (Sommerlad 2003). A retrospective UCLP case note review by six northern UK cleft units (NorCleft) reported post-incisive fistula prevalence at age three to be 11% (Moar et al. 2016). While the results from these two studies cannot be extrapolated to a national average, both of their estimates were far lower than the finding by the present study.

The CCUK study recruited 74.7% of the eligible nonsyndromic UCLP children, and that was similar to the 73% achieved in the original CSAG study (Persson et al. 2015). A similar recruitment rate suggested a similar sampling bias such as from families who refused to participate for reasons including international migration. Different fistula response rates between surgeons and pediatric dentists also indicated possible observer bias. Though CCUK required surgeons to collate historical information from the medical files in addition to clinical findings on the day of the research clinic, whereas the dentists were not, this could contribute to the differences in fistula reportings between the two specialties.

Inconsistent fistula definitions were an issue in the earlier literature especially concerning the separation of intentional (perialveolar) fistulas from the non-intentional post-incisive variety (Cohen et al. 1991; Emory et al. 1997; Muzaffar et al. 2001). While CCUK did not provide an exact definition for and how to assess for a fistula, it is widely accepted in the UK that a fistula is an unintentional connection of mucosal surfaces between the oral and nasal cavities arising after primary palatoplasty. The same definition applied in this study and is comparable to the

definitions adopted by the two previous meta-analysis and systematic reviews (Hardwicke et al. 2014; Bykowski et al. 2015). In keeping with this definition and with Cohen et al., all perialveolar fistulas were assumed most likely intentional and excluded from our analyses. Most publications of fistula rates are from individual surgeons or an institution's results, where those authors set their own definition and assessment method of a fistula. While we have not improved on this methodology, we have increased the assessment age to five years with two independent clinicians using the same data recording design.

A minority of early postoperative wound breakdowns appearing as fistulas may spontaneously heal if small (Richard et al. 2006). This led to criticism of some earlier studies where follow-ups were short or inconsistent. The follow-up in this study was 5-years, which was expected to negate any temporal effect of spontaneous fistula closure and give a realistic measure of fistula prevalence.

Pin-hole or very small fistulas may remain asymptomatic and undocumented until mentioned by parents at age 3-5. Alternatively, some surgeons will purposively leave even visible anterior fistulas, which may be intentional, for closure until the time of the secondary bone grafting procedure. So it is perhaps only the larger fistulas impacting on speech or intrusive nasal regurgitation that will be recorded and repaired by some surgeons because only a third of all fistulas in CCUK proceeded to repair. Other potential factors not considered include the surgeon's preference and parental pressure to close the fistula. This study recorded all the fistulas found by two different specialties (surgeon and dentist), and it is clear that the UK outcomes are higher in fistula numbers by this cumulative experience of observation over five years of follow-up. It is this unpublished cumulative experience of fistula documentation that is probably not discovered by the previous systematic reviews. It may be disheartening for the UK cleft community to recognize this result, but it is the true position from which to seek to improve outcomes in the future.

A study in the USA recently attempted a national estimate of the number of revision cleft palate surgeries from the Kids' inpatient database based on ICD-9 codes (Thompson et al. 2017). It found 2000 (36.8%) of 5431 hospital admissions were cleft palate revision surgeries in 2009. With similar ratios for the years 2003 and 2006, there is a consistent practice of about 58% of all cleft palate repairs having a revision surgery between age 3 and age 20. Although it was not possible to separate fistula repair from speech surgery by ICD-9, the authors were able to exclude alveolar bone graft and lip/nose surgeries (personal communication). Assuming approximately 20-30% will be for speech, it seems likely that at least another 20-30 % will be for fistulas. If these estimates are accurate, the USA also has a fistula repair requirement that is not dissimilar to the UK.

The literature had reported the effect of a surgeon's skill in cleft surgery on fistula prevalence. While a minority of studies suggested experience level to be independent of fistula prevalence (Muzaffar et al. 2001; Rohrich and Gosman 2004), most studies supported the notion that less experience correlated with higher fistula encounters (Cohen et al. 1991; Emory et al. 1997; Losee et al. 2008; Yong et al. 2010; Losken et al. 2011; Amirize et al. 2017). Sitzman et al. recently showed a five-fold variation among surgeons and hospitals in the use of secondary surgery for cleft palates in the USA, and the most attributable factors in the delivery of care were unexplained differences amongst surgeons and hospitals themselves (Sitzman et al. 2018b). That the surgeon is an important determinant of fistula prevalence is confirmed by the 24% variation in observed fistula prevalence in our study. Only one surgeon was an outlier for high fistula prevalence amongst 29 surgeons, and thus unlikely to be the sole cause of the high national fistula prevalence. Interestingly Sitzman et al. also suggests a four-fold hazard ratio of a need for secondary surgery in children who have their palate repaired before aged nine months and that may be true for many of the UK children (Sitzman et al. 2018b).

Limitations

The assignment of Pittsburgh classification to fistulas was essential for reducing ambiguity in fistula locations and facilitated ordinal analysis. Its original publication acknowledged challenges in distinguishing between Pittsburgh IV and V, due in part to unclear medical documentation (Smith et al. 2007). The reliability of the Pittsburgh recordings by different surgeons has recently been assessed by the Americleft task force surgeon subgroup, and found to be reliable when assessing a fistula broadly within the secondary palate but less reliable for specific zones (Sitzman et al. 2018a). The crossover from the primary to the secondary palate is less clear cut in the presence of a repaired UCLP. This study encountered similar challenges requiring the careful separation of Pittsburgh VI from V, and 25% of non-perialveolar fistulas did have a connection to a perialveolar fistula. Many surgeons would not plan for an intentional fistula to be this far posterior, but it remains possible that some surgeons do and thus a quarter of the reported non-intentional fistulas in this study could be considered to be intentional by some.

UCLP repair techniques and cleft severity are suspected of influencing fistula prevalence (de S Amaratunga 1988; Rohrich and Gosman 2004; Parwaz et al. 2009; Landheer et al. 2010). Surgical techniques, in general, were difficult to compare as often a named technique was not performed as originally described. In order to improve specificity when assessing for a potential cause and effect relationship between repair techniques and fistulas, it was more feasible to compare the effect of techniques by grouping them according to repair stages. Despite a trend that the Oslo sequence produced the lowest fistula prevalence while Schweckendiek the highest, there was no statistical difference (p = 0.25). Some subgroups in the comparison did have small numbers. Sequences with higher fistula prevalence can be a reflection of greater cleft width and the technical approach adopted to repair it. However as CCUK data did not include cleft width, it was not possible to study this.

Anterior-posterior descriptions of cleft severity not limited to the LAHSAL system (a modified LAHSHAL classification) are already routine practice in the UK, but there is no widely established method or practice to record cleft width (Kriens 1989; Shah et al. 2011). Analyses of fistulas by size, site, and symptoms of speech and or fluid/food regurgitation, in the context of the patient experience, is also in need of better definition and methodology of data collection. Cleft surgeons and their institutions should work towards unifying these concepts and prospectively audit their fistula outcomes.

The findings in this study were based on results no more than a decade on from service improvements owing to the CSAG report and will benefit from further validation by prospective studies on a similar national scale to evaluate continuing improvements in cleft care, of which fistula prevalence is one measure as will be patient-centered outcomes.

Conclusion

In a centralized multi-disciplinary service that had resulted in improved outcomes for facial growth, speech, and facial aesthetics, the prevalence of fistulas following UCLP repair is high. While a third of UCLP children with fistula had fistula surgery, it seems likely that the majority of the non-operated fistulas are small and without functional impairment at age five. Surgeons should explain the frequency of non-perialveolar fistulas following primary palatoplasty to the families and the findings of this study taken into account. The association of fistulas with palatoplasty techniques and subsequent workload warrants further investigation as do the effect of cleft width on fistula prevalence and the impact of fistulas on subsequent speech.

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