

Academy of Breastfeeding Medicine
BREASTFEEDING INFANTS WITH CLEFT LIP AND/OR PALATE
Annotated bibliography

Introduction: A total of 164 studies were initially reviewed. A few studies originally presented in the bibliography were later excluded from this annotated review on the basis that, after careful examination, they did not contain information relevant to the topic area. Descriptions of programs, discussion of incidence, prevalence or management of infants with cleft lip and/or palate in general (where feeding/breastfeeding was not mentioned/included), description of the manufacture or types of orthopaedic plates (palatal obturators), and studies that focused on congenital malformations with results for infants with cleft lip and/or palate not reported separately from the group all fall into this category, and are not included below.

Additionally, a large amount of literature in this area constitutes professional/expert opinion (Level III). As much of this literature is repetitive, the recommendations of the authors have been summarized, rather than individually annotated (see attached document). Level III studies that are 'data-driven' (e.g., descriptive studies, case reviews) have been individually annotated below. For the purposes of this bibliography, the abstracts have been summarized (where written in English), however it was not always possible to classify the level of evidence on the basis of the English abstract

In total, 56 studies are individually annotated below, and 45 (representing expert opinion) are summarized in the attached document. For the purpose of this summary and annotated bibliography, the following shortened forms are used:

CL= cleft lip only

CP= cleft palate only

CLP= cleft lip and palate

CL/P= cleft lip and/or palate (usually when multiple groups are included, or the specific population is not adequately described.

PRS= Pierre Robin Sequence

Reference	Comment	Level of Evidence
Glenny AM, Hooper L, Shaw WC, Reilly S, Kasem S, Reid J. Feeding interventions for growth and development in infants with cleft lip, cleft palate or cleft lip and palate (Review). Cochrane Database of Systematic Reviews . 2005(4):CD003315. Study type: Systematic review	Review of feeding interventions for infants with CL/P. The focus of the review is all feeding interventions, not just breastfeeding. With regard to breastfeeding, the authors conclude that there is weak evidence for the use of breastfeeding over spoon feeding following surgery for CL (based on Darzi).	I
Prahl C, Kuijpers-Jagtman AM, van't	Infants with complete unilateral CLP were randomised to intervention (passive	I

<p>Hof MA, Prah-Andersen B. Infant orthopedics in UCLP: Effect on feeding, weight, and length: A randomized clinical trial (Dutchcleft). Cleft Palate Craniofac J. 2005;42(2):171-177</p> <p>Study type: Randomized controlled trial.</p>	<p>maxillary plates) and non-intervention groups (n=27 in each group). Bottle feeding velocity, weight and length for age and weight for length measures were taken, and compared to reference norms. Results: no significant differences between the groups were found, although mean weight and height for age scores of both groups were significantly lower than the reference norms.</p>	
<p>Marques IL, de Sousa TV, Carneiro AF, de B. A. Peres SP, Barbieri MA, Bettiol H. Robin sequence: A single treatment protocol. Jornal de Pediatria (Rio J). 2005;81(1):14-22.</p> <p>Study type: Review of literature.</p>	<p>The authors reviewed recent literature on PRS to determine a treatment protocol for this syndrome. With regard to feeding, they indicate that the breathing difficulties of infants with PRS lead to difficulties in co-ordinating sucking, swallowing and breathing. Additionally, glossoptosis presents a barrier to suction, as the tongue tends to position posteriorly; and the cleft of the palate leads to poor suction. Usually, infants with PRS cannot be fed orally, and require NG tube feeding.</p>	I
<p>Reid J. A review of feeding interventions for infants with cleft palate. Cleft Palate Craniofac J. 2004;41(3):268-78.</p> <p>Study type: Systematic review.</p>	<p>A systematic review of feeding interventions for infants with CP. The focus is on all feeding interventions, not just breastfeeding. With regard to breastfeeding, the author concludes that there is weak evidence (mostly case studies or expert opinion) that infants with CL only experience little difficulty with breastfeeding, while infants with CP or CLP have more difficulty. There has been little empirical evaluation of the use of obturators to facilitate breastfeeding.</p>	I
<p>Masarei AG. An investigation of the effects of pre-surgical orthopaedics on feeding in infants with cleft lip and/or palate [PhD]. University College; 2003</p> <p>Study type: Randomized control trial.</p>	<p>Investigation of the use of presurgical orthopaedics (PSO) in feeding infants with unilateral CLP or isolated CP. Infants (N=34) were randomised to PSO or non PSO groups. Outcome measures were oral motor assessment and anthropometry at 12 months, and parent report of feeding at 3, 6 and 12 months, physiological feeding measures, and oral motor assessment at 3 and 6 months. Results: PSO had no significant effect on feeding or growth at 12 months. Non-statistically or clinically significant differences were found between groups at 6 months, with PSO group showing improvement.</p>	I
<p>Weatherley-White RC, Kuehn DP, Mirrett P, Gilman JI, Weatherley-White CC. Early repair and breast-feeding for infants with cleft lip. Plast Reconstr Surg. 1987;79(879-885).</p> <p>Study type: Descriptive</p>	<p>60 mothers with infants undergoing lip repair surgery offered the option of breastfeeding immediately following surgery, resulting in three groups: breastfeeding for 6 weeks (n=16), cup or syringe feeding (n=22) and breastfeeding for less than 6 weeks then changing to bottle (n=22). Only one complication (partial wound dehiscence) was noted in a cup fed infant. At 3 months, breastfeeding infants gained 67% in weight compared to pre-operatively, cup/syringe fed group gained 47% and the group that changed to bottle before 6 weeks gained 66%. With CL infants only, 1 and 3 month weight gains were significantly greater for breastfed infants.</p>	II-1
<p>Gopinath VK, Muda WA. Assessment of growth and feeding practices in children with cleft lip and palate. Southeast Asian J Trop Med Public Health.</p>	<p>Infants with CLP (n=60) and a normal control group (n=161) were compared for feeding practices, growth (at baseline at 6 months old), and number of infections. Mothers of normal infants were significantly more positive about breastfeeding, infants with CLP were more susceptible to infections and grew at a significantly</p>	II-2

<p>2005;36(1):254-258.</p> <p>Study type: Naturalistic comparison study.</p>	<p>lower rate than normal infants. Of the 20 mothers with infants with CLP up to 24 months old, 15 had attempted breastfeeding, but only 8 were successful. 90% were bottle feeding at the time of the study, and 73% used formula milk.</p>	
<p>Erkkila AT, Isotalo E, Pulkkinen J, Haapenen ML. Association between school performance, breast milk intake and fatty acid profile of serum lipids in ten-year-old cleft children. J Craniofac Surg. 2005;16(5):764-9.</p> <p>Study type: Cohort descriptive</p>	<p>Ten year old children with CL/P were studied (n=53) to determine fatty acid profiles of serum lipids. Profiles were compared to parent reported breastfeeding in infancy, and correlated to school performance. Shorter periods of breast milk intake (<1 month) was associated with lower school performance for children with CL/P.</p>	<p>II-2*</p>
<p>AnianssonG,SvenssonH,Becker M, Ingvarsson L. Otitis media and feeding with breast milk of children with cleft palate. Scand J Plast Reconstr Surg Hand Surg. 2002;36(1):9-15.</p> <p>Study type: Retrospective analysis with control group.</p>	<p>Incidence of acute and secretory otitis media in children aged 6-10 years with three groups: CP/CLP (N=48), CL (N=15) and normal controls (N= 21). Data were obtained retrospectively from medical records. Questionnaires about feeding with breastmilk were used to correlate OM with duration breast milk feeding (not necessarily breast fed). Findings: significant correlation during 0-18months between increased duration of feeding with breastmilk and lower OM incidence across the whole group. Strong but non-significant correlation between longer feeding and OM for CP/CLP group. Children with CP and CLP had a higher incidence (62%) of OM than controls (14%) or CL only (0%) at 0-18 months, and again at 0-6 years (90% CP or CLP, 80% CL, 48% controls).</p>	<p>II-2*</p>
<p>Chen Z, Chen J, Wu J, Zhang X. Factors involved in intelligent development of children with cleft lip and palate [Chinese]. Hua Xi Kou Qiang Yi Xue Za Zhi. 2001;19(3):174-177.</p> <p>Study type: Descriptive cohort with comparison group</p>	<p>Summary of abstract (article in Chinese): IQ and developmental quotient, and factors impacting upon these, of 152 children with CL/P, compared to 80 healthy children. Findings: Children with CL or CP only had higher IQs than those with CLP. The mean IQ value of those fed with breast milk was higher than those fed with formula.</p>	<p>II-2**</p>
<p>Choi, BH, Kleinheinz, J., Jooz, U., & Komposch, G. Sucking efficiency of early orthopaedic plate and teats in infants with cleft lip and palate. International Journal of Oral Maxillofacial Surgery 1991; 20:167-169</p> <p>Study type: Comparative group study</p>	<p>Intra-oral negative pressure (IONP) was measured in infants with CL/P (some with orthopaedic plates) and normal infants (n=28 total). All infants with CLP or CP were unable to create any IONP with or without an orthopaedic plate. Infants with CL and infants with repaired CL/P were able to create IONP using regular Nuk or Cleft Nuk nipple, and the mean IONP was similar to that of normal infants.</p>	<p>II-2</p>
<p>Paradise JL, Elster BA, Tan L. Evidence</p>	<p>Data collected on 315 infants with CL/P relating to breast milk intake and otitis</p>	<p>II-2</p>

<p>in infants with cleft palate that breast milk protects against otitis media. Pediatrics. 1994;94(6 Pt 1):853-60.</p> <p>Study type: Prospective and retrospective cohort.</p>	<p>media was analysed. Data on 224 infants were retrospective, based on previously collected data (therefore blinded in relation to this study)- data on a further 91 infants were collected for the study (unblinded). Findings: 54 (17.1%) infants were fed breast milk for varying periods- usually through compressible feeder. These infants were significantly more likely to have visits with no middle ear effusion in one or both ears.</p>	
<p>Cohen M, Marschall MA, Schafer ME. Immediate unrestricted feeding of infants following cleft lip and palate repair. J Craniofac Surg. 1992;3(1):</p> <p>Study type: Retrospective analysis of two protocols</p>	<p>Retrospective analysis of two postoperative (lip and palate) feeding protocols, with main outcome measure of wound dehiscence, scar appearance, palatal fistulae, early speech outcome. A total of N=80 infants, with approximately equal numbers in each group. Protocols were for A restricted tube or syringe feeding following surgery, or B for unrestricted bottle or breast feeding immediately after lip and following day after palate repair. Findings: No cases of wound dehiscence for the group using protocol B. There were no differences in final scars or early speech outcome across the groups. An 'overall impression' of weight gain (not objectively measured) was that infants using protocol B showed better weight gain and nutritional status than those with restricted feeding.</p>	II-2
<p>Avedian LV, Ruberg RL. Impaired weight gain in cleft palate infants. Cleft Palate J. 1980;12(1):24-6</p> <p>Study type: Respective survey with comparison</p>	<p>A survey of parents of children with CP (N=37), asking for actual weight records during the first 6 months of life, history of illnesses. Figures were plotted against standardised weight curves. Findings: A low birth weight (median 30th percentile) was recorded for CP children. Median weight decreased to 25th percentile at 3-5 months, and increased to 30th percentile at 6 months. Conclusions: weight fall of may be related to early feeding difficulties.</p>	II-2*
<p>Pandya AN, Boorman JG. Failure to thrive in babies with cleft lip and palate. Br J Plast Surg. 2001;54(6):471-5.</p> <p>Study type: Retrospective cohort review and prospective cohort audit (before and after with intervention).</p>	<p>In part A, charts of 147 children with CL/P undergoing primary cleft surgery were reviewed to identify incidents of failure to thrive. Findings: FTT rates were 32% for unilateral CLP, 38% for bilateral CLP, and 49% for CP alone. FTT for infants with Pierre Robin Sequence was 100%. In part B, changes to the protocol of the centre were instituted, including a feeding support nurse and supervised airway management for infants with PRS. 68 referrals were prospectively audited to determine changes in FTT rates. Rates showed a decrease to 9% for unilateral CLP, 20% for bilateral CLP and 26% for CP, and 40% for PRS.</p>	II-3
<p>Turner L, Jacobsen C, Humenczuk M, Singhal VK, Moore D, Bell H. The effects of lactation education and a prosthetic obturator appliance on feeding efficiency in infants with cleft lip and palate. Cleft Palate Craniofac J. 2001;38(5):519-24.</p> <p>Study type: Prospective baseline reversal design.</p>	<p>Baseline data on time and volume taken with breastfeeding and bottle feeding infants (N=8) with CL/P were recorded. Interventions were lactation education and application of prosthetic obturator, and data (time and volume) recorded. The obturator was then removed, and data again recorded. The obturator was returned and lactation support provided. Findings: In all infants, feeding times decreased significantly when fitted with obturator, following lactation education. Milk intake increased significantly with obturator. Infants were able to drink up to four times as much milk per minute when the obturator was in place.</p>	II-3

<p>Kogo M, Okada G, Ishii S, Shikata M, Iida S, Matsuya T. Breast feeding for cleft lip and palate patients, using the Hotz-type plate. Cleft Palate Craniofac J. 1997;34(4):351-3.</p> <p>Study type: Descriptive analysis of use of plate and success of breastfeeding.</p>	<p>Breastfeeding of 10 babies using the modified Hotz-type plate, with outcomes of sucking rate, milk intake, duration of use of plate assessed. Findings: Mean sucking rate 2.2g/min, mean total amount 22g of milk. Supplemental feeding still needed to produce weight gain. No reduction in weight gain compared to babies who were bottle fed with the Hotz plate. 4/10 babies continued to breastfeed with the plate, only one continued to 14 months (when weaned)- others gave up for various reasons (reduced supply of milk, baby taking long to feed). Intra oral negative pressure was still weak even with the plate.</p>	II-3
<p>Barbosa da Silva E, Barbosa Furia CL, Queiroz de Moraes Silveira Di Ninno C. Breast feeding in cleft lip and palate neonates: Study of feeding difficulties and methods [Spanish]. Rev CEFAC, Sao Paulo. 2005;7(1):21-28.</p> <p>Study type: Descriptive</p>	<p>Summary of abstract (article in Spanish): 50 mothers of infants with CLP were interviewed to discover feeding methods and difficulties. Findings: Breastfeeding was more successful in less complex clefts. Difficulty with sucking was a common reason for ceasing breastfeeding.</p>	III
<p>Garcez LW, Giuliani ER. Population-based study on the practice of breastfeeding in children born with cleft lip and palate. Cleft Palate Craniofac J. 2005;42(6):687-93.</p> <p>Study type: Descriptive cohort study</p>	<p>Interviews with 31 mothers of infants with CL/P used to determine frequency and duration of breastfeeding during the first year of life, and difficulties with breastfeeding. Participants were mothers of all children born in 2001 and 2002 with CL/P, within one city in Brazil. All mothers initiated breastfeeding, 67% initiated it exclusively (no other feeding method). Breastfeeding was maintained exclusively for a median of 15 days. Infants with CL only were more likely to be breastfed exclusively, and also breastfeed for significantly longer than those with CP or CLP- median breastfeeding for the CL group was similar to the general population. 1 infant with CLP and one with CP were exclusively breastfed, while 4 CLP and 1CP infants were fed using both direct breastfeeding with supplementary bottle feeding.</p>	III
<p>MacDonald SK. Caring for your newborn with cleft lip and/or cleft palate. Accessed at Prescription Parents at http://www.samizdat.com/pp2.html on 20 December 2005.</p> <p>Study type: Descriptive survey and expert</p>	<p>Suggestions for caring for newborns with CL/P, based on a questionnaire sent to members of Prescription Parents (study details not included). Results: a) "the mother must feel considerable commitment to breast feeding"; b) Infants with CL only may find it difficult to latch to the breast, mothers should remain as still as possible during feeding, and allow the baby to 'gum' the areola to stimulate 'let down' reflex; c) Infants with CP may need the breast supported in their mouth with the mother's forefinger and thumb, so that the nipple reaches to the back of the tongue without requiring the infant to maintain this through vacuum; and d) Infants with CP feed more easily when the breast is full.</p>	III
<p>Johansson B, Ringsberg KC. Parents' experiences of having a child with cleft lip and palate. J Adv Nurs. 2004;47(2):165-173.</p>	<p>Interviews of twenty families of children with CLP (2 girls, 8 boys), from one centre. Children aged from 1.5 months to 4.5 years, and had either unilateral CLP, bilateral CLP or bilateral CL. Families reported different varying experiences relating to breastfeeding, and were concerned with being given straightforward and consistent information about feeding. For example, if breastfeeding is likely to be difficult, this</p>	III

Study type: Qualitative phenomenological	should be stated and acknowledged by the whole care team (even if breastfeeding is still attempted or encouraged). Feeding is identified as a 'major issue' for parents.	
da Silva Dalben G, Costa B, Gomide MR, Teixeira das Neves LT. Breast-feeding and sugar intake in babies with cleft lip and palate. Cleft Palate Craniofac J. 2003;40(1):84-7. Study type: Descriptive.	Interviews of caregivers of infants with CL/P (n=200) enrolled at specialist craniofacial hospital, regarding breast feeding, sugar intake, food used, food offered in bottle, frequency of bottle use. Findings: 81% of infants had never been breastfed. Where breastfeeding had been attempted, 78.8% reported ceasing due to inability to suck. Babies with CP were less likely to be breastfed than babies with CL only. 84% of children who were breastfed for at least one month had CL only	III
Heinzel N, Baltzer J. Aspects of birth and breastfeeding from children with cleft palate [German]. Zentralblatt fur Gynakologie. 2003;125(10):393-7. Study type: Expert opinion.	Summary of abstract (article in German): The authors describe aspects of breastfeeding infants with CP and the importance of breastfeeding for nutrition	III**
Livingstone VH, Willis CE, Abdel-Wareth LO, Thiessen P, Lockitch G. Neonatal hypernatremic dehydration associated with breast-feeding malnutrition: a retrospective survey. Can Med Assoc J. 2000;162(5):647-52. Study type: Retrospective cohort review (descriptive)	Retrospective chart review of a cohort of infants who had developed hypernatremic dehydration when breastfed. 21 cases of hypernatremic dehydration were identified, and 'risk factors' leading to this state were identified, including: a) technical difficulties with 'latching' and positioning; b) sucking, swallowing and breathing difficulties of infant; c) facial or oral abnormalities, including CP; and d) restricted access to breast (not fully breastfed).	III
Stockdale HJ. Long-term expressing of breastmilk. Breastfeed Rev. 2000;8(3):19-22 Study type: Expert opinion/case study	The experience of a midwife whose baby was born with a bilateral CLP and was unable to breastfeed, despite prolonged attempts.	III
Cruz MJ, Kerschner JE, Beste DJ, Conley SF. Pierre Robin sequences: Secondary respiratory difficulties and intrinsic feeding abnormalities. Laryngoscope. 1999;109(1632-1636). Study type: Retrospective descriptive audit	Retrospective chart audit of management of infants with PRS, including methods for feeding management (n=47). 5 infants required long-term NG feeding and gastrostomy tube placement was required by 12 infants. Swallowing abnormalities not related to airway obstruction were diagnosed in 11%.	III

<p>Sykes L, Essop R. A feeding adaptation by an infant with a cleft palate. SADJ. 1999;54(8):369-370.</p> <p>Study type: Case study</p>	<p>Description: Report of infant with large CP and no obturator. This infant occluded her cleft with her own thumb under the bottle teat, pushing the thumb into the cleft.</p>	<p>III</p>
<p>Frischknecht K. Cleft palate prematurity feeding problems? Paper prepared for International Certification of Breastfeeding and Lactation Consultant 1998-1999; 1998.</p> <p>Study type: Case study</p>	<p>Case report of an infant with a large cleft of the hard and soft palate, born prematurely and with respiratory distress syndrome, bradycardia and mild pulmonary valve dysplasia. The infant was fitted with a palatal obturator at 26 days old. Although fed expressed milk via a bottle, the infant was introduced to the breast and seemed to enjoy the experience of suckling. The mother's breasts were large and soft, and she had a good let-down reflex and this was felt to aid the infant receiving some milk at the breast. The infant was gradually able to feed completely from a bottle with the palatal obturator in place. Breastfeeding, even though it was not the main method of nutrition for this infant, was felt to be important for security and bonding for the infant and mother.</p>	<p>III</p>
<p>Crossman, K. Breastfeeding a baby with a cleft palate: a case report. J Hum Lact. 1998;14(1):47-50</p> <p>Study type: Case study</p>	<p>Case report of breastfeeding an infant with a large cleft extending from behind the alveolar ridge to the uvula. Infant was unable to form a seal on the breast and could not compress the nipple against the palate to strip milk. Milk was expressed directly into her mouth at each feed for 5 minutes at each breast, with supplementary feed of expressed milk via squeeze bottle and soft teat. After fitting with a dental plate, the infants suckling and compression of the nipple increased. Postoperatively, the infant was not allowed to suck for 3 weeks, and was fed via spoon and cup. After 3 weeks, she was placed on the breast and fed successfully, with supplement from a cup.</p>	<p>III</p>
<p>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(6):526-32.</p> <p>Study type: Descriptive questionnaire</p>	<p>Questionnaires about feeding methods and perceptions of care sent to parents of infants with CL/P born in South East Wales, with 100 responses (64%). The majority of mothers intended to breastfeed prior to birth. Bottle feeding was most common after birth (in hospital) and once at home. 90% of infants with CP and 85% of infants with CLP were bottle fed at home, while 50% of infants with CL were at least partially breastfed. Only 3% of infants with CP and 5% of infants with CLP were breastfed (partially or totally) at home. Only 7% of CL, 13% of CP and 15% of CLP bottle fed infants were given expressed breast milk.</p>	<p>III</p>
<p>Chapados C. The child born with a cleft lip and palate [French]. Can Nurse. 1997;93(1):31-36.</p> <p>Study type: Descriptive</p>	<p>Summary of abstract (article in French): 8 families of infants with CL/P were studied using interviews and questionnaires to determine how nurses and practitioners can best support families. Findings: The first three months after birth are crucial, and feeding information and support is important</p>	<p>III</p>

<p>Lee J, Nunn J, Wright C. Height and weight achievement in cleft lip and palate. Arch Dis Child. 1997;76(1):70-2.</p> <p>Study type: Descriptive cohort single centre</p>	<p>Cohort study of weight and height achievement in children with cleft CL/P (n=83). Findings: 63% were reported to have had feeding difficulties (including children up to 4 years). Isolated CP were more likely to have feeding difficulties than CL only or CLP. Nasal regurgitation and vomiting were common across groups, most problems resolved following palate repair. All children with clefts showed early growth faltering, with subgroups identified- children with syndromes, and children with severe clefts were more likely to show growth faltering. All groups showed a 'catch up' later in infancy.</p>	III
<p>Skinner J, Arvedson JC, Jones G, Spinner C, Rockwood J. Post-operative feeding strategies for infants with cleft lip. Int J Pediatr Otorhinolaryngol. 1997;42(2):169-78</p> <p>Study type: Retrospective description of feeding strategies</p>	<p>Retrospective single-centre analysis of post-operative recommendations for feeding (n=42 infants with CL), compliance with recommendations, and complications related to feeding post-surgery. Findings showed that feeding recommendations varied, with majority being told to use cup feeding for 6 weeks (N=28), only 2 caregivers were told that their baby could be nipple fed immediately post-surgery. Nearly all caregivers complied with recommendations- those that did not changed from cup to nipple feeding earlier than recommended to them. There was no relationship found between post-operative surgical complications and feeding strategies.</p>	III
<p>Trenouth MJ, Campbell AN. Questionnaire evaluation of feeding methods for cleft lip and palate neonates. Int J Paediatr Dent. 1996;6(4):241-4.</p> <p>Study type: Questionnaire descriptive</p>	<p>Evaluation of feeding methods used by mothers of infants with CL/P (n= 25), and difficulties with feeding. 12 had attempted breastfeeding, but none were able to breastfeed successfully. Most mothers experienced difficulties with feeding generally, and over ¼ had not established a feeding pattern when the baby was 2 months old.</p>	III
<p>Lang S, Lawrence CJ, L'E Orme R. Cup feeding: An alternative method of infant feeding. Arch Dis Child. 1994;71:365-369.</p> <p>Study type: Expert opinion, descriptive case study</p>	<p>Cup feeding was used in addition to breastfeeding an infant with unilateral CLP. Authors note that cup feeding may be a better 'supplementary' method of feeding infants who are partially breastfed, or who will be breastfed, so that nipple confusion caused by using bottles, does not occur.</p>	III
<p>Shprintzen RJ. The implications of the diagnosis of Robin sequence. Cleft Palate Craniofac J. 1992;29:205-209.</p> <p>Study type: Descriptive</p>	<p>Syndromic diagnosis and features of infants with Robin sequence (n=100) are described. Isolated Robin sequence was rare (17%), the most common syndrome associated with PRS was Stickler syndrome (34%). Failure to thrive in PRS is associated with upper airway obstruction, rather than presence of CP. Therefore modified feeding recommended for infants with CP do not translate to infants with PRS.</p>	III
<p>Wellman CO, Coughlin SM. Preoperative and postoperative nutritional management of the infant with cleft palate. J Pediatr Nurs.</p>	<p>Questionnaire design exploring management of infants with CP- sent to teams across the US. Responses (53%) indicated that recommended postoperative feeding strategies were mostly cup or syringe feeding. Feeding techniques were of more interest to the teams than nutritional intake.</p>	III

1991;6(3):154-158. Study type: Descriptive		
Goldberg WB, Ferguson FS, Miles RJ. Successful use of a feeding obturator for an infant with a cleft palate. Spec Care Dentist. 1988;8(2):86-89. Study type: Case study	Use of a palatal obturator for an infant with a small cleft to the soft palate. After the obturator was inserted and fitted correctly, the infant was observed to close his lips around the nipple more effectively and suck more efficiently. Increased weight gain and feeding efficiency were noted.	III
Clarren SK, Anderson B, Wolf LS. Feeding infants with cleft lip, cleft palate, or cleft lip and palate. Cleft Palate J. 1987;24(3):244-249. Study type: Description of cohort (single centre)	Observations of feeding difficulties in 143 infants with CL/P, including 23 with PRS from 1 specialist craniofacial centre in the USA. Infants with CLP (n=53) required milk delivered directly into the mouth. Supplementary nursing systems were sometimes successful. Infants with CP (n=17) whose cleft was narrow or posterior were able to breastfeed. Infants with larger or anterior clefts were not usually able to breastfeed. Infants with soft palate clefts: the authors believed that most infants in this category were able to feed normally, and only 6 were seen for difficulties during the study period. Infants with PRS (23) did not breastfeed well, due to posterior tongue placement. Infants with CL (14) were usually able to breastfeed, sometimes experienced air leakage from the cleft space, requiring occlusion with a finger or thumb. Breastfeeding was more effective than bottle-feeding as the breast moulded to the defect.	III
Balluff MA, Udin RD. Using a feeding appliance to aid the infant with a cleft palate. Ear Nose Throat J. 1986;65:50-55 Study type: Before and after comparison	Effect of obturator placement and feeding education on weight gain and time period before surgical repair in 7 infants with CL/P. Results: An average weight increase of 4.8 grams per day occurred in infants following placement of the obturator, compared to the total average weight gain of infants from birth to surgery. The trend of reduced weight gain in infants with CL/P is not significantly impacted by the placement of an obturator. Average daily weight gain following insertion of appliance was not compared to average before, as these measurements were not considered accurate. Parents indicated decreased feeding times, increased ease of feeding, improved sucking and decreased nasal regurgitation. Infants were bottle fed throughout the study.	III
Jon Jones JE, Henderson L, Avery DR. Use of a feeding obturator for infants with severe cleft lip and palate. Spec Care Dentist. 1982;2(3):116-120. Study type: Descriptive	Describes construction and use of feeding obturators for infants (N=11) with unilateral or bilateral CLP. Prior to obturator use, all parents had experienced difficulty bottle feeding using cross cut nipples. Obturators were inserted at average of 7.7 weeks of age, and used for min 8 months. Parent reports indicated less escape of liquids through the nose following, increased feeding success (using bottles). Increased comfort with feeding process and increased ease of feeding were reported.	III
Styer GW, Freeh K. Feeding infants with cleft lip and/or palate. JOGN Nursing. 1981;10(5):329-32.	Convenience sample survey (N=25) of members of a local parent group. Feeding difficulties (not limited to breastfeeding) noted by respondents included excessive length of feeds (> 45 minutes), and infants choking/gagging as they feed. Expert	III

Study type: Descriptive (survey), expert opinion	opinion (of authors) on feeding infants with cleft is given.	
Raz Rzek MKA. Prosthetic feeding aids for infants with cleft lip and palate. J Prosthet Dent. 1980;44(5):556-561. Study type: Descriptive	Infants with CP or CLP using three different types of protheses are described. Outcomes of sucking/feeding are referred to but not reported in detail. The authors conclude that for infants with complete unilateral clefts of lip, palate and alveolus, prosthesis allowed adequate suckling. For infants with incomplete unilateral clefts, (not extending to alveolar ridge), prosthesis assisted feeding and stopped the infant's tongue from interfering with the cleft site.	III
Shah CP, Wong D. Management of children with cleft lip and palate. Can Med Assoc J. 1980;122:19-24 Study type: Retrospective cohort review	Medical records reviewed for 358 infants seen at one centre. Feeding difficulties are evident with infants with CP (with or without CL) but not CL only. If the palatal cleft is wide, a palatal obturator may facilitate feeding, by preventing nasal regurgitation. Specific data for this statement are not given.	III
Grady E. Breast feeding the baby with a cleft of the soft palate: success and its benefits. Clin Pediatr (Phila). 1977;16:978-981. Study type: Expert opinion/case study	Based on her own experience of breastfeeding a baby with cleft of soft palate, the author describes success, at 4 months of total breast feeding. Recommendations include holding the nipple in the infant's mouth to make up for lack of suction. *As no suction is being applied (due to inability to form a vacuum, the mother may not be aware that the baby is feeding successfully. Weight, thriving etc should be monitored.	III
Hemingway L. Breast feeding a cleft-palate baby. Medical Journal of Australia. 1972;2(11):626. Study type: Case report	Report of an infant with CP (extent not described) who was able to breastfeed following fitting with a palatal obturator. Initially, the infant was bottle fed, and once it was discovered that the infant was able to suck effectively (though weakly), it was introduced to the breast, and exclusively breastfed.	III
Williams AC, Rothman BN, Seidman IH. Management of a feeding problem in an infant with cleft palate. J Am Dent Assoc. 1968;77:81-83. Study type: Case study	A case of an infant using a palatal obturator to facilitate feeding in an infant with CP. After unsuccessful feeding prior, an obturator was fitted, and (presumably) bottle feeding recommenced. The infant's intake increased from 33cc per feeding, requiring nasogastric feeds to thrive, to 80cc using the obturator.	III
Lifton JC. Methods of feeding infants with cleft palates. J Am Dent Assoc. 1956;53:22-31. Study type: Expert opinion with case studies	Explains different methods used for feeding infants with CP. Conclusions: Infants with CP will not be able to suck from the nipple because negative pressure cannot be created. Case studies provide examples of the successful use of obturators for feeding infants with CP, using bottles.	III
Pini JG, Peres SPDBA. Nourishment of the infant with cleft lip and palate lactant: Nursing and nourishing introduction	Summary of abstract (article written in Spanish): Feeding history of children aged 2-4years with CL/P recruited from one hospital (N=60) was reviewed to determine feeding methods during the first year of life and current height and weight.	**

<p>[Spanish]. Revista de Nutricao. 2001;14(3):195-199.</p> <p>Study type: Cohort (?)</p>		
<p>Araruna RdC, Vendruscolo DM. Nutrition of children with cleft lip and cleft palate, a bibliographic study [Portuguese]. Revista Latino-Americana de Enfermagem. 2000;8(2):99-105.</p> <p>Study type: review of literature?</p>	<p>Summary of abstract (article in Portuguese): A bibliographic review with the aim of describing children with CLP, with emphasis on feeding. Concludes that breastfeeding should be used where possible because of the benefits (e.g., long term outcomes) and possible negative outcomes of not breastfeeding (e.g., psychological).</p>	<p>**</p>
<p>Khar'kov LV, Iakovenko LN. The risk factors in assessing the overall and local status of children with congenital cleft upper lip and palate before cheiloplasty. [Russian] Stomatologiia (Mosk). 1997;76(6):47-9.</p> <p>Study type: Descriptive Cohort (?)</p>	<p>Summary of abstract (article in Russian): Nutritional status of 2000 children with CLP before lip repair; conclusion that obturators facilitate breastfeeding.</p>	<p>**</p>
<p>Campo-Paysaa A. Treatment of labio-palate clefts. Pediatric. 1987;42(9):697-703.</p> <p>Study type: Descriptive</p>	<p>Summary of abstract (article in French). Describes the need for training of tongue movement during swallowing, for infants with CL/P. Long-term follow-ups of children who have undergone such training are reported</p>	<p>**</p>

*This study was difficult to rank using the US Preventative Services Task Force standards, and may in fact sit at either II-2 or II-3.

**This study is difficult to rank as the entire article is not yet available to us in English.

Summary of evidence and areas of research deficiency:

1. Can infants with CL breastfeed successfully?

Conclusion: Abundant weak (level III) evidence that infants with CL are able to breastfeed, moderate evidence (level II-2) that infants with CL do not experience difficulty creating negative pressure necessary to breastfeed successfully, but this was measured using a bottle. Weak (level III) evidence that infants with CL may find it easier to breastfeed than to bottle-feed.

2. Can infants with CP breastfeed successfully?

Conclusion: Abundant weak (level III) evidence that infants with CP experience difficulty with breastfeeding, effected by the size and location of the cleft. There is level II-2 evidence that infants with CP are unable to create negative pressure when feeding using a bottle. Weak evidence (level III) that breastfeeding is possible. Evidence from expert opinion suggests that modification to the feeding position may improve feeding efficiency and supplemental feeding is usually necessary.

3. Can infants with CLP breastfeed successfully?

Conclusion: Weak (level III) evidence that infants with CLP experience difficulties breastfeeding and may require modification to feeding position and supplemental feeding. Stronger evidence (level II-2) that breastfeeding is sometimes possible with these infants.

4. Is there evidence that palatal obturators facilitate breastfeeding success with infants with CLP or CP?

Conclusion: In support of the use of obturators, there is weak (level III) and moderate (level II-3) evidence that obturators facilitate suction in infants with CLP or CP and may facilitate breastfeeding and/or bottle-feeding. Against the use of obturators, there is strong evidence (level I) that the use of obturators does not facilitate feeding, weight gain or parent reported ‘difficulties’ with breastfeeding, and moderate evidence (level II-2) that obturators do not facilitate the infant’s generation of negative intra-oral pressure.

5. Is there evidence to guide assessment and management of breastfeeding in infants with CL/P?

Conclusion: Weak (level III-expert opinion) evidence for management strategies, such as recommendations for positioning of infants and supplementation. Moderate (level II-3) evidence supports lactation advice as a management strategy to facilitate breastfeeding. There is currently little information to guide assessment of breastfeeding in infants with CL/P.

6. Is there evidence for additional benefits of breastfeeding for infants with cleft lip and/or palate (CL/P), as compared to the normal population?

Conclusion: Moderate to weak (level II-2 and below) evidence that breast milk carries extra benefits for infants with CLP, such as protection against otitis media, facilitating healing post-surgery, and providing less irritation to nasal and oral mucosa than artificial milk. There is moderate (level II-2) evidence that breast milk is associated with higher IQ and better school performance outcomes in the long-term. However, it should be noted that infants could be fed expressed breast milk by bottle or other means in order to gain these benefits. There is weak (level III) evidence that breastfeeding is beneficial to infants with CL/P in developing oral facial musculature and thereby promoting speech, helping the mother and baby psychologically (e.g., bonding), and pacifying infants post-surgery.

7. Is there evidence to indicate when it is safe to commence breastfeeding following surgery for lip or palate?

Conclusion: Strong (level I to level II-2) evidence that unrestricted breastfeeding immediately following CL repair is safe (in terms of wound complications) and more effective for weight gain than other feeding methods (e.g., spoon, cup or syringe). Level II-2 evidence exists that unrestricted bottle or breastfeeding immediately following surgery for lip or palate does not cause dehiscence, effect scarring, or early speech outcome.

8. Is there evidence to indicate whether infants with CP as part of a syndrome are able to breastfeed?

Conclusion: Weak (Level III) evidence that infants with PRS are unable to breastfeed and sometimes cannot feed orally at all mainly due to airway obstruction, but also due to abnormal oral configuration (glossoptosis and cleft palate).

Suggestions for Future Research

Much of the evidence for breastfeeding babies with CL, CP or CLP is weak. Little is known about techniques that might promote direct breast feeding or support direct breastfeeding with complementary feeding by bottle or other method. The benefits of breast milk consumption for babies are well described and there may be additional benefits for those with cleft conditions relating to otitis media, IQ and school performance although the latter require further investigation. There is a need for prospective, controlled studies that explore initiation, duration and outcomes of breastfeeding for these infants. It is important to learn more about how breastfeeding positions and techniques might assist the successful feeding of babies with different types of clefts and co-morbidity, success rates and benefits for both the baby (e.g., weight, health, wellbeing) and mother. Both short and long-term outcomes need to be examined.

***US Preventive Services Task Force Ranking of Evidence from Scientific Studies**

- I Evidence obtained from at least one properly randomized controlled trial.
- II-1 Evidence obtained from well-designed controlled trials without randomization.
- II-2 Evidence obtained from well-designed cohort or case-control analytic studies, preferably from more than one center or research group.
- II-3 Evidence obtained from multiple time series with or without the intervention. Dramatic results in uncontrolled experiments (such as the results of the introduction of penicillin treatment in the 1940s) could be regarded as this type of evidence.
- III Opinions of respected authorities, based on clinical experience, descriptive studies and case reports; or reports of expert committees.

The Academy of Breastfeeding Medicine, Inc.

February 2007

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Supported in part by a grant from the Maternal and Child Health Bureau, Department of Health and Human Services.