The Nature of Feeding in Infants With Unrepaired Cleft Lip and/or Palate Compared With Healthy Noncleft Infants

A.G. Masarei, B.App.Sc., Ph.D., M.R.C.S.L.T., D. Sell, Ph.D., M.R.C.S.L.T., F.R.C.S.L.T., A. Habel, M.B.Ch.B., F.R.C.P., M.R.C.P.C.H., Michael Mars, D.Sc.(Hon.), Ph.D., B.D.S., F.D.S., D.Orth., F.R.C.S.L.T., B.C. Sommerlad, M.B., F.R.C.S.(Eng.), A. Wade, B.Sc., Ph.D., C.Stat., I.L.T.M.

Objective: **Feeding difficulties are reported widely in infants with cleft lip and/or palate. There is, however, a paucity of objective information about the feeding patterns of these infants. This study compared patterns of feeding in infants with unrepaired cleft lip and palate with healthy noncleft infants of a similar age.**

Setting: **North Thames Regional Cleft Centre. The noncleft cohort was recruited from West Middlesex University Hospital, a general hospital with similar demographics.**

Participants: **Fifty newborn infants with nonsyndromic complete unilateral cleft lip and palate or a cleft of the soft and at least two thirds of the hard palate who were referred to the North Thames Regional Cleft Centre participated. Parents of 20 randomly selected, noncleft infants agreed to participate.**

Main Outcome Measures: **Feeding patterns were rated using the Neonatal Oral Motor Assessment Scale. Additional objective information was collected using the Great Ormond Street Measurement of Infant Feeding (Masarei et al., 2001; Masarei, 2003).**

Results: **Infants with nonsyndromic complete unilateral cleft lip and palate or a cleft of the soft and at least two thirds of the hard palate had less efficient sucking patterns than their noncleft peers had. They used shorter sucks (mean difference, 0.30 second; $p < .0005$), a faster rate of sucking (mean difference, 34.20 sucks/second; $p < .0005$), higher suck-swallow ratios (mean difference, 1.87 sucks/swallow; $p < .0005$), and a greater proportion of intraoral positive pressure generation (mean difference, 45.97% positive pressure; $p < .0005$).**

Conclusions: **This study demonstrated that the sucking patterns of infants with nonsyndromic complete unilateral cleft lip and palate or a cleft of the soft and at least two thirds of the hard palate differ from those of their noncleft peers.**

KEY WORDS: *feeding, infants, pressure generation, sucking patterns, unrepaired cleft palate, unrepaired complete unilateral cleft lip and palate*

Feeding difficulties in the cleft lip and palate population were first reported in 1619 by Fabricus of Aquapendente (cited in Jones, 1988). He recognized that infants with cleft lip and palate were unable to suck and often died of malnutrition. The first published descriptions of the nature of feeding problems were by Zickefoose (1957) and Tisza and Gumpertz (1962), and since then there have been only three more reports (Shelton et al., 1966; Brogan et al., 1987; Clarren et al., 1987). In addition, there are several review articles providing an overview of feeding patterns (Spriesterbach et al., 1973; Wolf and Glass, 1992; Glass and Wolf, 1999; Arvedson and Brodsky, 2002). The majority of reports are based on clinical observation and parental report and focus almost entirely on the oral stage of swallowing (Zickefoose, 1957; Tisza and Gumpertz, 1962; Clarren et al., 1987). They identify an inability to generate sufficient suction to extract milk from the nipple or teat, infants ''chewing the nipple'' in an attempt to compensate for their structural deficiency, nasal regurgitation and sticky or pasty foods sticking in the cleft, and frequent burping. There are reports of infants giving up and refusing to finish feeds

Dr. Masarei was previously Specialist Speech and Language Therapist, Great Ormond Street Hospital for Children NHS Trust, London, United Kingdom. Dr. Sell is Lead Speech and Language Therapist, North Thames Regional Cleft Centre, and Head of Speech and Language Therapy Department, Great Ormond Street Hospital for Children NHS Trust, London, United Kingdom. Dr. Habel is Consultant Pediatrician, Dr. Mars is Lead Orthodontist, and Dr. Sommerlad is Lead Consultant Plastic Surgeon, North Thames Regional Cleft Centre, Great Ormond Street Hospital for Children NHS Trust, London, United Kingdom. Dr. Wade is Senior Lecturer in Medical Statistics, Centre for Paediatric Epidemiology and Biostatistics Institute of Child Health, London, United Kingdom.

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Address correspondence to: Dr. Anthea Masarei, 31 Parry Street, Fremantle, Western Australia, 6160. E-mail amsppath@bigpond.net.au.

(Zickefoose, 1957; Tisza and Gumpertz, 1962). There is some disagreement about whether these infants have impairment at the pharyngeal stage of the swallow. Some authors report symptoms that may suggest reduced airway protection, such as choking when taking liquids and an increased incidence of chest infections (Zickefoose, 1957; Tisza and Gumpertz, 1962). In contrast, others report that infants with cleft lip and/or palate (CL/P) swallow normally (Shelton et al., 1966; Brogan et al., 1987; Clarren et al., 1987).

Brogan et al. (1987) objectively investigated the feeding patterns of infants with cleft lip and palate using videofluoroscopic assessment of feeding, the technique that is widely regarded as the ''gold standard'' assessment of swallowing in infants and children. This study, however, only reported on the oral stage, focusing exclusively on tongue positioning, and found that infants with cleft lip and palate have normal tongue movements during feeding. Masarei (2003) reported the results of a consecutive series of videofluoroscopic studies conducted on infants with unilateral cleft lip and palate (UCLP) or a cleft of the soft and at least two thirds of the hard palate (ICP). Oral and pharyngeal stage abnormalities were reported for all infants assessed.

One of the greatest concerns for parents of infants born with CL/P is the establishment of safe and efficient feeding. As many as 63% of infants born with CL/P experience feeding difficulties (Clarren et al., 1987). Among the consequences of feeding difficulties are poor growth and development (Ranalli and Mazaheri, 1975; Avedian et al., 1980; Jones, 1988; Felix-Schollaart et al., 1992; Lee et al., 1997), altered mother-child bonding (Speltz et al., 1994, 1997; Coy et al., 2002), decreased emotional well-being of the family, and increased burden of care (Adams et al., 1999). Another serious consequence of feeding problems is aspiration, which may lead to recurrent chest infections (Zickefoose, 1957; Taniguchi and Moyer, 1994).

In the United Kingdom, infants with CL/P usually are fed with an unrepaired cleft palate for at least the first 6 months of life, and surgical repair generally occurs between 6 months and 1 year of age (Clinical Standards Advisory Group Committee, 1998). Parents often report difficulties feeding their infants (Trenouth and Campbell, 1996; Oliver and Jones, 1997; Johansson and Ringsberg, 2004).

Given the paucity of information available about the nature of feeding in infants with nonsyndromic unrepaired CL/P, the aim of this study was to systematically investigate the extent and nature of feeding patterns, as compared with a cohort of similarly aged noncleft infants.

Methods

Context of the Study

This study was carried out within the North Thames Regional Cleft Centre (NTRCC) over a 4-year period. The West Middlesex University Hospital (WMUH) was chosen as the base for recruiting the noncleft cohort, because it fell within the catchment area of the NTRCC and had a similar ethnic and socioeconomic diversity.

Inclusion and Exclusion Criteria for Infants With UCLP or ICP

All infants referred to the NTRCC who were diagnosed with nonsyndromic UCLP or ICP were eligible for inclusion in the study.

Feeding can be affected by many factors, including medical conditions such as cardiac anomalies, prematurity, and neurological impairment. Therefore, infants who met the inclusion criteria but were born earlier than 34 weeks gestation, required cardiac surgery, and/or were diagnosed at birth with neurological impairment and/or a syndrome were excluded.

Inclusion and Exclusion Criteria for Noncleft Cohort

Infants born at the WMUH who were term and were born without any diagnosed congenital abnormality, confirmed by the pediatrician's routine examination within the first 24 hours, were eligible for inclusion in the noncleft cohort.

Procedure

Following the attainment of written informed consent, all recruited infants underwent feeding assessment in their first month of life. This was carried out in their home or, in a small number of cases of infants with UCLP or ICP, at the hospital.

The assessment included a short interview to collect demographic data, a medical history designed to identify risk factors for feeding and/or developmental problems, and a feeding history including information about whether the infant was being breast- or bottle-fed, designed to identify symptoms indicative of feeding difficulties.

All feeding assessments were carried out at a time when a routine feed was due. Each infant's sucking pattern was assessed and was rated as normal, disorganized, or dysfunctional using a standardized observational scale, the Neonatal Oral Motor Assessment Scale (NOMAS) (Braun and Meyer Palmer, 1990; Meyer Palmer et al., 1993). A speech and language therapist who was not involved in the study but was experienced in the assessment and management of pediatric feeding and was certified to administer the NOMAS made the ratings, based on video-recordings of the feed.

Infants also were assessed using the Great Ormond Street Measurement of Infant Feeding (GOSMIF) (Masarei et al., 2001; Masarei, 2003). This technique allowed reliable, objective assessment of several aspects of infant feeding that are reported to contribute to feeding efficiency, including measurement of pressures generated during bottle-feeding; identification of features of sucking patterns, such as number of sucks generated within each sucking burst, length of sucking bursts, and rate of sucking; and identification of swallows through auscultation (Masarei, 2003).

The first 5 minutes of each infant's feed was assessed with

TABLE 1 Demographic Details of Recruited Infants

		Noncleft Cohort n = 20	*Unilateral Cleft Lip and Palate n = 33*	*Isolated Cleft Palate n = 16*
Sex	Boys	8	21	9
	Girls	12	12	7
Gestational age (weeks)	Mean	39.64	39.76	39.75
	SD	1.34	1.32	1.23
	Minimum	37	36	38
	Maximum	42	42	42
Birth weight (kg)	Mean	3.20	3.38	3.38
	SD	0.46	0.44	0.54
	Minimum	2.12	2.33	2.43
	Maximum	4.08	4.10	4.20
Age at assessment (years)	Mean	0.06	0.04	0.04
	SD	0.01	0.02	0.02
	Minimum	0.03	0.01	0.01
	Maximum	0.09	0.09	0.08
Ethnic origin	Indian	5	1	4
	Bangladeshi	0	2	0
	Other Asian	0	2	0
	White / U.K.	12	23	12
	White / European	0	3	0
	Turkish	2	1	0
	Black African	1	1	0
Father's occupational classification	1	1	0	3
	2	4	8	3
	3	3	6	1
	5	2	5	5
	8	5	2	2
	9	4	3	2
	Unemployed or student	1	6	0

the GOSMIF. Each infant was fed by a parent or main caregiver. A 5% glucose solution was offered in preference to formula in order to ensure that sucking patterns were not influenced by the taste or nutritional content of the infant's familiar milk. A specified set of rules was used to select the first three ratable sucking bursts for each infant (Masarei, 2003). Length of sucking bursts, lengths of individual sucks or peak-to-peak intervals, rate of sucking, suck-swallow ratios, and pressure generation within the feeding bottle each were selected for measurement as indicators of feeding efficiency. Sucking burst length was defined as the time from the first identifiable peak to the final identifiable peak on the sucking trace. The length of individual sucks was defined as the time between two consecutive peaks on the sucking trace. The rate of sucking was defined as the number of sucks generated per minute. The suck-swallow ratio described the number of sucks produced prior to triggering of a swallow during bottle-feeding. To successfully transfer liquid from the breast or bottle, infants must generate a combination of positive and negative pressures. This study measured percentage of pressure generated above the baseline pressure in the bottle, reflecting the amount of compression the infant used in transferring milk from the bottle to the mouth.

For the purposes of statistical analysis, the median assessment of each of the three sucking bursts studied per infant was used. However, in the graphical presentations, three measures per infant are shown in order to illustrate the variability of the measures within each infant. In some cases it was not possible to measure three sucking bursts and therefore, there may be fewer than three points shown. The use of *t* tests enabled comparisons between the combined cleft groups (UCLP and ICP) and the noncleft cohort for the medians of the three or fewer measures per infant.

RESULTS

A consecutive series of 60 parents whose newborn infants met the study inclusion criteria were approached. Thirty-three infants with UCLP and 16 infants with ICP were included in the study. Visits were made to the WMUH on six occasions. Inpatient mothers of 38 noncleft infants who met the inclusion criteria were approached regarding participating in the study. Parents of 20 infants agreed to participate.

Table 1 shows the demographic information. There were higher proportions of boys in the UCLP and ICP cohorts (UCLP, n = 20; ICP, n = 9; noncleft, n = 8). The gestational ages were similar across all groups (UCLP = 39.76 weeks; ICP = 39.75 weeks; noncleft = 39.64 weeks). Similarly, there was little difference between the mean birth weights of the infants across the groups (UCLP = 3.38 kg; ICP = 3.38 kg; noncleft = 3.20 kg). The mean age at assessment for infants with UCLP and ICP were 0.04 years. The noncleft infants were slightly older, with assessment at 0.06 years. Based on reported developmental changes in feeding patterns (Bosma, 1986; Qureshi et al., 2002), it was not thought that this small difference in the ages of infants at assessment would be clinically significant. The majority of infants were from Caucasian/U.K. origins, with the rest in the main of Asian origin.

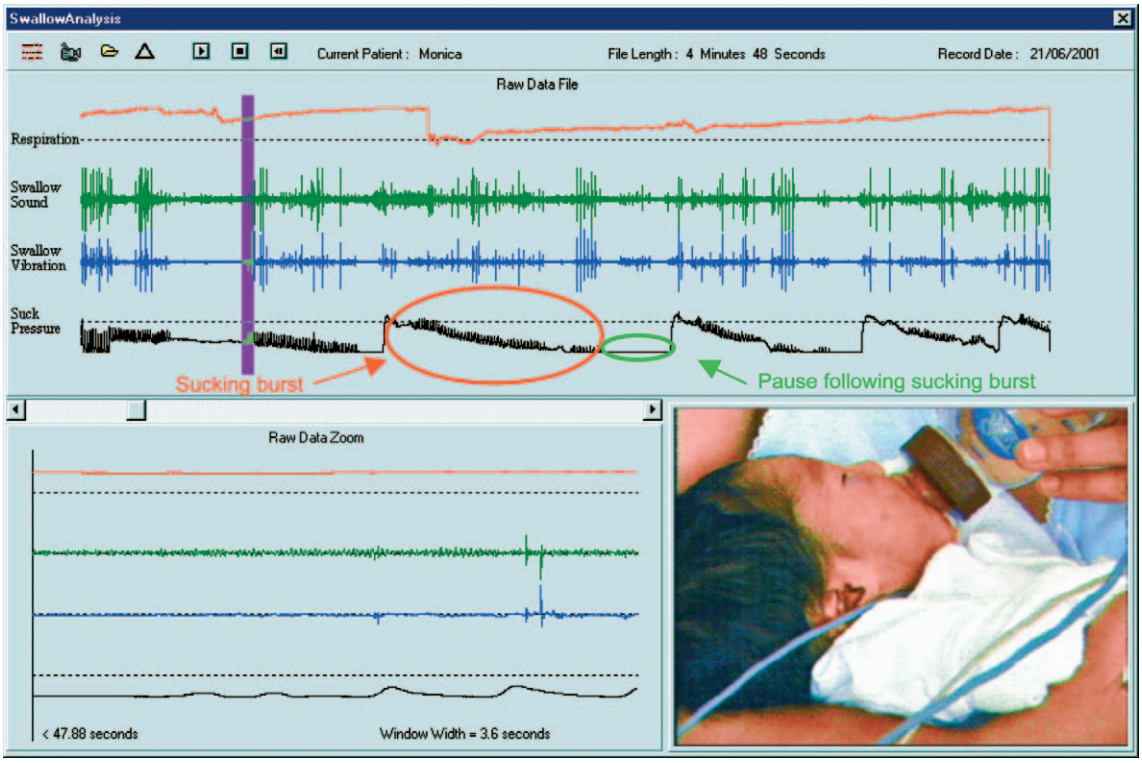

FIGURE 1 Screen shot of an infant's full 5-minute GOSMIF assessment.

NOMAS

All infants in the noncleft cohort were rated as having normal oral motor skills for feeding. In contrast, the majority of infants in the cleft cohort were rated as having disorganized (n = 30) or dysfunctional (n = 10) feeding. Two infants were rated as having normal oral motor skills for feeding and seven video-recordings were unratable.

GOSMIF Analysis

Overall Patterns of Sucking

Figure 1 is a screen shot of an infant's full 5-minute GOSMIF assessment. The top half of the screen shows a respiration trace (shown in red), swallow sounds used to identify when a swallow occurred (shown in blue and green), and a sucking trace (shown in black). In the lower half of the screen, the data within the purple band (spanning across all traces in the top half of the screen) is expanded. A video image of the infant is shown in the right lower corner. A typical sucking burst is shown within the red oval and a pause following this sucking burst is shown within the green oval. Three distinct sucking patterns were found. All noncleft infants produced an organized and rhythmic sucking/pause pattern over the 5-minute assessment (Fig. 1). In contrast, infants with UCLP or ICP used either of two patterns. In one, the infants did not produce any identifiable sucking burst, but used a continuous and disorganized pattern (Fig. 2); in the other, they generated a short but rhythmic sucking burst/pause pattern. This pattern was maintained for the start of the study, generally 2 or 3 minutes, followed by disorganization (Fig. 3).

Length of Sucking Bursts

The infants with UCLP or ICP used shorter sucking bursts (mean = 8.97 seconds) than did the noncleft cohort (mean = 13.28 seconds) (Fig. 4). This difference was statistically significant ($p = .002$, 95% confidence interval = −7.02 to −1.60).

Rate of Sucking

Infants with UCLP or ICP sucked at an average rate of 109.26 sucks per minute, in contrast to the noncleft infants, who sucked at an average rate of 75.07 sucks per minute (Fig. 5). There was a mean difference of 34.20 sucks per minute. This difference was significantly different ($p < .0005$, 95% confidence interval = 27.30 to 41.09).

Length of Individual Sucks/Peak-to-Peak Intervals

As might be expected, given the faster rate of sucking, infants with UCLP and ICP used shorter sucks than did the noncleft cohort, with the cleft cohort using a mean length of sucks of 0.57 seconds and the noncleft cohort using a mean length of 0.87 seconds (Fig. 6). The mean difference in the length of individual sucks between the cleft and noncleft cohort was

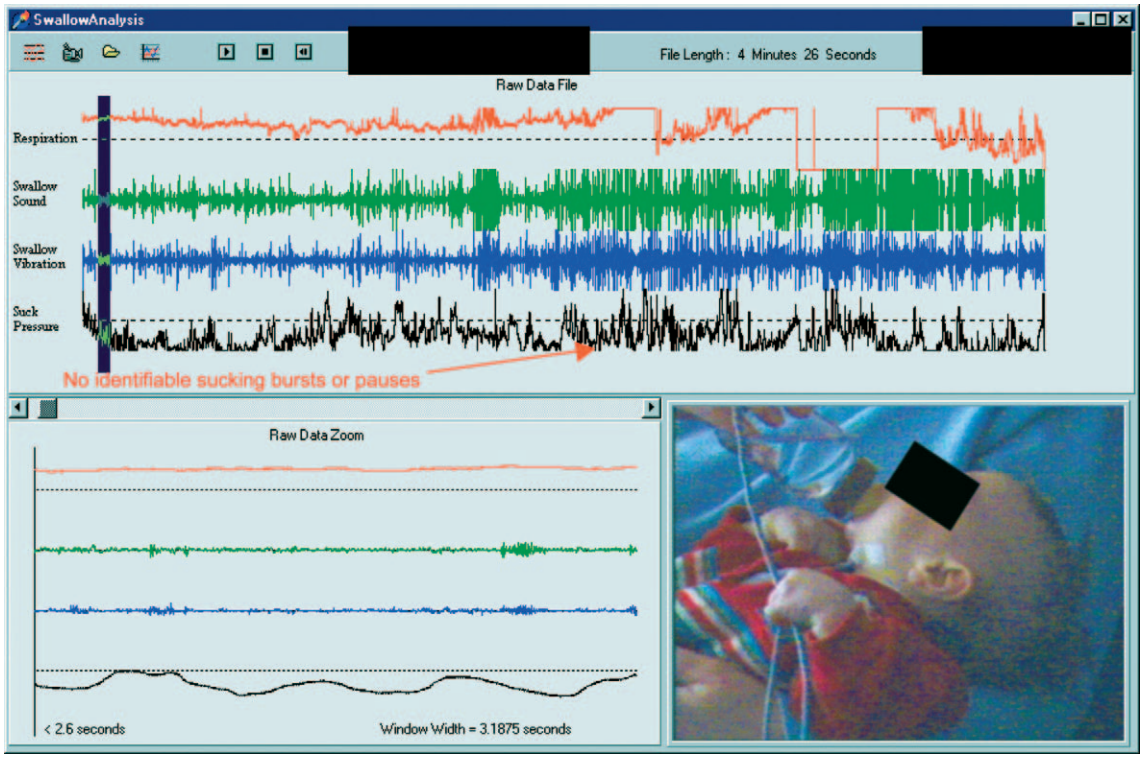

FIGURE 2 Some infants did not produce any identifiable sucking burst, but used a continuous and disorganized pattern.

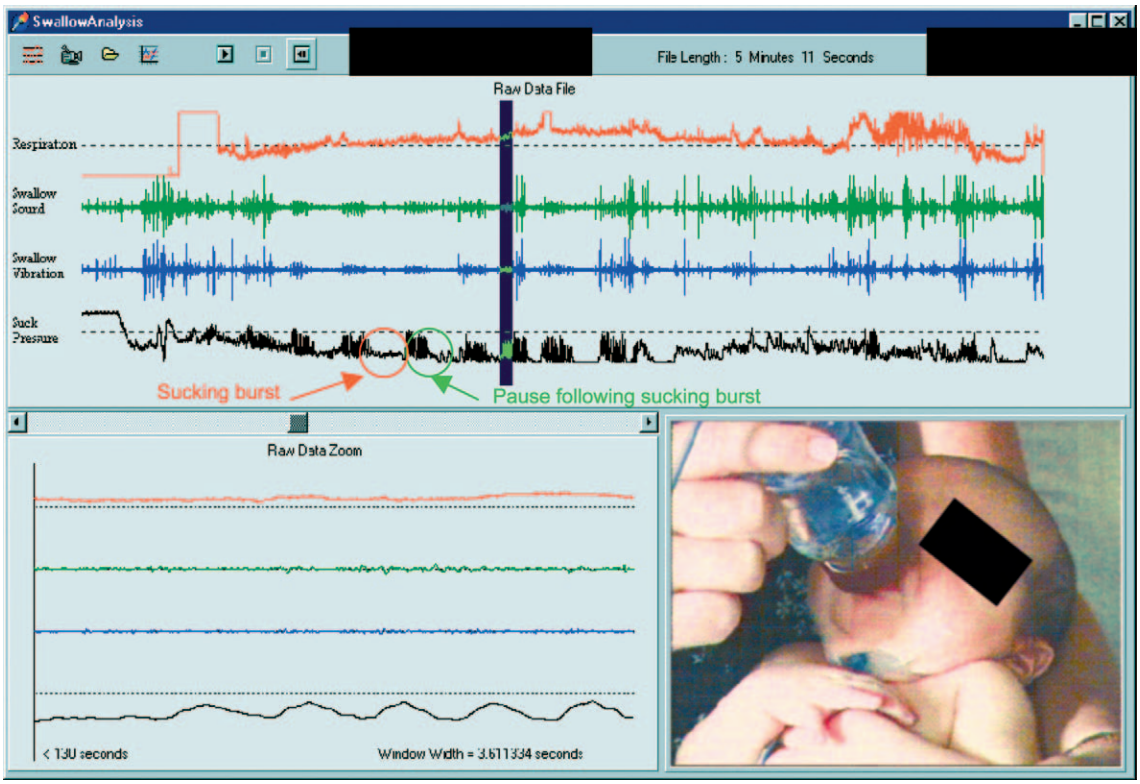

FIGURE 3 Second sucking pattern characterized by the generation of a short but rhythmic sucking burst/pause pattern.

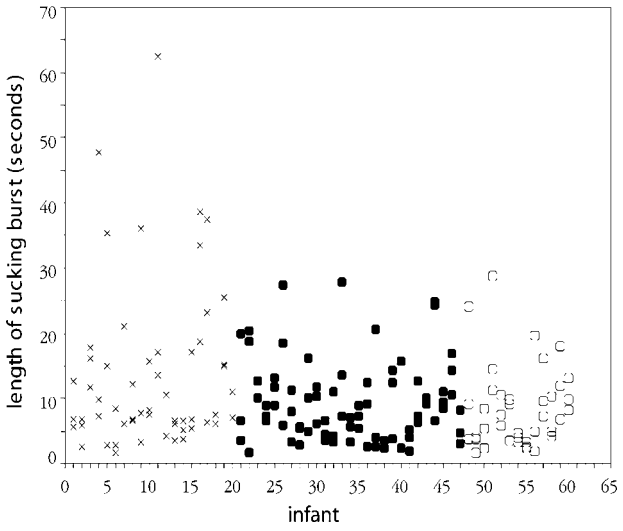

FIGURE 4 Length of sucking bursts (× = noncleft infants; ● = infants with unilateral cleft lip and palate; ○ = infants with isolated cleft palate extending two thirds into the hard palate).

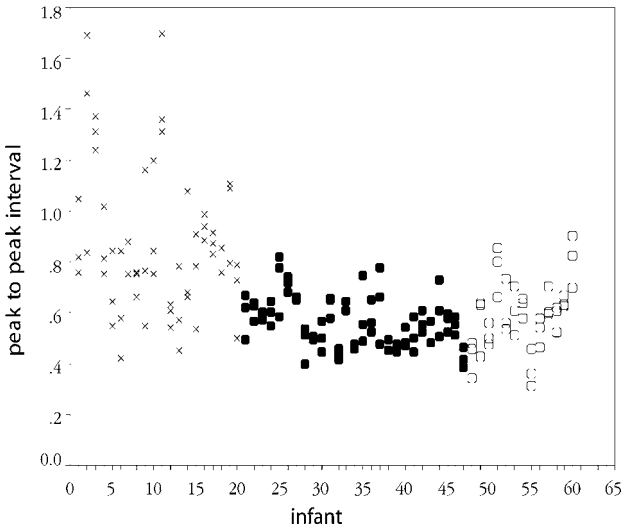

FIGURE 6 Length of individual sucks (× = noncleft infants; ● = infants with unilateral cleft lip and palate; ○ = infants with isolated cleft palate extending two thirds into the hard palate).

−0.30 seconds, and this difference was statistically significant ($p < .0005$, 95% confidence interval = −0.36 to −0.24).

Pressure Generation

Infants in the noncleft cohort generated a small proportion of positive pressure (mean = 25.71%), whereas the infants in the cleft cohort generated a mean of 71.68% (Fig. 7). The difference between the cohorts (45.97%) was statistically significant ($p < .0005$, 95% confidence interval = 36.61 to 55.33).

Suck-Swallow Ratio

Infants in the cleft cohort produced higher mean suck-swallow ratios (2.97:1) than did the infants in the noncleft cohort (1.20:1) (Fig. 8). This difference (1.87) was statistically significant ($p < .0005$, 95% confidence interval = 1.34 to 2.40).

Discussion

The results of this trial demonstrate the differences between the feeding patterns of infants with UCLP and ICP, as compared with those of a noncleft cohort. The NOMAS feeding assessment suggested that the majority of infants with UCLP and ICP had disorganized or dysfunctional feeding patterns. The NOMAS classified infants' feeding patterns based on a series of observations of tongue and jaw movements (Braun and Meyer Palmer, 1990). On closer examination of these individual items, it was evident that there were several common features resulting in the classification of disorganized and dys-

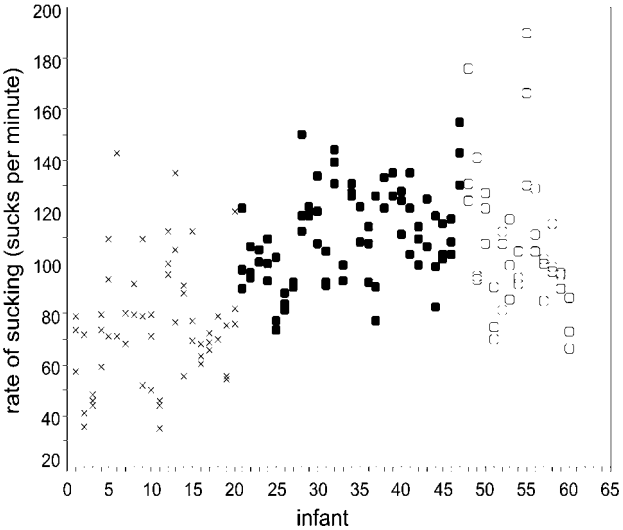

FIGURE 5 Rate of sucking (× = noncleft infants; ● = infants with unilateral cleft lip and palate; ○ = infants with isolated cleft palate extending two thirds into the hard palate).

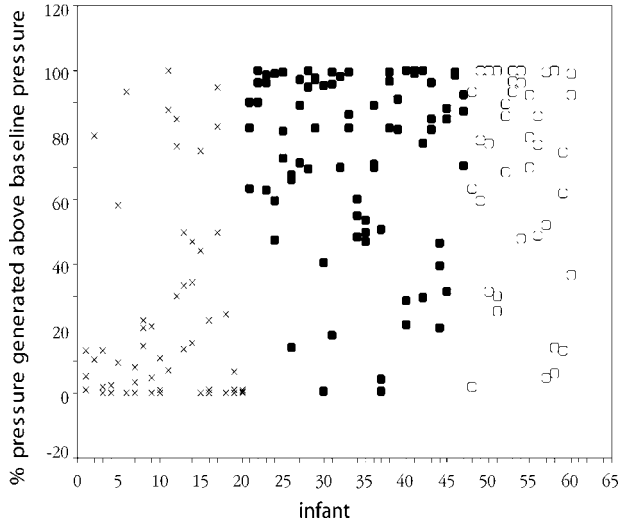

FIGURE 7 Pressure generation (× = noncleft infants; ● = infants with unilateral cleft lip and palate; ○ = infants with isolated cleft palate extending two thirds into the hard palate).

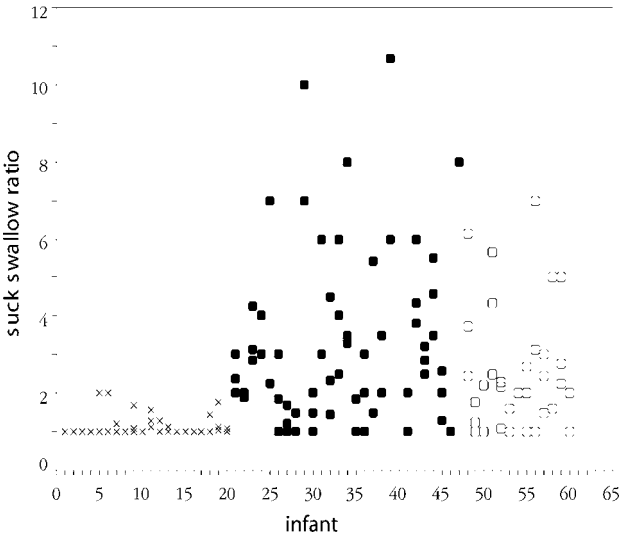

FIGURE 8 Suck-swallow ratio (× = noncleft infants; ● = infants with unilateral cleft lip and palate; ○ = infants with isolated cleft palate extending two thirds into the hard palate).

functional feeding in the cleft cohort. These features included inconsistent range of tongue and jaw movements, arrhythmical tongue and jaw movements, and altered rate of sucking. When the patterns were examined more closely with the GOSMIF, further differences between the cleft and noncleft cohort were evident. The measures of sucking selected for analysis (length of sucking burst, rate of sucking, length of individual sucks or peak-to-peak intervals, percentage of positive pressure generated, and the suck-swallow ratio) are thought to be related to feeding efficiency (Arvedson et al., 2002). All of these measures are closely related to the motor components of feeding. Infants with CL/P used shorter sucking bursts, but their rate of sucking was significantly faster and the length of individual sucks was shorter than that of the noncleft infants. There may be several explanations for this difference. Several authors have reported that when the flow rate of bottle feeds was increased, the infant's rate of sucking slowed and the length of individual sucks lengthened (Ardran and Kemp, 1951; Christensen et al., 1976; Mathew, 1988, 1991; Mathew and Bhatia, 1989; Eishima, 1991). Conversely, if the flow rate was decreased, the rate of sucking increased and the length of individual sucks decreased (Ardran et al., 1958a; Christensen et al., 1976; Mathew and Bhatia, 1989; Eishima, 1991; Mathew, 1991). It was, therefore, not unexpected that in infants with CL/P, where the bolus size was smaller and the flow rate slower, the rate of sucking was faster and the length of individual sucks was shorter.

To transfer fluid from the breast or bottle, infants must generate a combination of positive and negative pressures (Ardran et al., 1958a, 1958b; Colley and Creamer, 1958; Logan and Bosma, 1967; Wolf and Glass, 1992; Arvedson and Brodsky, 2002). Infants with CL/P do not, however, have the intraoral structures to allow negative pressure generation. Predictably, in this study the group of noncleft infants generated more negative pressure during GOSMIF assessment than did the cleft cohort. The predominance of positive pressure generation seen in the cleft cohort supports the suggestion of several authors that infants with CL/P use a characteristic chomping or biting action on the teat (positive pressure generation) in an attempt to transfer milk (Zickefoose, 1957; Tisza and Gumpertz, 1962; Clarren et al., 1987).

In this study, the cleft cohort produced higher suck-swallow ratios than did the noncleft cohort. This increase in the number of sucks required to obtain an adequate bolus was probably an indicator of inefficient suck and the increased attempts at transferring fluid from the bottle before having a sufficient size bolus to trigger a swallow.

Conclusion

The results of this study suggest that the nature of feeding difficulties in infants with CL/P is more complex than being purely a reduced ability to generate the intraoral pressures required for breast- or bottle-feeding. It seems likely that many of the differences seen were related to the cleft infants' attempts to compensate for their anatomical abnormalities. In recommending the use of assistive feeding devices and safe swallowing strategies, it is therefore imperative that thorough and, where possible, objective assessments/reviews are administered, evaluating the impact of recommendations on feeding patterns.

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