

# A Prospective, Longitudinal Study of Feeding Skills in a Cohort of Babies With Cleft Conditions

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**Objective:** To examine the natural history of feeding skills in babies with clefts and identify risk factors and predictors of poor feeding.

**Participants:** Sixty-two babies with clefts were examined at 2 weeks, 3 months, and 14 months of age.

**Main Outcome Measures:** Feeding ability, oral motor function, and feeding efficiency were assessed. Univariate analyses were used to determine whether oral motor function and sequelae varied according to feeding ability or cleft condition. Multivariable logistic regressions were used to determine risk factors for poor feeding.

**Results:** Poor feeding skills were detected in one third of newborns. The prevalence of poor feeding reduced to 19% at 3 months of age and 15% at 14 months of age. Oral motor dysfunction and sequelae (particularly nasal regurgitation) were more commonly observed in babies with poor feeding skills irrespective of comorbidity. The main risk factor for poor feeding was a diagnosis of syndrome or Pierre Robin sequence (PRS). At 2 weeks of age, babies with syndrome or PRS were 15 times more likely to have poor feeding skills than their nonsyndromic counterparts. When syndrome or PRS was controlled for, babies with cleft palate and cleft lip and palate were equally likely to have poor feeding skills. Parental report of feeding efficiency was predictive of poor feeding in young babies.

**Conclusions:** Poor feeding skills are relatively common in newborns with cleft palate and cleft lip and palate. Treatment for feeding problems may be needed beyond the first year of life, especially for babies born with PRS or a syndrome.

KEY WORDS: *cleft palate, feeding, infancy*

More than 30 years ago a review of clinical research in cleft lip or palate (CL/P) concluded that there was lack of knowledge about feeding problems (Spriestersbach et al., 1973). In the intervening years, little has changed. Descriptive epidemiological studies necessary to document the prevalence, characteristics, and natural history of feeding problems have yet to be undertaken.

Preliminary information about the nature of feeding prob-

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lems in CL/P can be extracted from studies that have utilized a variety of designs, data collection methods, and measurement systems. Two retrospective investigations have examined the prevalence of feeding problems and demonstrated that babies with cleft palate (CP) and combined cleft lip and palate (CLP) have significantly more difficulty feeding than those with cleft lip (CL) (Drillien et al., 1966; Jones, 1988). Jones (1988) conducted a retrospective chart investigation in the United Kingdom and reported that 25% of babies with nonsyndromic CL/P ( $n = 202$ ) had significant feeding problems. Among these, babies with CP were reported to have slightly more difficulty feeding than those with CLP. Moreover, babies with CL fed well, which is consistent with parental perceptions of feeding ability reported by others (e.g., Oliver and Jones, 1997) and expert opinion (Shah and Wong, 1980; Wolf and Glass, 1992; Glass and Wolf, 1999; Bannister, 2001; Miller and Kummer, 2001).

Reports describing feeding skills in CL/P are often contradictory and lacking in detail. For example, two studies have reported abnormal oral motor function (OMF) (specifically tongue position) in participants with CP and CLP (Malek and

**TABLE 1** Cross Tabulation of Participant Characteristics and Demographic Data by Cleft Type at Entry Into the Study\*

	Total n (100%)	Cleft Lip n (%)	Cleft Palate n (%)	Cleft Lip and Palate n (%)
Total recruited	62	11 (17.74)	36 (58.06)	15 (24.19)
Type of cleft condition				
Hard and soft palate	18	NA	18 (100)	NA
Soft palate only	18	NA	18 (100)	NA
Unilateral	20	11 (55.00)	NA	9 (45.00)
Bilateral	4	0	NA	4 (100)
Gender				
Male	33	7 (21.21)	17 (51.52)	9 (27.27)
Female	29	4 (13.79)	19 (65.52)	6 (20.69)
Comorbidity				
Isolated cleft	39	10 (25.64)	18 (46.15)	11 (28.21)
Syndrome	4	0	2 (50.00)	2 (50.00)
PRS	7	NA	7 (100)	NA
PRS + other anomalies	5	NA	5 (100)	NA
Plus other birth defects	7	1 (14.29)	4 (57.14)	2 (28.57)

\*NA = not applicable.

Psaume, 1983; Campo-Paysaa, 1987), but this is contradicted by data from Brogan et al. (1987), who examined nine babies by using videofluoroscopy and reported no discernible differences between cleft and noncleft babies regarding tongue position and movement during bottle feeding. Furthermore, there is consensus in the literature that feeding sequelae (such as excessive air intake, nasal regurgitation, fatigue, coughing, choking and gagging on fluids, prolonged feeds, and discomfort) are commonly seen in babies with CL/P (Styer and Freeh, 1981; Jones et al., 1982; Clarren et al., 1987; Carlisle, 1998), but there is little indication as to whether or not these are more frequently associated with poor feeding or whether they occur sporadically across the population.

Descriptions of many different feeding techniques and devices for infants with cleft conditions are found in the literature. Reid (2004) reviewed the literature and appraised the evidence for a range of feeding interventions and found that very few were evidence based. Only 5 of the 55 papers reviewed were data driven. Further examination revealed that many of the interventions were based on a common assumption, namely, that babies with CP and CLP have feeding problems because of impaired suction. Although several researchers have demonstrated that CP does interfere with both suction and compression (Choi et al., 1991; Mizuno et al., 2002), impairment of other skills may also be critical to feeding success. This is certainly the case in other pediatric populations. For example, some preterm babies have abnormal OMF (Lau and Schanler, 1996), altered sucking performance (Medoff-Cooper et al., 1993; Gewolb et al., 2001), and oropharyngeal problems (Newman et al., 2001), which contribute to poor feeding. In CL/P it is also feasible that other feeding skills may be similarly affected; therefore, investigation beyond impaired suction and compression is warranted. The investigation of feeding difficulties in babies with CL/P is also important because of the potential impact on the baby (Felix-Schollaart et al., 1992; Neiman and Savage, 1997), family (Field and Vega-Lahr,

1984; Endriga and Kapp-Simon, 1999; Young et al., 2001), and health care resources (CSAG, 1998).

In summary, there has been no prospective population study of the prevalence, natural history, and characteristics of feeding in babies with cleft conditions. Therefore, the aim of this investigation was to prospectively examine the natural history of feeding skills in a cohort of babies with CL/P and to determine the characteristics, risk factors, and predictors of poor feeding during the first 14 months of life.

## MATERIALS AND METHODS

### Design

A prospective, longitudinal study of babies with CL/P was undertaken. Baseline measures of feeding skills were recorded at 2 weeks of age and repeated at 3 months and 14 months of age. Ethics approval was granted from the Royal Children's Hospital Ethics in Human Research Committee (20017A) and the Human Ethics Committee at La Trobe University. Informed consent was obtained from each of the families before commencement of the study.

### Participants

This investigation aimed to recruit all babies born in Victoria between January 2001 and July 2002. The prevalence of CL/P varies between 1.3 and 1.7 cases per 1000 live births in Australia; in Victoria, where the study was conducted, the incidence is approximately between 105 and 110 live births per year (Riley and Halliday, 2000). Sixty-two babies with a variety of cleft conditions were recruited (Table 1). Of the 62 recruited babies, 11 had CL, 36 had CP, and 15 had CLP. Families were mainly referred to the study from the cleft palate team at the Royal Children's Hospital, Melbourne, but some were referred via CleftPaLS (the local parent support group).

**TABLE 2 Summary of Feeding Ability Protocols\***

	Feeding Ability		
	Good	Satisfactory	Poor
Neonates†	Breast or bottle feeding established in 48 hours Feeding time for quota is $\leq 20$ minutes Consistent weight gain of $\geq 200$ g/wk beyond 1 week of life	2 to 7 days to establish feeding (breast or bottle) Feeding time for quota is 20 to 40 minutes Weight gain $< 200$ g/wk beyond the first week of life	More than 7 days to establish feeding Feeding time for quota is 40 to 60+ minutes Irregular weight gain or loss beyond the first week of life
Infants (3 months)	$< 20$ minutes to take bottle and WHZ $< 1$ SD below the mean	$< 40$ minutes to take bottle or WHZ $< 2$ SDs below the mean or both	$> 50\%$ of feed delivered via nasogastric tube, or $> 40$ minutes to take bottle or WHZ $> 2$ SDs below the mean or both
Toddlers (14 months)	Cup feeding established (may still be breast or bottle feeding) Managing solids such as adult textures/finger foods consistently	Breast/bottle feeding predominantly, starting to cup feed Managing solids to at least a soft solids consistency	Enteral feeding continuing with solids or, if fully oral, primarily having pureed solids

\* WHZ = weight-for-height Z-score.

† Modified from original work by Jones (1988).

The participants ranged from 9 to 25 days of age at entry into the study (mean = 15.9, standard deviation [SD] = 3.45). The majority of participants were first-born singletons and the product of a full-term pregnancy. Birth weights reflected gestational age, with most participants clustering around the local average for this growth index. Approximately 18% of the cohort had a positive prenatal diagnosis of CL/P and 30.7% reported a positive familial history of CL/P in either first- or second-degree relatives.

Over half of the sample had a cleft condition in isolation (63%), that is, in the absence of a broader syndrome or co-existing congenital anomaly. Seven babies were classified as having nonsyndromic Pierre Robin sequence (PRS), and another five were classified as having PRS plus an additional congenital defect or syndrome (PRS+) such as Sticklers syndrome or spondyloepiphyseal dysplasia. Another four syndromic babies were identified. Two babies in the CP group had oculoauriculaovertebral dysplasia (Goldenhar syndrome) and Van der Woude syndrome, and two babies in the CLP group had lobar holoprosencephaly and multiple hormone deficiency.

A validation process using data from the Victorian Perinatal Data Collection Unit's Birth Defects Register (Riley and Halliday, 2000) was conducted to establish how representative the sample was of the Victorian CL/P population. The results of the validation process indicated that there were no significant differences between the proportion of babies with CL (chi-square(1) = 0.7670,  $p = .38$ ) and the proportion of babies with CLP (chi-square(1) = 1.9875,  $p = .16$ ) reported for the Victorian population and study participants. However, there was an overrepresentation of CP participants (chi-square(1) = 3.9216,  $p = .048$ ) within the study cohort and an overabundance of babies with PRS+ (Fisher exact,  $p = .01$ ) compared with prevalence rates in Victoria for previous years.

At the successive data collection appointments scheduled when participants reached 3 months and then 14 months of age, a proportion of the participants was lost to follow-up. The rate of attrition was 12.9% between the first and second appointments and then a further 27.8% between the second and third appointments. The overall attrition rate was 37%.

## Procedure

Families were visited at home for the first two appointments and attended the Royal Children's Hospital (Melbourne) for the third appointment. All appointments commenced with a structured interview designed to capture demographic, medical, and feeding information, which was recorded on the Cleft Palate Feeding Survey. Direct assessments of specific feeding behaviors were then undertaken. The outcomes in terms of OMF, feeding sequelae, and feeding efficiency are reported in this paper. Sucking performance (e.g., suction, compression, suck width), breast-feeding patterns, and growth and development are the subjects of forthcoming papers.

At each appointment, a protocol to classify feeding ability was applied to participants to determine the prevalence of good, satisfactory, and poor feeders within the cohort. The protocol for neonates (2 weeks of age) was modeled on parameters described by Jones (1988), including time taken to establish feeding (days), time to feed (minutes), and weight gain (grams). Three versions of the protocol were necessary to accommodate the advancing age and feeding skills of the participants (Table 2).

At the first appointment, a scheduled bottle or breast feed was videotaped, and OMF was measured by using the Neonatal Oral Motor Assessment Scale (Palmer et al., 1993). This instrument categorizes normal, disorganized, and dysfunctional sucking behavior.

Oral motor function was not assessed at the second data collection appointment because there were no standardized tests available for 3-month-old infants. At the third appointment, OMF was measured by using the screening version of the Schedule for Oral Motor Assessment (Reilly et al., 2000).

Results of intra- and interrater reliability for each of the tools confirmed agreement between two raters ranging from 88% to 100%, which was considered good to excellent.

Mothers of participants in the current study were asked whether coughing, choking, gagging, nasal regurgitation, or a wet or gurgly voice quality (i.e., feeding sequelae) occurred

during or immediately after feeding. Parental responses were recorded on the Cleft Palate Feeding Survey.

Feeding efficiency was calculated from parental report of the time taken to feed the baby (minutes) and quota consumed (milliliters) and expressed as milliliters per minute (mL/min) (Lau et al., 1997; Mizuno and Ueda, 2001; Mizuno et al., 2002). Feeding efficiency was calculated as mL/min under conditions where participants were fed with their own bottle and preferred milk. Participants who were nil by mouth or fully breast fed were excluded from this assessment.

### Statistical Methods

Descriptive statistics were used to determine the prevalence of babies with good, satisfactory, and poor feeding skills at each of the data collection appointments. The composition of each of the feeding skills groups was examined for cleft type (CL, CP, CLP) as well as syndrome or PRS. The associations among feeding ability, OMF, and feeding sequelae were examined by chi-square or Fisher exact tests (Schwartz and Polgar, 2003). Multivariate logistic regression analyses were undertaken to confirm the predictors of poor feeding.

The measure of feeding efficiency (mL/min) gathered via parental report showed promise in discriminating babies with poor feeding skills from those with adequate (good or satisfactory) feeding skills. Therefore, a receiver operating characteristic (ROC) curve analysis was undertaken to determine the discriminatory accuracy of this measure (mL/min) as a screening tool. A ROC curve expresses the relationship between the sensitivity and specificity for a given test (Fletcher et al., 1988).

### RESULTS

There was a fairly even distribution of feeding skills across the neonatal sample ( $n = 62$ ). Thirty-one percent ( $n = 19$ ) of participants had good feeding skills, 37% ( $n = 23$ ) had satisfactory feeding skills, and 32% ( $n = 20$ ) had poor feeding skills. Only 9 of the 20 poor feeders were feeding orally. These babies were slow to establish bottle feeding (taking between 7 and 14 days to manage full oral feeds) and slow to consume quota (median = 55 minutes; interquartile range [IQR] = 53, 60; range = 41 to 90 minutes). Furthermore, they had a history of poor weight gain beyond the first week of age. Of the remaining 11 poor feeders, 8 continued on full enteral feeds via nasogastric tube, and 3 were on partial oral feeds (with more than 50% of the daily quota obtained via enteral means). The association between cleft condition and neonatal feeding skill was significant (Fisher exact,  $p = .001$ ). Babies with CL were typically good feeders ( $n = 9/11$ ) and never poor feeders. Fourteen percent of CP participants were good feeders, 42% were satisfactory feeders, and 44% were poor feeders. Participants with CLP were relatively evenly distributed across the three feeding skills groups (33% recorded good feeding ability, 40% recorded satisfactory feeding ability, and 27% recorded poor feeding ability).

Poor feeders were heterogenous regarding syndrome diagnosis. Thirty-five percent of poor feeders had nonsyndromic CL/P, and the remaining 65% had various syndromes and PRS.

By 3 months of age, 44 of the 54 remaining infants were adequate feeders—31.5% were good feeders and 50% were satisfactory feeders. Ten participants (18.5%) were poor feeders. Five of these were nil by mouth and fed via nasogastric tube. Four were partially orally fed, receiving at least 50% of nutritional and hydration requirements via nasogastric tube and the remainder via oral intake. One poor feeder was fully orally fed. However, this participant took more than 60 minutes to feed and had a weight-for-age Z-score more than two SDs below the reference mean. Nine of the 10 poor feeders had either PRS or PRS+. The 10th participant (the only oral feeder) was classified as nonsyndromic unilateral CLP.

At 14 months of age, only 6 (15%) of the remaining 39 participants were classified as poor feeders. Five of these had PRS(+) and one had lobar holoprosencephaly. Two PRS(+) participants were fed via gastrostomy tube with some food offered orally. The remaining four poor feeders were fed orally but had not advanced to age-appropriate consistencies such as cut-up foods or finger food. They were fed puree solids and were drinking from a bottle or spout cup.

Most participants lost to follow-up had adequate (good or satisfactory) feeding skills at the time of attrition. Moreover, neonates with adequate feeding skills who continued in the study rarely (only 1 of 39) worsened, and poor feeders showed a trend of improvement over time. These observations were used first to impute data for the 23 participants who were lost to follow-up (see Table 3) and second for comparison with the actual data gathered at each of the appointments.

Table 3 illustrates the natural history of feeding skills across the study period. Imputed feeding ability classifications are written in text in the cells where participants were lost to follow-up. Comparison of imputed data with actual data revealed striking similarities: 32% of newborns ( $n = 20/62$ ) in the cohort had poor feeding skills, and this reduced to 19% (actual  $n = 10/54$ , imputed  $n = 12/62$ ) at 3 months of age. By 14 months of age, analysis of actual data suggested that only 15% ( $n = 6/39$ ) of the remaining cohort had poor feeding ability, which is almost the same rate yielded from the imputed dataset (16%,  $n = 10/62$ ).

Abnormal OMF was identified in 31% ( $n = 15/62$ ) of neonates and later in 15% ( $n = 6/39$ ) of toddlers aged 14 months. Eighteen of the 19 neonates with good feeding skills demonstrated normal OMF during nutritive sucking. One good feeder had disorganized OMF, which was characterized by arrhythmic jaw and tongue movements and an inability to sustain a suckle pattern beyond 2 minutes. A similar result was obtained for the satisfactory feeders, with 22 of the 23 having normal OMF. One satisfactory feeder had disorganized OMF, which was again characterized by arrhythmic jaw and tongue movements. Oral motor function was variable among the poor feeders. Seven of 20 poor feeders had normal OMF, and a further 7 had disorganized OMF. Those with normal OMF were nonsyndromic, and those with disorganized OMF had PRS(+). Un-

**TABLE 3 The Progression of Feeding Skills Across the First 14 Mo of life—Including Imputed Data**

#	Cleft	Syndrome	Neonates (2 weeks)	Infants (3 months)	Toddlers (14 months)
1	CL	-			
2	CL	-			
3	CL	-			
4	CL	-			
5	CL	-			
6	CL	-			
7	CLP	-			
8	CLP	-			adequate
9	CP	-			adequate
10	CP	-			
11	CLP	-			
12	CLP	-			
13	CL	-			adequate
14	CP	-			adequate
15	CLP	-			adequate
16	CP	-			adequate
17	CL	-		adequate	adequate
18	CP	-		adequate	adequate
19	CL	-		adequate	adequate
20	CL	-			
21	CP	-			
22	CP	Van der Woude			
23	CLP	-			
24	CLP	-			
25	CL	-			
26	CP	-			
27	CP	-			
28	CP	-			
29	CP	-			
30	CLP	-			
31	CLP	-			
32	CP	PRS+			adequate
33	CP	-			adequate
34	CP	-			adequate
35	CP	-			
36	CP	-			
37	CP	-		adequate	adequate
38	CP	-		adequate	adequate
39	CLP	-		adequate	adequate
40	CP	-			
41	CP	PRS			
42	CLP	-			
43	CP	-			
44	CP	-			
45	CP	-			
46	CLP	-			
47	CP	Holoprosencechaly			
48	CP	-			adequate
49	CP	PRS			adequate
50	CLP	-			adequate
51	CLP	MHD			adequate
52	CP	PRS+			poor
53	CP	PRS			poor
54	CP	PRS			
55	CP	PRS+			
56	CP	PRS+			
57	CP	PRS+			
58	CP	PRS			
59	CP	PRS			
60	CP	PRS			
61	CP	-		poor	poor
62	CLP	GoldenHar		poor	poor

Legend:

Good
Satisfactory
Poor
Imputed value

Poor = 20/62; 32%	Poor = 12/62; 19%	Poor = 10/62; 16%
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**Note.** Adequate feeding ability was a broad classification indicating either good or satisfactory feeding ability

**TABLE 4** Sensitivity and Specificity of Cut-Points for Feeding Efficiency of Neonates and 3-Month-Old Infants\*

	<i>Cut-Point, mL/min</i>	<i>Sensitivity, %</i>	<i>Specificity, %</i>	<i>Correctly Classified, %</i>	<i>LR+</i>	<i>LR-</i>
Neonates	<2.2	100.00	86.84	90.00	7.60	0
Infants	<3.3	100.00	97.14	97.50	35.00	0

\* LR+ reports the likelihood ratio for a positive test result, and LR- reports the likelihood ratio of a negative test result. Detailed data describing the results for other cut-points are available from the authors on request.

like the good and satisfactory feeders, some poor feeders ( $n = 6/20$ ) were classified as having dysfunctional OMF. These participants had lobar holoprosencephaly ( $n = 1$ ), Goldenhar syndrome ( $n = 1$ ), multiple hormone deficiency ( $n = 1$ ), and PRS(+) ( $n = 3$ ). Examples of oral motor dysfunction (OMD) included minimal jaw excursions and absence of both jaw and tongue movements for significant periods of time. The association between OMF and feeding ability reached statistical significance with good and satisfactory neonatal feeders more likely to have normal OMF compared with poor feeders (Fisher exact,  $p < .001$ ).

At 14 months of age, normal OMF was recorded for 85% ( $n = 33/39$ ) of all the participants, and OMD was recorded for the remaining 15% ( $n = 6/39$ ). Ninety-six percent of good feeders and 88% of satisfactory feeders had normal OMF, whereas only 20% of poor feeders achieved the same result. A positive association between OMF and toddler feeding skill was confirmed (Fisher exact,  $p = .001$ ).

Mothers of poor neonatal feeders reported a higher proportion of nasal regurgitation, wet or gurgly phonation, coughing, choking, gagging, and distress during feeding compared with mothers of good and satisfactory feeders. A statistically significant result was detected by Fisher exact tests between feeding ability and nasal regurgitation ( $p = .001$ ), wet or gurgly phonation ( $p = .016$ ), coughing ( $p = .014$ ), choking ( $p = .001$ ), gagging ( $p = .037$ ), and distress ( $p < .001$ ).

At 3 months of age, sequelae were reported for more than half of the participants ( $n = 40/62$ , 65%), whereas 23% ( $n = 14/62$ ) were confirmed as asymptomatic. Missing data were registered for 13% ( $n = 8/62$ ) of the sample. Statistically significant associations were confirmed by Fisher exact statistical tests for feeding ability and nasal regurgitation ( $p = .031$ ), coughing ( $p = .009$ ), choking ( $p = .011$ ), gagging ( $p = .015$ ), and distress ( $p = .002$ ).

By 14 months of age, only 13% ( $n = 8/62$ ) of the cohort were reported to have persistent sequelae, usually nasal regurgitation, associated with feeding episodes. Half of the toddlers were now reported to be asymptomatic ( $n = 31/62$ ). The outcome for the remaining 37% ( $n = 23/62$ ) of the cohort was unknown. The association between nasal regurgitation and feeding ability was just significant ( $p = .046$ ). Nasal regurgitation was reported for 5 of 39 toddlers (1 good feeder, 2 satisfactory feeders, and 2 poor feeders).

After controlling for the independent variables of cleft condition, birth weight, Apgar score, and gestational age, the presence of syndrome or PRS(+) was statistically significant in identifying poor neonatal feeders ( $p = .001$ ). The odds of be-

ing a poor feeder were almost 15 times greater when a diagnosis of syndrome or PRS(+) was made compared with nonsyndromic, non-PRS(+) counterparts. At 3 months of age, the presence of a syndrome or PRS(+) continued to be a statistically significant predictor of poor feeding ( $p = .005$ ), with the odds of being a poor feeder 53 times greater for infants with a positive diagnosis of syndrome or PRS(+) compared with counterparts with nonsyndromic CL/P. By 14 months of age, only syndromic or PRS(+) participants were classified as poor feeders.

A diagnosis of syndrome or PRS(+) cannot always be made immediately after a baby is born. Therefore, a logistic regression analysis was performed to determine whether or not poor feeding skills could be predicted from the independent variable cleft type (CP, CLP) when other variables such as syndrome were not controlled. The results confirmed that there was no significant effect for cleft type ( $p = .24$ ); thus, neonates with CP or CLP were equally likely to have poor feeding skills.

To determine whether the feeding efficiency (mL/min) measure was a useful clinical index for identifying poor feeders, its precision against a reference test (the neonatal feeding ability protocol) was examined by a ROC curve analysis. The feeding efficiency (mL/min) measure identified all neonates with poor feeding ability (sensitivity = 100%) when a cut-point of <2.2 mL/min was chosen for selection of cases. The corresponding specificity (86.84%) and positive predictive value (88%) were high (Table 4). Furthermore, the area under the ROC curve was 0.99 (95% confidence interval = 0.96, 1.0), confirming excellent overall performance of the measure.

Results of a second ROC analysis suggested that the feeding efficiency measure retained high discriminatory accuracy when used with 3-month-old infants. A cut-point of <3.3 mL/min resulted in 100% sensitivity, 97.14% specificity, and 97% positive predictive value in identifying infants with poor feeding skills (Table 4).

## DISCUSSION

Our data revealed that poor feeding diminished but did not entirely resolve during the 14 months of investigation. From an initial prevalence of 32% in neonates, the frequency reduced to 19% in infants (3 months of age) and 15% in toddlers (14 months of age). The neonatal rate was comparable with results reported by Drillien et al. (1966), who classified 32% ( $n = 47/149$ ) of babies with CL, CP, CLP, and PRS as having severe feeding difficulty according to length of time taken to feed. Not surprisingly, the rate of poor feeding was much high-

er than the 1% reported for otherwise healthy neonates (Motion et al., 2001). The trend of persistent feeding problems reported in the current study supports previous parental reports of poor intake and prolonged feeding times continuing well beyond the first 3 months of life (Zickefoose, 1957; Drillien et al., 1966; Trenouth and Campbell, 1996).

By 3 months of age, all but one of the nonsyndromic participants was an adequate feeder. This should be reassuring to the families of babies with nonsyndromic CL/P and to professionals alike. However, the path for syndromic and PRS(+) participants was different. Some participants demonstrated adequate feeding by 3 months of age, but others had persistently poor feeding to 14 months of age (the end point for review in the current study). All toddlers described as poor feeders had a confirmed syndrome or PRS(+) diagnosis. These data provide important information for service planners. First, the prevalence of poor feeding in a cohort of CL/P newborns may be significant even when syndromic and PRS(+) participants are excluded. Second, nonsyndromic CP and CLP neonates are equally likely to need feeding assistance in the first months of life, but by 3 months of age poor feeding will likely resolve. Third, participants with syndromes or PRS(+) are more complex and may require feeding support services for much longer, even beyond 14 months of age.

Oral motor dysfunction, which may have contributed to poor feeding, was mainly confined to participants with PRS(+) and recognized syndromes. Oral motor dysfunction is thought to arise primarily from neurological immaturity or impairment (Miller-Schuberth, 1994) but may also arise from structural abnormalities (Glass and Wolf, 1999). It is likely that both of these hypotheses were applicable to the current CL/P cohort, which included syndromic and nonsyndromic participants. However, further investigation is required to understand the basis of OMD in babies with cleft conditions.

Sequelae were commonly associated with feeding episodes in neonates with CL/P, particularly those with poor feeding skills. At least some of the behaviors persisted in half of the cohort to 3 months of age and in a small percentage of participants to 14 months of age. Nasal regurgitation was the most commonly reported behavior at each of the three appointments. Elimination of nasal regurgitation is desirable because the regurgitation may lengthen the duration of a bottle or breast feed (Sidoti and Shprintzen, 1995), is unpleasant for the baby, and is potentially stressful for the mother (Zickefoose, 1957).

The best mechanism for detecting feeding problems in babies with CL/P remains uncertain and is probably dependent on local issues such as geography, human resources, and the size of the CL/P population. We believe that a diagnostic feeding assessment of all new babies with CL/P is unrealistic and indeed unnecessary. However, universal surveillance with a screening tool could be an efficient way to identify "at risk" babies, who should then be referred onward for verification of diagnosis by a suitably qualified feeding specialist. The screening test described in the current study was easily applied but has limitations.

First, the measure was derived from parental report and is

potentially unreliable (Reilly et al., 1996; Stallings et al., 1996). Second, replication with a large sample is required to yield precise estimates of sensitivity and specificity (Gigerenzer, 2002). Third, and most importantly, there are limited normative data available for feeding problems in the general population, and this affects the diagnostic accuracy of the screening test itself (Coughlin and Pickle, 1992). In these circumstances it is conventional to choose a cut-point with high specificity and sacrifice some sensitivity and in doing so reduce the incidence of false positives. This ameliorates consequences such as psychological harm associated with incorrect selection and unnecessary consumption of services (Gigerenzer, 2002). However, we choose to retain a high sensitivity in this investigation because the risk of missing true cases (babies with poor feeding skills) was viewed as unacceptable because of the potential impact on growth, development, and well-being (Field and Vega-Lahr, 1984; Felix-Schollaart et al., 1992; Neiman and Savage, 1997; Endriga and Kapp-Simon, 1999; Young et al., 2001). Furthermore, incorrectly identifying some adequate feeders as poor feeders was unlikely to cause psychological or physical harm or consume excessive health resources. Anecdotal evidence from our cohort also suggested that mothers welcomed the opportunity to have their babies' feeding skills examined whether there was a problem or not. This was probably because there is high parental concern about feeding in the neonatal period (Young et al., 2001).

The validation process confirmed that the sample was well distributed for CL, CLP, and PRS. However, there was an overrepresentation of CP and an unexpectedly high rate of PRS(+). Furthermore, most participants were recruited via the Royal Children's Hospital, so they were more typical of a clinical sample than the broader Victorian population. Because participants with PRS(+) had a greater risk of poor feeding skills compared with other members of the cohort, and because they were overrepresented in the current sample, the prevalence figures reported for poor feeding skills may reflect an overestimation of the true prevalence in the Victorian population. Nevertheless, the high prevalence of poor feeders in this clinical cohort suggests that early detection and management of feeding difficulties is important.

## CONCLUSION

Poor feeding skills were prevalent in a clinical cohort of newborns with CL/P and in some cases persisted to 14 months of age. Oral motor dysfunction, feeding sequelae, and poor feeding efficiency were characteristic of the problem. Babies with identifiable syndromes and PRS were greatly at risk of early, and in some cases persistent, feeding problems.

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## REFERENCES

- Bannister P. Early feeding management. In: Watson ACH, Sell DA, Grunwell P, eds. *Management of Cleft Lip and Palate*. London: Whurr; 2001:137–147.
- Brogan WF, Foulner DM, Turner R. A videoradiographic investigation of the position of the tongue prior to palatal repair in babies with cleft lip and palate. *Cleft Palate J*. 1987;24:336–338.
- Campo-Paysaa A. Treatment of labio-palate clefts. *Pediatric*. 1987;42:697–703.
- Carlisle D. Feeding babies with cleft lip and palate. *Nurs Times*. 1988;94:59–60.
- Choi BH, Kleinheinz J, Joos U, Komposch G. Sucking efficiency of early orthopaedic plate and teats in infants with cleft lip and palate. *Int J Oral Maxillofac Surg*. 1991;20:167–169.
- Clarren SK, Anderson B, Wolf LS. Feeding infants with cleft lip, cleft palate, or cleft lip and palate. *Cleft Palate J*. 1987;24:244–249.
- Coughlin SS, Pickle LW. Sensitivity and specificity-like measures of the validity of a diagnostic test that are corrected for chance agreement. *Epidemiology*. 1992;3:178–181.
- CSAG report—Clinical Standards Advisory Group. *Cleft Lip and Palate*. London: HMSO; 1988.
- Drillien CM, Ingram TTS, Wilkinson EM. *The Causes and Natural History of Cleft Lip and Palate*. Baltimore: Williams and Wilkins; 1966.
- Endriga MC, Kapp-Simon KA. Psychological issues in craniofacial care: state of the art. *Cleft Palate Craniofac J*. 1999;36:3–11.
- Felix-Schollaart B, Hoeksma JB, Prahl-Andersen B. Growth comparison between children with cleft lip and/or palate and controls. *Cleft Palate Craniofac J*. 1992;29:475–480.
- Field TM, Vega-Lahr N. Early interaction between infants with cranio-facial anomalies and their mothers. *Infant Behav Dev*. 1984;7:527–530.
- Fletcher RH, Fletcher SW, Wagner EH. *Clinical Epidemiology: The Essentials*. 2nd ed. Baltimore: Williams and Wilkins; 1988.
- Gewolb I, Vice F, Schweitzer-Kenney E, Taciak V, Bosma J. Developmental patterns of rhythmic suck and swallow in preterm infants. *Dev Med Child Neurol*. 2001;43:22–27.
- Gigerenzer G. *Reckoning With Risk: Learning to Live With Uncertainty*. London: Penguin Books; 2002.
- Glass RP, Wolf LS. Technology. Feeding management of infants with cleft lip and palate and micrognathia. *Infants Young Child*. 1999;12:70–81.
- Jones BW. Weight gain and feeding in the neonate with cleft: a three-center study. *Cleft Palate J*. 1988;25:379–384.
- Jones JE, Henderson L, Avery DR. Use of a feeding obturator for infants with severe cleft lip and palate. *Spec Care Dent*. 1982;2:116–120.
- Lau C, Schanler RJ. Oral motor function in the neonate. *Clin Perinatol*. 1996; 23:161–178.
- Lau C, Sheena H, Shulman R, Schanler R. Oral feeding in low birth weight infants. *J Pediatr*. 1997;130:561–569.
- Malek R, Psaume J. Nouvelle conception de la chronologie et de la technique chirurgicale du traitement des fentes labiopalatines. Resultats sur 220 cas. *Ann Chir Plast Esthet*. 1983;28:237–247.
- Medoff-Cooper B, Verklan T, Carlson S. The development of sucking patterns and physiologic correlates in very-low-birth-weight infants. *Nurs Res*. 1993; 42:100–105.
- Miller CK, Kummer AW. Feeding problems of infants with cleft lip/palate or craniofacial anomalies. In: Kummer AW, ed. *Cleft Palate and Craniofacial Anomalies: The Effects on Speech and Resonance*. San Diego: Singular Thomson Learning; 2001:103–127.
- Miller-Schuberth L. The role of occupational therapy in diagnosis and management. In: Tuchman DN, Walter RS, eds. *Disorders of feeding and swallowing in infants and children: pathophysiology, diagnosis and treatment*. San Diego, CA: Singular Publishing Group; 1994:115–130.
- Mizuno K, Ueda A. Development of sucking behavior in infants who have not been fed for 2 months after birth. *Pediatr Int*. 2001;43:251–255.
- Mizuno K, Ueda A, Kani K, Kawamura H. Feeding behavior of infants with cleft lip and palate. *Acta Paediatr*. 2002;91:1227–1232.
- Motion S, Northstone K, Emond A, and the ALSPAC study team. Persistent early feeding difficulties and subsequent growth and developmental outcomes. *Ambul Child Health*. 2001;7:231–237.
- Neiman GS, Savage HE. Development of infants and toddlers with clefts from birth to three years of age. *Cleft Palate Craniofac J*. 1997;34:218–225.
- Newman LA, Keckley C, Petersen MC, Hamner A. Swallowing function and medical diagnoses in infants suspected of dysphagia. *Pediatrics*. 2001;108: E106.
- Oliver RG, Jones G. Neonatal feeding of infants born with cleft lip and/or palate: parental perceptions of their experience in South Wales. *Cleft Palate Craniofac J*. 1997;34:526–532.
- Palmer MM, Crawley K, Blanco IA. Neonatal Oral-Motor Assessment scale: a reliability study. *J Perinatol*. 1993;13:28–35.
- Reid JA. A review of feeding interventions for infants with cleft palate. *Cleft Palate Craniofac J*. 2004;41:268–278.
- Reilly S, Skuse D, Poblete X. Prevalence of feeding problems and oral motor dysfunction in children with cerebral palsy: a community survey. *J Pediatr*. 1996;129:877–882.
- Reilly S, Skuse DH, Wolke D. *SOMA: The Schedule of Oral Motor Assessment*. London: Whurr Publishers; 2000.
- Riley M, Halliday J. *Birth Defects in Victoria, 1983–1998. Perinatal Data Collection Unit*. Melbourne: Victorian Department of Human Services; 2000.
- Schwartz M, Polgar S. *Statistics for Evidence-Based Health Care*. Croydon: Tertiary Press; 2003.
- Shah CP, Wong D. Management of children with cleft lip and palate. *Can Med Assoc J*. 1980;122:19–24.
- Sidoti EJ, Shprintzen RJ. Pediatric care and feeding of the newborn with a cleft. In: Shprintzen RJ, Bardach J, eds. *Cleft Palate Speech Management: A Multidisciplinary Approach*. St. Louis: Mosby; 1995:63–73.
- Spiestersbach DC, Dickson DR, Fraser FC, Horowitz SL, McWilliams BJ, Paradise JL, Randall P. Clinical research in cleft lip and cleft palate: the state of the art. *Cleft Palate J*. 1973;10:113–165.
- Stallings VA, Zemel BS, Davies JC, Cronk CE, Charney EB. Energy expenditure of children and adolescents with severe disabilities: a cerebral palsy model. *Am J Clin Nutr*. 1996;64:627–634.
- Styer GW, Freeh K. Feeding infants with cleft lip and/or palate. *J Obstet Gynecol Neonatal Nurs*. 1981;10:329–332.
- Trenouth MJ, Campbell AN. Questionnaire evaluation of feeding methods for cleft lip and palate neonates. *Int J Paediatr Dent*. 1996;6:241–244.
- Wolf LS, Glass RP. *Feeding and Swallowing Disorders in Infancy: Assessment and Management*. Tucson: Therapy Skill Builders; 1992.
- Young JL, O’Riordan M, Goldstein JA, Robin NH. What information do parents of newborns with cleft lip, palate, or both want to know? *Cleft Palate Craniofac J*. 2001;38:55–58.
- Zickefoose M. Feeding problems of children with cleft palate. *Children*. 1957; 4:225–228.