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ORIGINAL ARTICLE

A Cross-Sectional Cohort Study of Speech in Five-Year-Olds With Cleft Palate ± Lip to Support Development of National Audit Standards

BENCHMARKING SPEECH STANDARDS IN THE UNITED KINGDOM

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Objective: **To develop national standards for speech outcomes and processes of care for children with cleft palate ± lip and to test the standards using national data.**

Design, Setting, and Participants: **In this large, multicenter, prospective cohort study, speech recordings of 1110 five-year-olds with cleft palate involvement (born 2001 to 2003) were collected by 12 cleft centers in Great Britain and Ireland. Recordings were analyzed by consensus by specialist speech and language therapists using the Cleft Audit Protocol for Speech–Augmented. Results were benchmarked against evidence-based process and speech outcome standards and statistical analysis undertaken.**

Results: **From the 1110 children audited, 48% (530) had speech within the normal range. This was not significantly different from the agreed standard of 50% ($P = .20$, CI = 45–50%). Sixty-six percent (734) had speech with no evidence of structurally related speech problems or history of speech-related secondary surgery. This was significantly below the standard of 70% ($P = .007$, CI = 62–69%). Sixty percent (666) had no serious cleft-related articulation errors. This was significantly better than the agreed standard of 50% ($P < .001$, CI = 67–73%). More than 80% of 2-year-olds received a specialist speech and language assessment against a benchmark of 100%.**

Conclusions: **Developing standards has facilitated more meaningful reporting of speech outcomes and treatment processes. Evidence-based standards were defined and extensively tested, enabling centers to compare their performance with national trends. One 5-year outcome standard was achievable; the other two standards will require modification through the mandatory annual national audit program.**

KEY WORDS: *cleft palate, audit, standards, speech outcomes, process, velopharyngeal insufficiency, articulation*

Approximately 870 babies (1/700) are born annually in England, Wales, and Northern Ireland with cleft lip and/or palate (CRANE Database, 2012). Cleft care is a complex, long-term multidisciplinary intervention involving surgery, audiology, ENT, orthodontics, nursing, dental health, clinical psychology, and speech and language therapy (SLT). Primary outcomes include speech, facial growth, and psychological well-being. Children with cleft palate ± lip (CP±L) are at high risk of developing speech difficulties requiring SLT (Vallino-Napoli, 2011), which may have long-term consequences for literacy and psychosocial development (Stothard et al., 1998; Johnson et al., 1999; McCormack et al., 2009; Chapman, 2011; Richman et al., 2012).

The Clinical Standards Advisory Group investigation of cleft services in the United Kingdom commissioned by the government (Department of Health, 1998a) found

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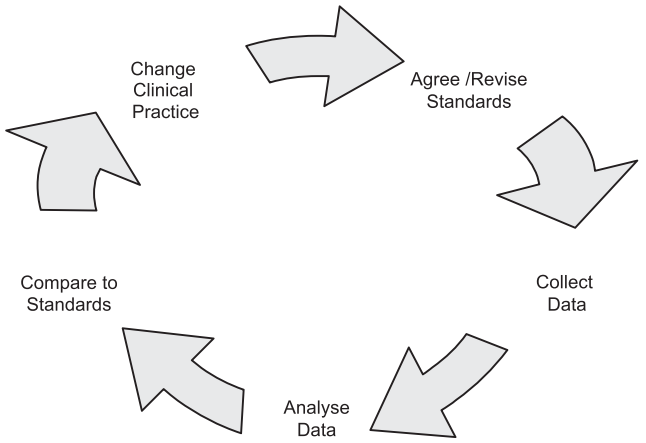

FIGURE 1 Audit cycle. Adapted from The National Institute of Clinical Excellence Guidelines (NICE; 2002).

"poor standards of care and poor clinical outcomes" including disappointing speech outcomes (Sell et al., 2001) compared with centers elsewhere in Europe. This resulted in the reorganization and centralization of cleft care. There were 57, predominantly low-volume units, offering primary cleft surgery in 1996. There are now 10 Regional Cleft Services in England, Wales, and Northern Ireland, with most treating more than 60 primary cases per year (CRANE Database, 2012) plus a managed clinical network in Scotland (CLEFTSiS, 2013). Recommendations for the new services were driven by the need to improve standards of care, with regular audit identified as a mandatory requirement (Department of Health, 1998a, 1998b). The recommendations stated that audit records including speech recordings, orthodontic models, photographs, dental records, and audiograms should be taken at ages 5, 10, and 15 or 18 years depending on cleft type (Craniofacial Society of Great Britain and Ireland [CFSGBI], 2005). Centers gradually began to collect audit records, but there was no process to evaluate these records systematically. John et al. (2006) recognized the "urgent need to introduce a continuous program of inter-center audit of cleft speech outcomes for all cleft patients in the UK" (p. 279).

The National Institute for Clinical Excellence (NICE; 2002) defined audit as "a quality improvement cycle which seeks to improve patient care through a systematic process of establishing best practice, measuring care against explicit criteria, taking action to improve care and monitoring to sustain improvement" (p. 5). They recommended four stages: selecting criteria, measuring performance against criteria, making improvements, and sustaining improvement. Each cycle aspires to a higher level of quality (Fig. 1). The Health Care Quality Improvement Partnership (2009) updated guidance on best practice in audit using a similar four-stage cycle. Audit is intimately intertwined with evaluation and research, both in definition and implementation. Audit may generate the need for service evaluation and research or vice versa. Interestingly, audit is frequently interpreted as meaning either "monitoring" or "research," and the stages of setting standards and making improvements are often overlooked (Ovretveit, 1998, p. 13; Bowling, 2009, p. 9).

The National Institute for Clinical Excellence (NICE) (2002) guidelines outlined two types of standards: process standards, which are explicit statements of how care is provided, and outcome standards, which measure the patient's physical or behavioral response to an intervention. NICE (2002) recommended that standards should be relevant, measurable, and evidence based and lead to improvements in care. Standards should be derived methodically using clinical practice guidelines, systematic literature reviews, professional consensus, and user involvement (Health Care Quality Improvement Partnership, 2009).

Although the Cleft Audit Protocol for Speech–Augmented (CAPS-A) was developed as an assessment tool for audit studies of speech (John et al., 2006), there were no explicit standards against which speech outcomes and treatment processes could be compared. John et al. (2006) suggested that the availability of CAPS-A as a "robust audit measure will enable standards to be set for cleft speech outcomes in the UK and Ireland" (p. 280).

There was professional consensus that speech outcome standards should initially focus on age 5, when children begin statutory education in the United Kingdom. This is also the earliest nationally agreed audit point minimizing the length of the audit loop (CFSGBI, 2005). There is strong evidence that maximizing communication skills before starting school is important socially, emotionally, and educationally and may have long-term socioeconomic consequences (Bishop and Adams, 1990; Stackhouse and Wells, 1997; Stothard et al., 1998; Johnson et al., 1999; Nathan et al., 2004, p. 17; Bowen, 2009, p. 73; McCormack et al., 2009; Marsh et al., 2010; Harulow, 2011).

The evaluation and comparison of speech outcomes to date are complicated by differing or flawed methodologies, and the lack of robust published studies reporting speech outcomes in cleft care is well documented (Peterson-Falzone, 1996; Lohmander and Olsson, 2004; John et al., 2006; Persson et al., 2006; Lohmander, 2011; Kummer et al., 2012). Lohmander and Olsson (2004) critiqued 88 papers reporting speech outcomes and found major concerns regarding the method for speech sample collection, documentation, and analysis, making comparison of published results difficult. Flaws included lack of information about ages at speech assessment, who conducted the assessment, the nature of the speech sample, and rater reliability. Similarly, Kummer et al. (2012) reported variability in methodology and inconsistency in success criteria when reporting outcomes for secondary speech surgery in the United States.

Experts have argued that perceptual studies of cleft palate speech, whether for audit or research purposes,

should be based on robust listening procedures (Peterson-Falzone, 1996; Sommerlad et al., 2002; Lohmander and Olsson, 2004; Sell, 2005; Brunnegard and Lohmander, 2007). This includes the following:

- Ratings from multiple listeners, who are trained and experienced SLTs
- Recorded speech samples
- Randomized and blindly assessed speech samples
- A study group with a narrow age span
- Reporting inclusions and exclusions
- Reporting interrater and intrarater reliability
- Consideration of the speech sample and the parameters analyzed (Dalston et al., 1988; Kuehn and Moller, 2000; Henningsson et al., 2008)

Studies of speech outcomes have also been criticized for their small heterogeneous samples and retrospective cohort designs (Lohmander, 2011). They have frequently been descriptive studies with no point of comparison. Where they have compared groups, sample size calculations are rarely reported and studies frequently lack rigorous methodological design (Roberts et al., 1991; Bearn et al., 2001).

Lohmander (2011) undertook a comprehensive critical review of published studies reporting speech outcomes. She identified 34 studies of speech outcomes based on assessment from audio/video recordings published in English-language journals from 1984 to 2010. Lohmander (2011) cautions that although these are the most reliable studies of speech outcomes in children born with cleft palate to date, they are still characterized by considerable heterogeneity. She critiqued these studies in terms of their sample size, levels of evidence, and sources of bias and summarized the speech outcomes in terms of percentage of patients who did not exhibit hypernasality, nasal emission, or articulation errors. Lohmander (2011) cited 12 studies including speech outcomes on 5-year-olds (Lohmander-Agerskov et al., 1995; Lohmander-Agerskov, 1998; Sell et al., 2001; Lohmander et al., 2002; Persson et al., 2002; Pigott et al., 2002; Lohmander et al., 2006; Persson et al., 2006; Lohmander and Persson, 2008; Lohmander et al., 2009; Nyberg et al., 2010; Havstam and Lohmander, 2011). These 12 studies involved a total of 680 subjects, of which 464 (68%) in 9 studies had unilateral cleft lip and palate (UCLP), 198 (29%) in 6 studies had cleft palate only (CPO), and only 18 cases (2.6%) in 3 studies had bilateral cleft lip and palate (BCLP). Of the 12 studies, 9 included fewer than 50 cases, and many suffered from selection or exclusion bias. The largest blinded study of speech outcomes to date and the only study involving more than 100 cases was conducted by Sell et al. (2001) on 238 five-year-olds with UCLP as part of the Clinical Standards Advisory Group Study (Department of Health, 1998a).

Speech outcomes for children with a cleft palate have been broadly described in three areas: (1) normal speech, (2) structurally related speech disorders, and (3) cleft-related articulation difficulties. Achieving normal speech by statutory school age is an important goal for the child, their family, and the cleft team (Kuehn and Moller, 2000). Normal speech is also the goal of secondary speech surgery (Kummer et al., 2012). Structural anomalies, such as velopharyngeal insufficiency (VPI) and fistulae, are common complications of primary cleft surgery. When this occurs, speech is often characterized by one or more of the following: hypernasality, nasal emission, nasal turbulence, and the passive or obligatory articulation patterns of weak nasalized consonants and nasal substitutions. Secondary speech surgery is frequently advised (Kummer et al., 2012). Finally, cleft-related articulation difficulties include a range of distinctive speech characteristics frequently associated with a history of cleft palate. Many of these, such as glottal and pharyngeal substitutions, may be the result of early mislearning and may persist despite the physical ability for adequate velopharyngeal closure. These are sometimes described as compensatory, maladaptive, or active characteristics and are distinguished from the passive or obligatory characteristics that result directly from structural problems with the articulatory or velopharyngeal mechanism (Harding and Grunwell, 1998; Peterson-Falzone et al., 2001). The speech outcome standards therefore focused on these three areas.

The two aims of the study were to develop national standards for speech outcomes and processes of care for children born with CP±L and to pilot the audit process by benchmarking against the standards using national data.

Method

Phase 1: Development of the National Standards for Speech

A working group of five British SLTs who were specialists in cleft and related disorders was established to develop process and outcome standards which were realistic, inclusive, measurable, and evidence based for children born with CP±L. Standards were drafted and then agreed upon following consultation with two key stakeholder groups (surgeons and specialist SLTs).

Process standards were developed to define when children with CP±L should have their first comprehensive assessment with a specialist SLT and when they should have speech records taken. These standards were based on clinical guidelines (Department of Health, 1998b; Royal College of Speech and Language Therapists, 2006) and are shown in Table 1.

Speech outcome standards were defined following a literature review. Of the 35 published studies reviewed that reported speech outcomes, ten studies were identified which reported speech outcomes from audio recordings at age 5 to 6 years following primary surgery, and a further four were cited in the critical review by Lohmander (2011). A summary of these 14 papers

TABLE 1 Summary of Results for Process Standards

Process Standards	*2001*	*2002*	*2003*
1. 100% of children with cleft palate (±cleft lip/alveolus) who are eligible are offered assessment by a specialist SLT by 27 months and this offer documented	Four centers met standard Range: 42%, 67%, 83%, 85%, 98%, 98%, 100%, 100%, 100%, 100% United Kingdom Median: 98%	Five centers met standard Range: 69%, 70%, 81%, 81%, 94%, 97%, 100%, 100%, 100%, 100%, 100%, United Kingdom Median: 97%	Seven centers met standard Range: 40%, 79%, 90%, 99%, 100%, 100%, 100%, 100%, 100%, 100%, 100%, United Kingdom Median: 100%
2. Speech records are taken in line with national audit recommendations and reported locally and nationally for all nonsyndromic children with UCLP, BCLP, or isolated CP who are able to complete audit. Any exclusions are reported with reasons	Six centers reported more than 50% eligible cases	12 centers met standard Nine centers reported more than 50% eligible cases	12 centers met the standard 11 centers reported more than 50% eligible cases
3a. All audit recordings are analyzed by consensus by at least 2 CAPS-A–trained listeners	Inconclusive	Eight centers met standard for 5-year-olds	10 centers met standard for 5-year-olds
3b. A minimum of 10 consecutive recordings per center involve a listener external to the center	Inconclusive	Six centers met standard	10 centers met standard
3c. All results are submitted to the U.K. national database for cleft (CRANE)	No centers met this standard as not mandatory until 2012–2013	No centers met this standard as not mandatory until 2012–2013	No centers met this standard as not mandatory until 2012–2013

which includes the 12 reviewed by Lohmander (2011) is provided in the appendix. The outcomes for resonance/VPI and articulation difficulties were collated and used to provide evidence for drafting the three speech outcome standards focused on achieving "normal" speech (outcome standard 1), the presence of speech difficulties which are likely to be the result of existing or previous structural anomalies (outcome standard 2), and the presence of cleft-related articulation difficulties (outcome standard 3). The perceptual speech criteria for the speech outcome standards were defined using the CAPS-A audit tool (John et al., 2006).

The three speech outcome standards, their definitions using CAPS-A, and the relevant literature are described in Table 2.

Phase 2: Benchmarking Against Speech Outcome and Process Standards

Benchmarking processes of care and speech outcomes against the agreed standards was undertaken by the SLT teams in the 12 regional cleft centers across Great Britain and Ireland for children with cleft palate involvement born between January 2001 and December 2003 (see the Acknowledgments section for centers). The initial pilot used speech recordings collected at age 5 years for children born in 2001. After reviewing these national data, further speech recordings were collected at age 5 years for children born in 2002 and 2003. One center withdrew for 2003 data.

The specialist SLTs collected and analyzed audio and video speech recordings according to the agreed national protocol (Sell et al., 2009) on a consecutive series of nonsyndromic children aged between 5;0 and 5;11 with cleft palate involvement (UCLP, BCLP, CPO) who were able to complete the assessment. Speech samples included conversation, rote speech, and sentence repetition. Each speech recording was analyzed by consensus by at least two (usually three) CAPS-A–trained SLTs from each center using the CAPS-A tool. Interrater and intrarater reliability of trained listeners using the tool had been previously established during the CAPS-A training program (Sell et al., 2009). An external listener was recruited by each center for at least 10 cases (approximately 25%) per year to ensure reliability and minimize listener drift. Results were benchmarked against the standards. Each regional SLT team collated their own data, calculated the raw scores and percentages for their center for each standard, and submitted these data centrally to the first author.

Children were excluded if they had a diagnosed syndrome, had a submucous cleft palate, had transferred centers between birth and their 5-year speech assessment, or were unable to comply with the assessment process. Children with Pierre Robin Se-

TABLE 2 Speech Outcome Standards, Their Definitions, and Rationales

Speech Outcome Standard 1: "*By 5-5;11 years over 50% of children with CP±L will have speech within the normal range.*"
Definition: Children achieve this standard if they are rated on CAPS-A as having normal resonance (0) or borderline-minimal hypernasality (1), nasal airflow rated as absent or occasionally heard on pressure consonants (0/1) and no cleft-related articulation errors other than dentalization, or one or two consonants affected by lateralization, palatalization, or double articulation involving velars (i.e., a green profile on CAPS-A). **Rationale:** From the literature, it was difficult to identify the proportion of children with cleft palate who have normal speech by age 5 years, not least because there is no single agreed upon definition of "normal speech." Lohmander (2011, p. 68) concluded that by age 4 to 5 years, 60% to 70% of children could be expected to have normal speech, although none of the studies reviewed specifically reported this parameter. Sell et al. (2001) reported that 49% of their cohort had intelligibility judged as normal (19%) or not different enough to provoke comment (30%). Based on this evidence, approximately 50% of children born with a cleft palate were expected to have normal speech by age 5, as reflected in the standard as compared with 85% to 90% of children without a cleft (Enderby and Davies, 1989; Johnson et al., 1999; Law et al., 2000; Department for Education, 2008).
Speech Outcome Standard 2a: "*By 5-5;11 years over 70% children with CP±L have speech with no evidence of a structurally related problem and have not had velopharyngeal surgery or fistula repair for speech.*"
Definition: Children achieve this standard if they have had no velopharyngeal revision surgery or fistula repair and on CAPS-A analysis they have hypernasality rated as absent or borderline (0/1), nasal airflow rated as absent or occasionally heard (0/1), and no passive cleft speech characteristics (John et al., 2006). **Rationale:** The term *structurally related speech problems* was used to describe hypernasality, nasal airflow, and those articulation difficulties that are likely to be the result of VPI or symptomatic fistulae often described as passive (Harding and Grunwell, 1998; John et al., 2006) or obligatory errors (Peterson-Falzone et al., 2005; Kummer, 2008). Prevalence figures quoted in critical reviews of the literature for VPI were used to inform this standard. These vary considerably. This variation may reflect real differences in treatment outcomes or differences in the definition of VPI, the age at which it is measured, and the assessment criteria used. Morris (1973) concluded that a VPI rate of 25% was typical following primary surgery, Enderby and Emerson (1995) reported VPI prevalence ranging from 5% to 40%, and Peterson-Falzone (1990) found the average prevalence of VPI reported was 16% rising to 40% if "strict criteria" were applied. More recent reports estimate 20% to 43% of children have VPI after primary palate repair (Cable et al., 2004). Sell et al. (2001) found that 29% of their 5-year-old cohort with UCLP had inadequate primary repair in terms of velopharyngeal function, with 18% having consistent hypernasality and another 11% having undergone velopharyngeal revision surgery. In the studies identified by Lohmander (2011), a median of 70% (range, 50% to 94%) of cases were reported to have no hypernasality and 67% (range, 36% to 100%) no nasal emission at age 5 to 6 years. In conclusion, the review showed that 20% to 40% of children with CP±L develop speech patterns related to structural problems. Consequently, the standard initially developed stated that 70% of 5-year-olds might be expected to have no speech symptoms or history indicative of structurally related speech difficulties. In 2002, sections 2b-d were added to provide additional information by dividing the children with structurally related speech problems into three subgroups. These have not been based on a literature review but reflect expert opinion only and therefore do not constitute part of the standard. The subgroups are the following: 2b. Percentage of children who have had velopharyngeal surgery or fistula repair and now have no evidence of a structurally related speech problem 2c. Percentage of children who have had velopharyngeal surgery or fistula repair and still have speech indicative of a structurally related problem 2d. Percentage of children who have not had velopharyngeal surgery or fistula repair but their speech is indicative of a structurally related problem
Speech Outcome Standard 3: "*By 5-5;11 years over 50% of children with CP6L have no cleft-related articulation difficulties requiring SLT and/or surgery.*"
Definition: Children achieve this standard if their CAPS-A analysis identifies no cleft speech characteristics other than dentalization; less than two consonants affected by lateralization, palatalization, or double articulation; and weak and/or nasalized consonants or nasal realizations (i.e., a green profile on CAPS-A for all active cleft speech characteristics [John et al., 2006] with no backing to velar/uvular, pharyngeal realizations, glottal realizations, active nasal fricatives, or gliding of fricatives or affricates). **Rationale:** Articulation errors of interest were those distinctive speech characteristics frequently associated with a history of cleft palate and described by CAPS-A as anterior oral, posterior oral, and nonoral cleft speech characteristics. Many of these have been described as compensatory, maladaptive, or active errors (Harding and Grunwell, 1998; Kummer, 2008; Peterson-Falzone et al., 2001, 2005). The great variability in terminology used to describe cleft-related articulation errors in the literature made comparability across studies challenging (Sell, 2005). Across the studies reviewed by Lohmander (2011), a median of 74.5% (range, 23% to 100%) of children were reported to have no articulation errors when assessed at age 5 to 6 years. Lohmander (2011, p. 68) also highlighted a number of frequently referenced studies using live speech assessment or chart reviews, which reported articulation outcomes in 5-year-olds with articulation proficiency of 26% to 66% (Dalston, 1990; Peterson-Falzone, 1990; Park et al., 2000; Hardin-Jones and Jones, 2005). Chapman (1993) found 5 year old cleft and non-cleft groups were similar in their use of phonologic processes. Sell et al. (2001) found that 34% of 5-year-olds with UCLP had at least one serious articulation error. Similarly, Persson et al. (2002) found that 36% of 5-year-olds with CPO had velopharyngeal frication or glottal or retracted articulation. The literature review indicated that at least 50% of 5-year-olds with cleft palate should have no significant cleft-related articulation difficulties requiring therapy and/or surgery, and therefore the standard was set at this level.

TABLE 3 Summary of National Data and Exclusions

	2001, n (%)	*2002, n (%)*	*2003, n (%)*	*Total*
Total cohort (CP±L)	Not collected	712	835	
Excluded with syndromes	Not collected	51/712 (7%)	65/835 (7.7%)	
Excluded for other reasons (e.g., late diagnosis, learning difficulties)	Not collected	120/712 (17%)	144/835 (17%)	
Total excluded from cohort	Not collected	171/712 (24%)	209/835 (25%)	
Total eligible for audit	Not collected	541	626	
Eligible cases (%) not submitted (e.g., DNA, technical issues, local audit decisions)	Not collected	192/541 (35%)	195/626 (31%)	
UCLP cases submitted	88/314 (28%)	121/365 (33%)	152/431 (35%)	361 (32%)
BCLP cases submitted	44/314 (14%)	54/365 (14%)	44/431 (10%)	142 (13%)
CPO cases submitted	182/314 (58%)	190/365 (52%)	235/431 (54%)	607 (55%)
Total number of submitted/eligible (%)	314	365/541 (67%)	431/626 (69%)	1110

quence but no identified syndrome were included. Exclusions and missing data were reported by centers. Data were not collected on surgical type or timing.

Speech outcome data were collected for 1110 five-year-olds (Table 3). The numbers of cases submitted per year increased over the 3 years as the study gained momentum. Individual centers contributed a mean of 34 cases per year, and most provided more than 50% of their eligible cases in 2002 (9 of 12 centers) and 2003 (11 of 11 centers). Of the cases submitted, 361 (32%) had UCLP, 142 (13%) had BCLP, and 607 (55%) had CPO, reflecting the prevalence of cases with cleft palate involvement reported in England, Wales, and Northern Ireland over the past 10 years (CRANE Database, 2012).

Data on exclusions were collected systematically for 2002 and 2003 births. Despite variability between centers, exclusion rates for the whole cohort were remarkably similar for the 2 years, with 24% (171) for 2002 births and 25% (209) for 2003 births. Nationally, for both years, 7% to 8% of children were excluded because of a diagnosed syndrome and 17% for other reasons including learning difficulties and late diagnoses. Twenty syndromes were identified over the 3 years. A further 35% (192) of children eligible for audit from 2002 births and 31% (195) from 2003 births were not submitted for other reasons including failure to attend, lost or damaged tapes, lack of time to analyze, or local audit decisions, leaving 67% (349) submitted for 2002 births and 69% (431) for 2003 births.

In the United Kingdom, research ethics committee approval is not required for clinical audit studies such as this (National Patient Safety Agency, 2013). However, the principles outlined in the Declaration of Helsinki were followed.

Statistics

The data from the 12 centers were collated, national results calculated, and statistical analysis completed. Means, medians, and ranges were calculated. Assuming the null hypothesis that there was no difference between the standards and the speech outcomes and a 5% significance level ($P < .05$), the binomial probability test (two-tailed) was used to determine if there were significant differences between the standards and speech outcomes nationally for each year and for the cumulative outcome over 3 years. Ninety-five percent confidence intervals were calculated. Data were reported by center if more than 50% of eligible cases were submitted.

Results

Process Standards

The process standards data (Table 1) proved difficult to interpret owing to differences in reporting. Some centers reported using simple binary yes/no responses, while others reported raw data and percentages. More consistent reporting using raw data and percentages has since been implemented for future annual audits.

Over the 3 years, results for process standards have steadily improved. For example, in 2001, only six centers submitted data on more than 50% of eligible 5-year-olds, but by 2003, this had increased to 11 centers. Nationally, a median of 97% to 100% of children per center were offered a specialist SLT assessment before 27 months of age. Similarly, 349 of 541 (67%) of nonsyndromic children with CP±L in the United Kingdom were offered a 5-year speech assessment in line with national audit recommendations in 2002, rising to 431 of 626 (69%) in 2003.

Outcome Standards

The annual national speech outcomes and cumulative national outcomes for the years 2001 to 2003 for all three standards are summarized in Table 4. The outcomes for individual centers (indicated by letters) compared with the national outcomes for each standard are shown in Tables 5, 6, and 7, and Figures 2 to 4.

Outcome Standard 1 states that "*By 5-5;11 years over 50% of children with CP±L will have speech within the normal range.*" Table 5 and Figure 2 show that 48% ± 3% of all 1110 five-year-olds studied had speech within the normal range. Statistical analysis using a two-tailed test and rejecting the null hypothesis at the 5%

TABLE 4 Annual and Cumulative National Speech Outcomes†

Outcome Standard	*National Speech Outcomes (% ± CI)*			
	2001 (n = 314)	*2002 (n = 365)*	*2003 (n = 431)*	*Cumulative 2001–2003 (n = 1110)*
1. By 5-5;11 years, over 50% of children with CP±L will have speech within the normal range	160/314 (51% ± 5%), $P = .96$	169/365 (46% ± 5%), $P = .40$	201/431 (47% ± 4%), $P = .18$	530/1110 (48% ± 3%), $P = .20$
2a. By 5-5;11 years over 70% of children with CP±L will have speech with no evidence of a structurally related problem and have not had velopharyngeal surgery or fistula repair for speech	237/314 (75% ± 5%), *$P = .036$	230/365 (63% ± 5%), **$P = .007$	267/431 (62% ± 5%), **$P < .001$	734/1110 (66% ± 3%), **$P = .007$
3. By 5-5;11 years over 50% of children with CP±L will have no cleft-related articulation difficulties requiring SLT and or surgery	190/314 (60% ± 6%), **$P < .001$	209/365 (57% ± 5%), **$P = .0045$	267/431 (62% ± 5%), **$P < .001$	666/1110 (60% ± 3%), **$P < .001$

† CI = confidence interval based on a 95% confidence level.
Rejecting the null hypothesis that the national speech outcome is the same as the standard at the 5% significance level (*$P < .05$) and at the 1% significance level (**$P < .01$).

Standard 1 - Normal speech
Goal: 50%
range: 25%-77%
average: 48%

TABLE 5 Results Nationally and by Center for Speech Outcome Standard 1: "By 5-5;11 Years Over 50% of Children With CP±L Will Have Speech Within the Normal Range"†

Year	*National Outcome (% ±CI)*	*No. Centers Achieved*	*Range of Outcomes by Center (% ±CI), (**median**)*
2001 (n = 314)	51% ± 5%, $P = .96$	3/6	46% ± 17%, 47% ± 14%, ***48% ± 14%, 50% ± 22%***, 64% ± 21%, 68% ± 13%
2002 (n = 365)	46% ± 5%, $P = .4$	5/8	25% ± 12%, 32% ± 14%, 49% ± 18%, ***55% ± 15%, 57 ± 16%***, 60% ± 21%, 71% ± 19%, 77% ± 31%
2003 (n = 431)	47% ± 4%, $P = .18$	5/11	26% ± 16%, 31% ± 13%, 33% ± 13%, 41% ± 13%, 47% ± 14%, ***48% ± 18%***, 55% ± 12%, 57% ± 16%, 64% ± 22%, 65% ± 20%, 70% ± 35%
Total (N = 1110)	48% ± 3%, $P = .20$		

† CI = confidence interval based on a 95% confidence level.
Rejecting the null hypothesis that the national speech outcome is the same as the standard at the 5% significance level (*$P < .05$) and at the 1% significance level (**$P < .01$).

Standard 2a - SRSD (VPI)
Goal: 70%
range: 65-90%
average: 65%

TABLE 6 Results Nationally and by Center for Speech Outcome Standard 2a: "*By 5-5;11 years over 70% of children with CP±L will have speech with no evidence of a structurally related problem and have not had velopharyngeal surgery or fistula repair for speech*"†

Year	*National Outcome (% ±CI)*	*No. Centers Achieved*	*Range of Outcomes by Center (% ±CI), (median)*
2001 (n =314)	75% ± 5%, *$P = .036$	4/6	65% ± 18%, 67% ± 13%,***70% ± 14%, 73% ± 21%,*** 73% ± 23%, 98% ± 7%
2002 (n = 363)	63% ± 5%, **$P = .007$	3/8	48% ± 18%, 59% ± 16%, 59% ± 17%, ***62% ± 16%, 66% ± 15%,***77% ± 31%, 80% ± 21%, 86% ± 16%
2003 (n = 431)	62% ± 5%, **$P < .001$	3/11	40% ± 14%, 48% ± 14%, 56% ± 22%, 59% ± 15%, 61% ± 16%, ***64% ± 19%,*** 64% ± 23%, 66% ± 14%, 73% ± 12%, 84% ± 18%, 90% ± 35%
Total (N = 1110)	65% ± 3%, **$P = .007$		

† CI = confidence interval based on a 95% confidence level.
Rejecting the null hypothesis that the national speech outcome is the same as the standard at the 5% significance level (*$P < .05$) and at the 1% significance level (**$P < .01$).

Evidence from this study suggests that speech outcome standard 2 may be set slightly high. Although the standard was achieved for 2001 births (75%), this was influenced by the exceptional results of one center, associated with one surgeon who then retired. Consequently, the results were not replicated in 2002 to 2003.

Standard 3 - Articulation
Goal: 50%
range: 53-90%
average: 60%

TABLE 7 Results Nationally and by Center for Speech Outcomes Standard 3: "*By 5-5;11 years over 50% of children with CP±L will have no cleft-related articulation difficulties requiring SLT and or surgery*"†

Year	*National Outcome (% ±CI)*	*No. Centers Achieved*	*Range of Outcomes by Center (% ±CI), (median)*
2001 (n = 314)	61% ± 6%, **$P < .001$	6/6	53% ± 13%, 59% ± 26%, ***59% ± 14%, 68% ± 18%,*** 68% ± 13%, 76% ± 17%
2002 (n = 363)	57% ± 5%, *$P = .045$	7/8	34% ± 14%, 54% ± 17%, 59% ± 16%, ***62% ± 15%, 63% ± 12%,*** 64% ± 21%, 71% ± 19%, 77% ± 29%
2003 (n = 431)	62% ± 5%, **$P < .001$	10/11	44% ± 20%, 52% ± 14%, 53% ± 15%, 55% ± 13%, 57% ± 15%, ***64% ± 13%,*** 64% ± 19%, 77% ± 12%, 77% ± 18%, 77% ± 22%, 90% ± 35%
Total (N = 1110)	60% ± 3%, **$P < .001$		

† CI = confidence interval based on a 95% confidence level.
Rejecting the null hypothesis that the national speech outcome is the same as the standard at the 5% significance level (*$P < .05$) and at the 1% significance level (**$P < .01$).

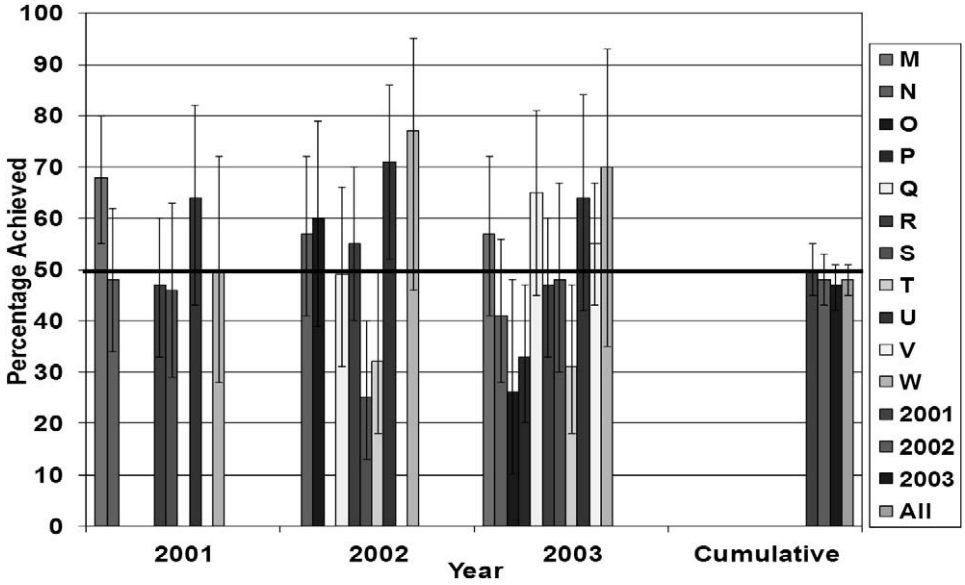

FIGURE 2 Results nationally and by center for Speech Outcome Standard 1: "*By 5-5;11 years over 50% of children with CP±L will have speech within the normal range*." Bars show results for each center (m-w) submitting more than 50% eligible cases by year (LHS) and annual national results (2001 to 2003) and total results (RHS). Whiskers show 95% confidence interval. Solid horizontal line shows the standard. If the confidence interval crosses the standard, the result is not significantly different from the standard.

significance level ($P < .05$) showed the national outcomes for 2001 (51% ± 5%, $P = .96$), 2002 (46% ± 5%, $P = .40$), 2003 (47% ± 4%, $P = .18$) and the cumulative outcome (48% ± 3%, $P = 0.20$) were not significantly different from the standard. However, Figure 2 demonstrates the variability in outcomes and wide confidence intervals for individual centers (15% to 20%) attributable to their small sample size (mean = 33 cases per year). The annual national data, based on a larger sample size (about 350 cases per year) had small confidence intervals under 5%. Despite the variation

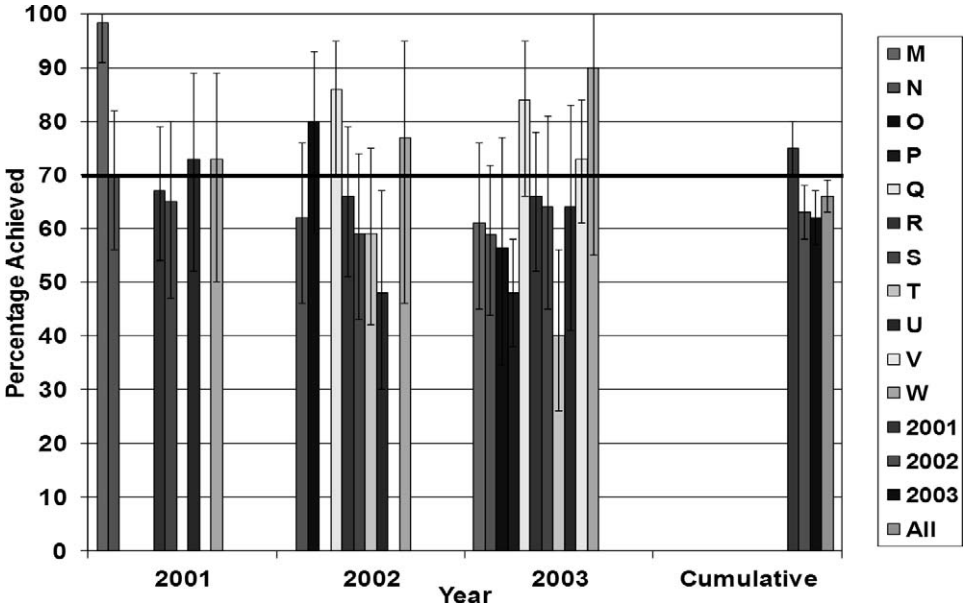

FIGURE 3 Results nationally and by center for Speech Outcome Standard 2a: "*By 5-5;11 years over 70% of children with CP±L will have speech with no evidence of a structurally related problem and have not had velopharyngeal surgery or fistula repair for speech*." Bars show results for each center (m-w) submitting more than 50% eligible cases by year (LHS) and annual national results (2001 to 2003) and total results (RHS). Whiskers show 95% confidence interval. Solid horizontal line shows the standard. If the confidence interval crosses the standard, the result is not significantly different from the standard.

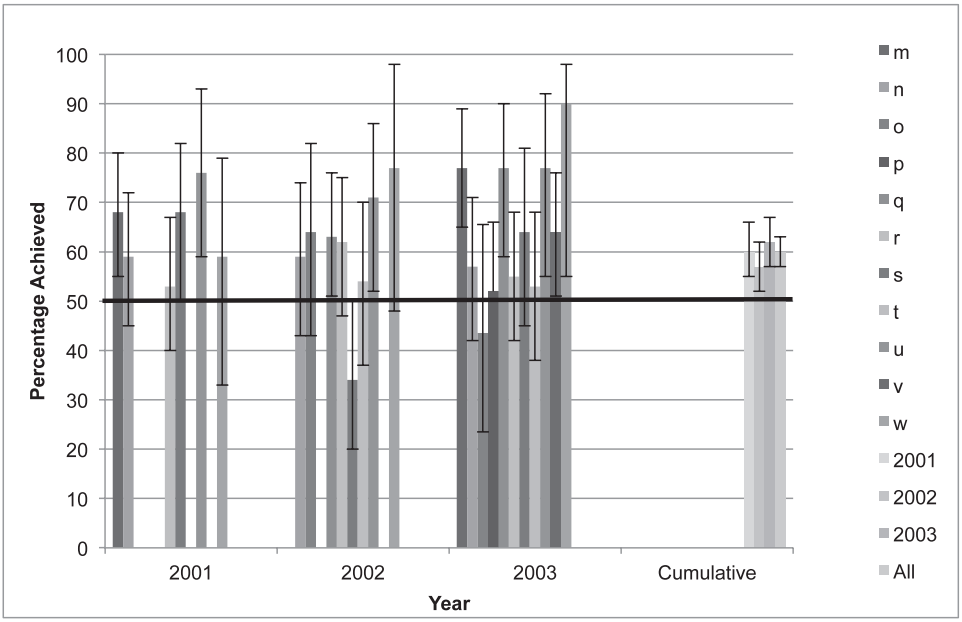

FIGURE 4 Results nationally and by center for Speech Outcomes Standard 3: "*By 5-5;11 years over 50% of children with CP±L will have no cleft-related articulation difficulties requiring SLT and/or surgery*." Bars show results for each center (m-w) submitting more than 50% eligible cases by year (LHS) and annual national results (2001 to 2003) and total results (RHS). Whiskers show 95% confidence interval. Solid horizontal line shows the standard. If the confidence interval crosses the standard, the result is not significantly different from the standard.

across centers, the national outcomes were very similar across all 3 years.

Outcome Standard 2a states that "*By 5-5;11 years over 70% of children with CP±L will have speech with no evidence of a structurally related problem and have not had velopharyngeal surgery or fistula repair for speech*." Table 6 and Figure 3 show that the confidence intervals for individual centers were again wide and outcomes were variable, but the national trend was more consistent and the confidence intervals smaller. For 2001 births, the standard was achieved with a national outcome of 75% ± 5%. This was influenced by the exceptional results of Centre M. This was not repeated for 2002 and 2003 births, for which the national outcomes were similar (63% ± 5%/62% ± 5%), giving a cumulative outcome for all 1110 cases of 66% ± 3%. Statistical analysis, assuming a 5% significance level, showed that although results for 2001 were significantly better than the standard ($P = .036$), results for 2002 ($P = .007$), 2003 ($P < .001$), and the cumulative outcome ($P = .007$) were significantly below the standard.

From 2002, information was also gathered on sections 2b-d, which divide the children with or a history of structurally related speech problems into three subgroups. Table 8 shows the national outcomes and range for each subgroup in 2002 and 2003. Again, over the 2 years, the results were remarkably similar. About 36% of the cohort each year had structurally related speech

TABLE 8 Additional Data for Children With Structurally Related Speech Problems

	2002	*2003*
2b. Percentage of children who have had velopharyngeal surgery or fistula repair and now have no evidence of a structurally related speech problem		
National outcome	10.6%	10.5%
Median by center	8%	9%
Range by center	0%–22%	0%–24%
2c. Percentage of children who have had velopharyngeal surgery or fistula repair and still have speech indicative of a structurally related problem		
National outcome	10.3%	10.6%
Median by center	9%	10.2%
Range by center	0%–19%	0%–22%
2d. Percentage of children who have not had velopharyngeal surgery or fistula repair but their speech is indicative of a structurally related problem		
National outcome	17%	16.7%
Median by center	15%	16%
Range by center	0%–9%	0%–29%
2b-d. Total percentage of children with a history of speech difficulties indicative of structural problems		
National outcome	37.9%	36%

problems. Of these, about 10% had secondary surgery that was successful in eliminating VPI (2b), while 10% had undergone secondary surgery but still showed speech patterns indicative of structural problems (2c). Meanwhile, about 17% had not undergone secondary surgery but had speech indicative of structural problems.

Outcome Standard 3 states that "*By 5-5;11 years over 50% of children with CP±L will have no cleft-related articulation difficulties requiring SLT and or surgery*." Table 7 and Figure 4 show that this was the most consistently achieved outcome standard over the 3 years. Eight centers achieved this standard over at least 2 years and four centers over all 3 years. Again, the national outcomes and cumulative data were very consistent with small confidence intervals. Statistical analysis assuming a 5% significance level showed that the national outcomes for 2001 (60% ± 6%, $P < .001$), 2002 (57% ± 5%, $P = .0045$), and 2003 (62% ± 5%, $P < .001$) and the cumulative outcome for all 1110 cases (60% ± 3%, $P < .001$) were all significantly higher than the standard.

Discussion

Following the Clinical Standards Advisory Group report and reorganization of cleft care in the United Kingdom (Department of Health, 1998a), regional cleft centers were mandated to conduct multidisciplinary audit. According to recommendations for audit (NICE, 2002; Health Care Quality Improvement Parternership, 2009), standards were required against which to compare the processes and outcomes of cleft care. Through this multicenter study, standards for speech have been defined and extensively tested on 1110 cases over 3 years.

These evidence-based standards for speech and the national outcome data now provide benchmarks against which UK cleft centers can compare their own processes and outcomes to complete the audit cycle and inform stakeholders. Speech recordings are now collected annually using the validated and reliable perceptual audit measure (CAPS-A; John et al., 2006) and systematically benchmarked against nationally agreed standards. The results are centrally collated, and an annual report is produced (Britton, 2009, 2011). More data are required before statistically meaningful conclusions can be drawn about the speech outcomes of any individual center.

Process Standards

NICE (2002) guidelines suggest the best way to improve care is to identify and measure process standards. Agreeing on and refining process standards provided an opportunity to generate statements about service delivery to support speech outcomes. Most children were offered specialist speech and language assessment by 2 years of age and a 5-year speech assessment in line with the agreed standards. Adherence to process standards improved over the 3 years. The process standards have begun to provide a valuable tool for benchmarking service delivery following which a shortage of CAPS-A–trained listeners and differences in timing of early assessments have been identified.

Process standards needed to be specific, objective, measurable, achievable, and realistic. Meaningful therapy standards proved difficult to define or use. Consequently, the standards defined focused on easily quantified but small aspects of the SLT role in cleft care relating to early assessment and audit while omitting other important areas such as the provision of therapy. Process standards need to be reevaluated over time, and further development of the process standards is recommended to reflect both the wider role of the SLT in cleft care and multidisciplinary processes influencing speech outcomes.

Speech Outcome Standards

Three speech outcome standards for 5-year-olds with CP±L were defined and comprehensively tested. Following initial feedback, they were all worded as positive statements. Standard 1 provides a benchmark for speech outcome for the entire multidisciplinary team, standard 2a is closely related to surgery, and standard 3 depends on surgery, SLT, and audiology.

Defining the outcome standards using published research proved difficult owing to the limitations of the current evidence base on speech outcomes in cleft palate. The standards were tested using consensus analysis of speech recordings for 1110 five-year-olds over 3 years. To the authors' knowledge, this is the largest cohort study of speech outcomes to date. The results demonstrated remarkably consistent annual national speech outcomes despite variability across centers. The results suggest that speech outcome standard 1 relating to normal speech is set at an appropriate level, speech outcome standard 2 relating to structural speech difficulties may be set slightly high, and speech outcome standard 3 regarding articulation was easily achievable and therefore could be raised. In other specialties in the United Kingdom such as cardiac surgery, standards are nationally determined based on the audit data collected rather than published research (Bridgewater et al., 2013). Given the national outcomes combined with the recent excellent review by Lohmander (2011), further refinement of the standards is anticipated.

Speech Outcome Standard 1 states that "*By 5-5;11 years over 50% of children with CP±L will have speech within the normal range*." From this cohort, 48% ± 3% had CAPS-A speech profiles within the normal range. This was not statistically different from the standard (*P*

= .2), providing evidence that this standard is set at an appropriate level. From the research evidence, Lohmander (2011, p. 68) concluded that 60% to 70% of children with cleft palate aged 4 to 5 years could be expected to have good speech. However, advising that, with treatment, children have a 50% chance of speech within the normal range by age 5 is currently more realistic in the United Kingdom.

Enabling children with cleft palate to have normal speech by age 5 could be argued to be the most important speech outcome measure for both the family and the cleft team. For the family, this goal is important both socially and educationally, while for the cleft team, it is a multidisciplinary outcome involving timely and effective primary surgery, well-coordinated multidisciplinary follow-up, proactive hearing management, effective speech and language therapy (if required), prompt and successful revision surgery (where necessary), and family commitment to care. For some children to achieve normal speech by 5 years, they may have already been treated for hearing difficulties, articulation problems, VPI, or fistulae.

Although most professionals working with patients with CP±L agree that a successful speech outcome should be defined as "normal" speech (Kummer et al., 2012), it is less clear how "normal speech" should be defined and by whom. Normal speech could be operationalized in many ways, but for the purposes of this study, it was defined using CAPS-A (John et al., 2006) assessed by specialist SLTs. It is recognized that this is a narrow perspective, and it would also be useful to consider the child's and parents' perspectives.

Speech Outcome Standard 2, regarding structure, proved the most difficult standard to define and achieve. This standard was redrafted following the initial pilot on 2001 births. Originally, it stated that "*By 5-5;11 years, the prevalence and history of VPI will be less than 29%*." However, the results from 2001 data indicated that although it was possible to identify children with speech indicative of structural problems, differential diagnosis of VPI is not possible from perceptual assessment alone. This standard was therefore reworded as "structurally related speech problems," which may be either VPI or fistula related and, like the other standards, redrafted as a positive statement. Supplementary sections 2b–d were also added. Speech Outcome Standard 2a now states that "*By 5-5;11 years over 70% of children with CP±L will have speech with no evidence of a structurally related problem and have not had velopharyngeal surgery or fistula repair for speech*." Sections 2b–d are not part of the standard but provide valuable additional information for a center looking at its outcomes. While surgeons strive to ensure primary surgery is as effective as possible, there will always be some children with evidence of structurally related speech problems for whom it is not appropriate to intervene before their 5-year assessment, either clinically driven or because of the family's wishes (subgroup 2d). Others will have undergone revision surgery for speech with (2b) or without (2c) successful outcomes for speech by age 5.

Evidence from this study suggests that speech outcome standard 2 may be set slightly high. Although the standard was achieved for 2001 births (75% ± 5%, $P = .036$), this was influenced by the exceptional results of one center, associated with one surgeon who then retired. Consequently, the results were not replicated in 2002 to 2003. These results have been independently verified in other studies but have not been referenced here in order to maintain the anonymity of centers. The national outcomes for 2002 births (63% ± 5%, $*P = .007$), 2003 births (62% ± 5%, $*P < .001$), and the cumulative results for all 1110 five-year-olds (66% ± 3%, $*P = .007$) were significantly below the standard. This outcome is lower than might be expected from published studies to date (Lohmander, 2011). Further data collection will help confirm whether this standard needs adjusting.

It is possible that the results have been affected by the known challenges associated with the perceptual rating of hypernasality, which is a key component of this standard (Lee et al., 2009; Sweeney, 2011). Whitehill (2010) concluded that there is a lack of reliability associated with rating hypernasality and stressed the importance of high-quality training. However, studies using the CAPS-A tool with its definitions and training program, as undertaken by all raters in this study, have reported satisfactory reliability (Sell et al., 2009). Judges were calibrated during the comprehensive training to use CAPS-A and interrater/intrarater reliability calculated before and after training. Ongoing calibration is achieved through the consensus listening process and ensuring an external listener from another center is involved in at least 20% of cases.

It is also possible that the results have been affected by overestimating the significance of minor nasal airflow errors. CAPS-A defined minor nasal emission or turbulence as "occasionally heard on pressure consonants, less than three examples on different sounds." Clinicians conducting the audit assessments reported concern that children whose only structurally related speech difficulty was clinically minor audible nasal emission or turbulence were rated as not achieving this standard. This has led to modifications of the nasal airflow sections on the CAPS-A audit tool (Sell et al., 2008). The modified version of CAPS-A has been used for speech audit from 2004 births onward. The impact of these modifications is currently unknown.

Speech Outcome Standard 3 states that "*By 5-5;11 years over 50% of children with CP±L will have no cleft-related articulation difficulties requiring SLT and or surgery*." In this study, 60% ± 3% of the total cohort

had no cleft-related articulation difficulties. This outcome was significantly higher than the standard ($P < .001$), suggesting the standard is easily achievable. Similarly, a median of 74.5% of children in the published studies of 5-year speech outcomes reviewed by Lohmander (2011) had no articulation errors. Given these findings, it is anticipated that this standard is likely to be raised.

It is important to highlight the relevance of differentiating obligatory speech errors from those that are the function of mislearning. Speech outcome standard 2 focuses on the obligatory or passive speech patterns that are likely to be the result of VPI or symptomatic fistulae while speech outcome standard 3 focuses largely on those active or compensatory speech patterns that are likely to be the result of mislearning and therefore require therapy. However, it is recognized that a clear distinction does not always exist and that compensatory articulations may persist despite no structural difficulties (Harding and Grunwell, 1998; Peterson-Falzone et al., 2005).

More data are required before robust statistical conclusions can be drawn for individual centers. However, interesting patterns have begun to emerge when comparing outcomes between the standards. For example, some centers with a high proportion of children with a history of structural speech problems (outcome 2a) still have a high proportion of children with speech within the normal range by 5 years (outcome 1). This may reflect the important role played by all members of the multidisciplinary cleft team in ensuring that even if a child's primary repair is not successful, the coordinated work of the SLT, audiologist, and surgeon can help ensure the child has normal speech by age 5.

Methodological Issues

Centers reported on a consecutive series of cases following consistent inclusion/exclusion criteria to minimize the number of confounding variables. Overall, 25% of cases were excluded from the total cohort. Although the range for individual centers was quite wide (15% to 43% exclusions), most excluded about 25% of cases. From those cases eligible for the audit, another 34% were not submitted for reasons specified on the data submission form such as equipment failure, damaged tapes, and uncooperative children.

Blinded, independent, perceptual analysis of speech data by specialist SLTs is argued to be the gold standard methodological approach when reporting audit and research outcomes (Sell, 2005). This project met most of the recommended criteria for listening procedures in perceptual studies of cleft palate speech (Lohmander and Olsson, 2004; Brunnegard and Lohmander, 2007; Henningsson et al., 2008). These included high-quality speech recordings and playback equipment, judgments from multiple raters, an external listener in 20% of cases per center, using a narrow age span in the study group, and reporting the exclusion of individuals with additional syndromes or cognitive delay. Intra- and inter-rater reliability was calculated when training listeners to use CAPS-A (Sell et al., 2009). Although speech recordings were not randomized or blindly assessed, consensus listening was used as a recognized method of reducing measurement error (Shriberg et al., 1984; Sell, 2005). The time-consuming task of consensus listening was justified by recognizing the importance of maintaining competencies of specialist SLTs in analyzing cleft speech (John et al., 2006). The outcomes of this audit may be influenced by the tendency for specialist SLT listeners to be overcritical of a child's speech (Sell, 2005; Havstam and Lohmander, 2011, p. 309).

Any system analyzing speech will be a socially constructed model reflecting an interpretivist epistemology (Heselwood, 2009; Benton and Craib, 2011). Speech outcomes could be defined by experts using a validated measurement tool or identified more functionally by parents, children, or untrained listeners. Sell (2005) commented that untrained listeners add real-life significance to clinical speech assessments. The judgments of lay listeners in identifying how distracting or acceptable they find a person's speech are now being recognized as increasingly important (Brunnegard et al., 2009; Whitehill, 2010). Although this study defined standards based on assessment by specialist SLTs, there is growing recognition of the importance of parent and child views on speech outcomes (Department of Health, 2008; Havstam et al., 2011). Kuehn and Moller (2000) advocated that speech outcome measures should include both professional and patient or caregiver judgments. Peers may also be an important "lay listener" to consider, since a child's self-esteem may be related to peer acceptance. Furthermore, the International Classification of Functioning Disability and Health for Children and Young People (World Health Organization, 2007) advocates describing the impact of impairment on an individual's functional performance (activity) and the social consequences (participation). Further development of the speech outcome standards should now give careful consideration to parent and child perception of the child's speech and collecting information on activity and participation as well as impairment (Havstam and Lohmander, 2011).

This study did not control for, or collect data on, primary surgery, and there was a range of uncontrolled variables including cleft type, surgical timing, surgical procedure, and hearing management. It is recognized that audit provides a lower level of evidence than clinical trials but has an important place in the evaluation of clinical practice and service delivery (Reilly et al., 2004; Sell, 2005; John et al., 2006).

The prevalence of different cleft palate types in this 3-year study reflected the prevalence reported by the

national register for England, Wales, and Northern Ireland (CRANE Database, 2012). Excluding cases with cleft lip only, of the children registered on the National CRANE database with cleft palate involvement from 2002 to 2011, 30% had UCLP, 13% had BCLP, and 58% had CPO (CRANE Database, 2012). Similarly, in this study, 32% (361) had UCLP, 13% (142) had BCLP, and 55% (607) had CPO. The United Kingdom appears to have a higher proportion of children with CPO than many countries. Following a comprehensive international review of cleft registries and a systematic literature review, Mossey and Modell (2012) found that overall, European and U.S. studies on nonsyndromic cleft prevalence suggest 30% to 35% of cases have UCLP, 20% to 25% have isolated CL and CP, and 10% have BCLP. However, they found wide variation in the reported prevalence of different cleft types between countries, which was most pronounced for CPO. While this may relate to a range of factors including clinical ascertainment and methodology for reporting, the authors argue that the differences identified between countries in Europe are probably real, with higher rates of CPO reported for Canada and parts of northern Europe. This includes the United Kingdom.

During this project, there was debate among the U.K. cleft teams about defining speech outcome standards by cleft type. The Clinical Standards Advisory Group recommendations mandated U.K. cleft teams to audit speech outcomes for all children with cleft palate involvement. Consequently, this project took a pragmatic approach and aimed to make the standards as inclusive as possible by including all cleft types. The literature shows a strong bias toward reporting outcomes for children with UCLP. Despite the fact that children with UCLP constitute only about 30% of those with cleft palate involvement in the United Kingdom (CRANE Database, 2012), 68% (464/680) of 5-year-olds reported in the review by Lohmander (2011) had UCLP. If mandatory audit involved only cases with UCLP, most of the cleft palate caseload would be excluded. Furthermore, with about 10 cases of UCLP per center annually, it would be difficult to collect enough cases to get statistically meaningful data. By benchmarking the standards using a representative cohort across cleft types, it is hoped they will more comprehensively reflect the speech outcomes for the whole nonsyndromic cleft caseload.

Few studies have compared speech outcomes across cleft types, and they report varying results. Lohmander (2011) identified 13 studies reporting speech outcomes separately for different cleft types. Of these, several report similar speech outcomes across cleft types (Pulkkinen et al., 2001; Timmons et al., 2001; Brunnegard and Lohmander, 2007), while others report variation (Persson et al., 2002; Nyberg et al., 2010). Hardin-Jones and Jones (2005) found a significant relationship between cleft type and speech outcomes. Further investigation is required to determine whether it is necessary to split speech outcomes by cleft type.

Teams also debated the inclusion criteria relating to children with syndromes. It might be argued that the only children who should be excluded were those unable to complete the speech audit task regardless of diagnosed syndrome. However, data used for audit reports needed to contain the minimum number of confounding variables while remaining as comprehensive as possible. Most of the evidence used to define the standards involved a nonsyndromic cleft population. However, two related recent studies found that children with isolated cleft palate and associated syndromes or malformations (such as Pierre Robin Sequence) have significantly higher rates of both VPI (64%) and articulation difficulties compared with children with nonsyndromic isolated cleft palate (VPI, 14%) aged 5 years (Persson et al., 2002) and a higher prevalence of persisting VPI and glottal articulation at the ages of 7 and 10 years (Persson et al., 2006). It was therefore agreed to exclude children with diagnosed syndromes from national outcome reporting.

Costs Versus Benefits

The process of undertaking high-quality speech audit according to NICE (2002) recommendations is extremely time-consuming and includes training listeners, collecting and analyzing the speech recordings, uploading the analyzed data onto local/national databases, collating and summarizing the data, and providing an annual report to clinicians and users. This investment must be weighed against the quality of speech outcome data obtained through this process. John et al. (2006, p. 280) noted that "the rating of 10 cases for consensus listening took an average of five hours, including breaks," and the quality of speech assessment is affected by fatigue (Stoel-Gammon 2001). The time required was borne out in this study. To rate 1110 cases over 3 years required 550 hours of listening. Assuming three listeners at each consensus listening session, this equates to 235 days of specialist SLT time. Initiatives under way to try to reduce the time required to complete the analysis while maintaining reliability include eliminating the rating of intelligibility of conversational speech due to concerns around its validity and the additional listening time taken for this parameter. Furthermore, alternative methods for measuring speech outcomes (such as patient self-ratings) at other mandatory audit ages are being explored. Nevertheless, it has to be recognized that perceptual speech assessment of cleft palate speech is time-consuming.

Future Developments

This is believed to be the first large-scale audit study of this type to be published. Having defined and extensively piloted standards for processes of care and speech outcomes, future work includes agreeing on a strategy for publication of center-specific outcomes, assessing the costs versus benefits of audit, and further developing the standards. Further work is needed to assess the variability in CAPS-A outcomes across cleft types and to collect more center-specific speech outcome data. A new 3-year phase using 2004 to 2006 births is now in progress using the revised CAPS-A tool. This will enable comparison of speech outcomes with the 2001 to 2003 cohort, investigation of the speech outcomes of individual centers over a 6-year period, and evaluation of the impact of the modifications to the CAPS-A tool.

It was the responsibility of individual centers to check that the raw data matched the summarized data submitted. Validation of the data will be improved in the future by submission of raw data to a national database, which is an important step before implementing the publication of center-specific speech outcomes.

The evidence from this study would suggest that nationally, speech outcomes across cleft types are reasonably consistent from year to year. However, there are indications that considerable variability exists between centers, and a key goal for the future will be to identify why this variation exists and how to improve outcomes. Contributing factors might include type and timing of primary surgery, intensity, timing and quality of SLT, and nature of hearing management. This broad-based multicenter audit of 1110 five-year-olds provides a strong benchmarking tool for future investigations.

The ultimate goal of audit is to share the data collected with patients and their families and other stakeholders to improve clinical outcomes. It would have been premature to name centers in this study because the development of the standards was at an early stage and it was important to refine the data collection process. Furthermore, collecting statistically reliable data for any one center (using a 95% confidence level) requires approximately 350 cases to achieve a confidence interval of approximately 10%. With centers currently submitting approximately 50 speech audit records per year across cleft types, it will take approximately 7 years to collect 350 cases. Speech outcomes should ideally be published alongside other outcomes in cleft care. A strategy for publishing speech outcomes alongside other outcomes for individual centers is now required and is under discussion in the United Kingdom following the lead of U.K. cardiac services (Bridgewater et al., 2013).

Normative data for CAPS-A would be useful. It is assumed that most children without cleft would have a "normal" profile on CAPS-A, but this has not been studied. Published studies have commented on the presence of unexpected speech difficulties in their control groups. Lohmander and Persson (2008) noted mild hypernasality, and Persson (2002) reported retracted articulation, hypernasality, nasal emission, and VPI demonstrated in the 5-year-old control groups.

Future development of the national standards should include the development of more process standards (e.g., for therapy) and ongoing refinement of the 5-year speech outcome standards. Adopting the audit model used in other specialties (Bridgewater et al., 2013), it is anticipated that the standards will not be set in stone based on the relatively limited evidence from research studies but constantly redefined based on the audit data collected. The need to develop a method for obtaining a parental/child judgment of speech outcome is also recognized. Finally, similar projects to develop and pilot speech outcome standards for the other mandatory audit ages are required.

While large-scale intercenter audit projects can help set benchmarks for measuring the outcomes of entire protocols, rigorous high-quality research is required alongside audit to enable us to continue to raise these standards in everyday practice.

Conclusions

As a result of this project, national standards for speech based on the literature and expert opinion have been defined and extensively tested. Results from 1110 children age 5 years with CP±L showed that 48% of cases had speech within the normal range, 66% had no evidence or history of structurally related speech difficulties, and 60% had no cleft-related articulation difficulties. The systems of data collection have been refined, and a continuous national program of intercenter audit of speech outcomes for all nonsyndromic 5-year-olds with cleft palate involvement is now in place. The standards provide evidence-based benchmarks and national data against which centers can compare their own speech outcomes and reflect on their clinical practice.

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- **Northern and Yorkshire** led by Stephanie Delvin, Jenny Nayak
- **Northern Ireland** led by Christine Hayden

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- **North Thames** led by Debbie Sell
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Appendix

Summary of Review of Speech Outcome Studies Involving 5-Year-Olds (Adapted From Lohmander, 2011)

Study	*Havstam et al. (2011)*	*Lohmander and Persson (2008)*	*Nyberg et al. (2010)*	*Pigott et al. (2002)*
Design	Retrospective case series (longitudinal correlation with attitudes age 10 years)	Prospective cohort (longitudinal)	Retrospective, cross-sectional (comparative: cleft type, cleft versus noncleft, technique)	Retrospective cross-sectional (comparative: three surgical techniques)
Level of evidence	IV	II	III	III
Subjects				
Number/possible	45/68 had 5 year records	N = 20/ni	86/133	66/139
Age at speech analysis	5, 7, 10 years	18/12, 3, 5, 7 years	4;0–6;0 (mean, 5 years)	5 years
Controls	No	10	18	
Cleft types	BCLP: 8, UCLP: 20, HCP:18, SCP:8	UCLP	HCP: 53, SCP: 33	UCLP
Exclusions	1× developmental delay	Additional anomalies	47 (30 missing audio)	Associated anomalies
Surgical procedure (no. of surgeons)	NI	Two stage (2)	One stage (4), two techniques	One stage (1), three techniques
Age at surgery	NI	SPC: 6 months HPC: 3–4 years	12–15 months	6 months
Secondary surgery rate	NI	4/20 after 5 years	17/86 (20%) after 5 years	6/66 9% by 5 years 2 for each technique
Fistula rate	NI	Unrepaired HP, all	6 (7%)	
Method of perceptual speech analysis				
Speech sample recorded	Audio	Audio	Audio	Audio
Speech sample	Standard sentences 1–2 minute conversation	Single words, sentence repetition	45 seconds–2 minutes, content varied	Single words, rote speech, conversation
Parameters analyzed	4: Ar, I, VPI, Imp.	5: Hyper, NA, WPC, G, ROA	7: Hyper, Hypo, WPC, NA, ROA, G, NPF	3: Ar, R, NE
Scales	Four-point scales	Five-point scales	Five-point scales	Five/seven-point scales
Reliability	Intrarater yes, interrater no	Interrater yes, Intrarater yes	Interrater yes, intrarater yes	Interrater yes (89%), intrarater no
Randomized	Yes	Yes	Yes	?
Blinded	Yes	Yes	Yes	?
External Raters	1	2	1	No
SLT Raters	2	2	4	*?2*
Multiple Raters	2	2	4	2
Structurally related speech outcomes aged 5 years				
Hypernasality	NI but 72% "normal (28%) + mild deviant (46%) VP function"	70% absent	HCP: 51% absent, SCP: 50% absent	39/66 59% absent
Nasal airflow	72% absent	90% absent	HCP: 61% absent, SCP: 53% absent	44/66 66% absent
Articulation-related speech outcomes (cleft speech characteristics) aged 5 years	59% absent	83% absent	HCP: 81% absent, SCP: 87% absent	30/66 45% absent
Intelligibility aged 5 years	70% good	82% good	NI	NI
Statistical analysis of data	Correlation/*t* tests	Significance testing	Significance testing	
Sample size calculation	None	None	None	None
Critique and bias	Selection bias	Small group	Selection bias, speech sample varied	Selection bias, randomization?
Notes and conclusions	35/45 (77%) "deviant" speech age 5 years		No SD difference with regard to technique	

Notes: (*L*) = include in Lohmander (2011); NI = no information; IVV = intravelar veloplasty; HPC = hard palate closure; SPC = soft palate closure; UCLP = unilateral cleft lip and palate; BCLP = bliateral cleft lip and palate; CPO = cleft palate only; HCP = hard cleft palate; SCP = soft cleft palate; Synd = syndrome. Scales: Ar = articulation; R = resonance; I = intelligibility; VPI = velopharyngeal incompetence; Imp = General impression as shown; Ac = acceptability; Hyper = hypernasality; WPC = weak pressure consonants; E = nasal emission; ROA = retracted oral articulation; G = glottals; NPF = nasopharyngeal friction; VF = velopharyngeal friction; CA = compensatory articulation. CAPS = Cleft Audit Protocol for Speech. Level of evidence as defined by Vallino-Napoli (2011).

Extended

Lohmander et al. (2009) in Lohmander (2011)	*Sell et al. (2001)*	*Lohmander-Agerskov et al. (1995) in Lohmander (2011)*	*Lohmander-Agerskov et al. (1998) in Lohmander (2011)*	*Lohmander et al. (2002) in Lohmander (2011)*
Retrospective longitudinal case series	Prospective cross-sectional case series	Retrospective case series, longitudinal	Retrospective case series, longitudinal	Retrospective case series, longitudinal
IV	IV	IV	IV	IV
55/65	238/326	15/18	21/35	22/22
5, 7, 10, 16, 19 years	5 or 12 years	5, 7 , 8.5, 10 years	13 months, 3, 5 years	5, 7 years
No	None			No
UCLP	UCLP	BCLP 5, UCLP 10	BCLP 5, UCLP 13, HCP 3	UCLP 22
	Syndromic, developmental delay			
Two stage (ni)	Various	Two stage (NI)	Two stage (NI)	Two stage (ni)
SPC 8 months, HPC 8 years	Various	SPC 6–8 months, HPC 8–9 years	SPC 8 months, HPC 8–9 years	SPC 2–12 months, HPC 8–9 years
0	16%	13%	0%	0%
NI	NI			
Audio	Audio	audio	audio	Audio
	Counting, sentence repetition, conversation			
	4: Hyper, Hypo, NA, Ar, I			
	Five-point scale (CAPS)			
Intrarater yes, interrater yes	Interrater yes, intrarater yes	Interrater yes, intrarater yes	Interrater yes, intrarater yes	Interrater yes, intrarater yes
Y	Yes	Y		Y
Yes	Yes	Yes		Yes
1	Yes	No	No	No
	Yes			
2	2	2	2	2
72% absent	70% absent	53% absent	78% absent	72% absent
68% absent	67% absent	67% absent	76% absent	40% absent
60	66% absent	53% absent	72% absent	75% absent
83	49% good	NI	NI	NI
	Frequencies and percentages			
	No			
Exclusion bias?	Selection bias	Small group	Small group, exclusion bias	Small group

Extended

Lohmander et al. (2006)	*Persson et al. (2002)*	*Persson et al. (2006)*	Chapman (1993)	Enemark et al. (1990)
Retrospective case series, longitudinal	Retrospective case series cross-sectional	Retrospective case series longitudinal	Cross-sectional	Longitudinal, prospective case series
IV	IV	IV	IV	IV
26/26	51/85	26/32	30	57 consecutive
5, 7, 10 years (5 year a/s before HP closed in some)	5 years	3, 5, 7, 10 years	3, 4, 5 years (20 each)	5 and 21 years
No	13 noncleft	17 noncleft	30 noncleft	No
UCLP 26	HCP: 26, SCP: 25 ± synd	HCP: 11, SCP: 15 ± synd	UCLP: 16, BCLP: 7, CPO: 7	UCLP
None	None	None	Other anomalies, sensorineural HL, developmental delay	Additional anomalies
Two stage (2), two procedures	Two stage (NI)	Two stage (NI)	NI	Two stage (1)
SP: 7 months, HPC: 38–89 months	SPC: 8 months, HPC: 2–3 years	SPC: 6 months, HPC: 4 years	Mean = 13.6 months	Lip and HP: 10 weeks, SP: 22 months
NI	2%	0 by 5 yrs, 8 (31%) by 10 yrs		13 (23%) by age 21
NI	NI	*2*		NI
Audio	Audio	Audio	Audio	Audio
Standardized sample: sentence repetition and spontaneous speech	Single word and sentence repetition	Vowel and sentence repetition	Single words (Goldman Fristoe Test of Articulation) spontaneous speech	Conversational speech, Danish Pressure Artic Test
6; Hyper, WPC, NE, ROA, G, I	7: VPI, hyper, WPC, VF, NE, G, ROA	6: VPI, Hyper, hypo, WPC, NA, CA (5)	Phonological analysis	3: R, A, VPI
Three/five-point	Five-point scale	Five-point scales	None	Three/seven-point scales
Interrater yes, intrarater yes	Interrater yes, intrarater yes	Interrater yes, intrarater yes	Interrater yes, intrarater yes	NI
Y	Yes	Yes	?	NI
Yes	Yes	Yes	?	
1	No	Yes	?	NI
1/4	*3*	*3*	1	NI
2	3	3	No	NI
72% absent	68% absent/mild, HCP: 69%/SCP: 94% absent	72% absent	NI	13/56 23% normal
36% absent	86% absent/mild, HCP: 67%/SCP:100% absent	62% absent	NI	
75% absent. (25% retracted oral, 10% glottal)	64% absent VFS, 92% absent glottal, 86% absent ROA	71% absent	No significant differences in phonological processes in cleft/noncleft children aged 5 years	3/56 5% normal
55% good	NI	NI	NI	
	Significance tests	Significance tests	Significance tests	Frequencies
No	No	No	No	No
Small group	Selection bias, no external rater	Small group, selection bias	Small sample	
Some 5 year a/s B4 HP closed	Cleft chn significantly higher VPI, hyper, WPC than noncleft, 35% mod-severe VPI, 14% nonsynd. Grp, 64% synd grp			15/56 26% VPI on radiographs at 5