

Presurgical Orthopedics and Satisfaction in Motherhood: A Randomized Clinical Trial (Dutchcleft)

Charlotte Prah, D.D.S., Birte Prah-Andersen, D.D.S., Ph.D., Martin A. Van't Hof, Ph.D., Anne M. Kuijpers-Jagtman, D.D.S., Ph.D.

Objective: To study the effect of infant orthopedics on satisfaction in motherhood.

Design: Prospective two-arm randomized controlled trial in parallel with three participating academic cleft palate centers. Treatment allocation was concealed and was performed by means of a computerized balanced allocation method.

Setting: Cleft palate centers of Amsterdam, Nijmegen, and Rotterdam, the Netherlands.

Patients: Two groups of infants with complete unilateral cleft lip and palate and no other malformations.

Interventions: Group IO+ (n = 27) wore passive maxillary plates during the first year of life, group IO- (n = 27) did not.

Main Outcome Measure(s): Mean satisfaction scores were obtained from completed questionnaires at 6, 24, and 58 weeks of age. A 4-point scale was used (1 = very satisfactory to 4 = very unsatisfactory).

Results: The range of the mean scores for the individual items on the questionnaires for both groups ranged between 1.1 and 2.4. No differences were found between groups. Mothers appear to be satisfied in motherhood, least satisfied with the available time for themselves, and very satisfied with hugging and walking their babies. No differences were found between groups.

Conclusions: Results from the present study show that infant orthopedics, with a passive plate during the first year of life, in children with a unilateral cleft lip and palate has no influence on the mothers' satisfaction in motherhood.

KEY WORDS: *cleft palate, infant orthopedics, multicenter study, personal satisfaction, randomized controlled trial, treatment outcome*

Dr. Prah is in private orthodontic practice part time and is Staff Member of the Department of Orthodontics Academic Center for Dentistry, Amsterdam, the Netherlands, Member of the Cleft Palate Team of the Vrije Universiteit, University Medical Center, Amsterdam, the Netherlands. Dr. Prah-Andersen is Professor Emeritus of Orthodontics, Academic Center for Dentistry Amsterdam and former Head of the Department of Orthodontics, Dijkzigt University Hospital Rotterdam, the Netherlands. Dr. Van't Hof is Professor Emeritus of Biostatistics at the Department of Preventive and Curative Dentistry, Radboud University Nijmegen Medical Center, Nijmegen, the Netherlands. Dr. Kuijpers-Jagtman is Professor and Chairperson of the Department of Orthodontics and Oral Biology and Head of the Cleft Palate Craniofacial Unit of the Radboud University Nijmegen Medical Center, Nijmegen, the Netherlands.

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Address correspondence to: Dr. Charlotte Prah, Academic Center for Dentistry ACTA, Department of Orthodontics, Louwesweg 1, 1066 EA, Amsterdam, the Netherlands. E-mail c.prah@acta.nl.

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In children born with cleft lip and palate (CLP), quality of life is already influenced at birth by the impact of the conditions on the infant, mother, and family. Not only does the birth of a child with CLP raise controversial emotions in the parents, especially in the mother (Isarin, 2002), but the parents also have to adjust to the loss of the anticipated perfect child (Bradbury and Hewison, 1994). The contradictory feelings, clashing interests between the parents and others, and the conflicting ideas about what is best for the child illustrate the complexity of the relationship between parents and child (Isarin, 2002). On top of this, the infant is bound to have reconstructive surgery of the cleft lip/nose and soft palate in the first year of life, and in half of the European CLP teams, infants also receive infant orthopedics (IO) (Shaw et al., 2000). This latter type of early treatment with palatal appliances was introduced about 50 years ago in order to narrow the cleft presurgically. Since its introduction, different type of appliances with different objectives have been introduced; many of these are still in use (Winters and Hurwitz, 1995; Kuijpers-Jagtman and Prah-Andersen, 2006). Opponents claim that there is insufficient scientific data to support the use of IO and that IO is expensive and not cost effective. Claimed advantages

of IO in the literature include facilitation of lip surgery due to the narrowed cleft, more normalized tongue tip function, fewer feeding problems, restoration of the symmetry of the maxilla and nose, straightening of the nasal septum, better speech development, and minimization of the severity of skeletal and dental deformities, thus fewer orthodontics and surgeries later on and cost-effective, psychological support for parents and psychological advantages for the child due to better child-parent interaction (Kuijpers-Jagtman and Prahl-Andersen, 2006). A positive treatment experience with IO during the early phases of care could affect the mother-child relationship positively. Psychological advantages and support for the mother and child due to more frequent child-parent-team interaction as well as active parent participation in treatment due to IO have been claimed and discussed (Huddart, 1973, 1987; Gnoinski, 1990; Larson et al., 1993). The claimed psychological advantage of IO would certainly be welcome, because parents of infants with CLP are reported to have higher levels of parental stress in infancy and toddlerhood (Pope et al., 2005). In addition, Speltz et al. (1993) found that mothers of children with CLP reported less favorable social support than did mothers of children with less-visible malformations. Perceived family support was, according to Bradbury (1994), the only significant variable related to parental adjustment. On the other hand, most children with CLP emerge from the first 2 years of life with secure maternal attachment (Maris et al., 2000).

Thus far, all evidence on “psychological effects of IO” has emerged from expert opinion but has never been evaluated scientifically. Therefore, in this study, the satisfaction in motherhood in relation to the care of their child with or without IO was studied in a randomized controlled trial design.

MATERIALS AND METHODS

A detailed description of the experimental design, eligibility, treatment assignment, treatment protocol, and operators can be found in a previous publication (Prahl et al., 2001). The most important issues and specific information for this part of the study are given below.

Experimental Design

The experimental design was a prospective two-arm randomized controlled trial conducted in parallel at three centers. Three Dutch academic cleft palate centers (Nijmegen, Amsterdam, and Rotterdam) participated after the study protocol was approved by the three local ethical committees. The inclusion criteria were complete unilateral cleft lip and palate (UCLP), infants born at term, both parents Caucasian and fluent in the Dutch language, and trial entrance within 2 weeks after birth. The choice for Caucasian infants was based on two important issues: bias due to ethnic differences regarding growth, speech, and

language development, as well as the impossibility to create additional treatment groups with adequate power due to the low prevalence of infants with UCLP from other ethnic backgrounds in the Netherlands. Exclusion criteria were other congenital malformations (except for syndactyly) and soft tissue bands. Parents of eligible infants were informed verbally about the trial and were invited to participate. When the parents agreed to participate, a written informed consent was signed. Subsequently, treatment allocation was concealed and was performed for each individual center by means of a computerized balancing allocation method (Zielhuis et al., 1990). Two experimental groups were formed, one with IO and one without. Between 3 and 6 months of age, all included children were checked and confirmed by the geneticist of their own CLP team as being nonsyndromic. In total, 54 infants with UCLP were included in the trial. In previous publications from this trial (Prahl et al., 2001, 2003, 2005, 2006), however, five infants were excluded from further evaluation due to the presence of soft tissue bands, and in one case the presence of a contralateral submucosal cleft lip was diagnosed. In this part of the clinical trial, satisfaction was assumed not to be influenced by the presence of these features. As result of this assumption, completed questionnaires from all 54 infants (27 in each treatment group) were included for evaluation. For further details on the treatment protocols in both groups, the reader is referred to Prahl et al. (2001).

Data Management

In order to measure the satisfaction in motherhood, a satisfaction questionnaire was used. The questions were derived from the experiences from a longitudinal study into mother-child attachment in CLP (Hoeksma and Koomen, 1987, 1991). The questionnaire was subdivided into four domains and contained 42 questions (items).

Domain 1: Interaction and Caretaking of the Baby (12 Items). These questions concern the pleasure experienced during interaction when cuddling, consoling, playing, walking, visiting, and caretaking when feeding, bathing, dressing, and changing, including interaction when baby was crying and getting the baby out of bed.

Domain 2: Comings and Goings of the Baby (10 Items). These questions concern feelings of the mother toward the comings and goings of the baby (i.e., sleeping, feeding, and the timing of both, digestion, conduct during the day, and reaction of the baby during interaction and when left to play by himself or herself.)

Domain 3: Motherhood and Life Outside (10 Items). The questions concern the feelings of the mother toward motherhood (specifically and in terms of quality of life) and life outside of motherhood (i.e., relationship with partner, own spirit, family spirit, and housekeeping; sufficient time for other occupations or pursuits than the baby within the house or outside the house; time for self and time for friends and relatives).

TABLE 1 Domain 1 (Interaction and Caretaking of the Baby), Mean Satisfaction Scores for Both Groups IO– and IO+, Number of Participants (n), Standard Deviation (SD), 95% Confidence Interval (95% CI), and *p* Value (*p*)* at T1, T2, and T3*

Age	IO	n	Mean	SD	95% CI	<i>p</i>
T1	IO–	26	1.30	0.25	–0.27, 0.28	.930
	IO+	23	1.42	0.26		
T2	IO–	24	1.30	0.21	–0.18, 0.10	.588
	IO+	23	1.33	0.25		
T3	IO–	19	1.42	0.27	–0.23, 0.16	.387
	IO+	18	1.46	0.31		

* IO– = treatment group without infant orthopedics (IO); IO+ = treatment group with IO; T1 = 6 weeks of age; T2 = 24 weeks of age; T3 = 58 weeks of age.

Domain 4: Support (10 Items). The questions concern the support from partner, relatives, acquaintances and friends, and others for mental support, caretaking, housekeeping; support, information, and advice from the cleft team, general physician, and other medical institutions; and adequate contact with other parents and parents of children with clefts.

For the response, a 4-point scale was used: 1 = very satisfactory, very happy, a lot of fun, more than adequate; 2 = satisfactory, happy, often fun, adequate; 3 = unsatisfactory, unhappy, not so much fun, inadequate; and 4 = very unsatisfactory, very unhappy, no fun, very inadequate.

It was decided to evaluate satisfaction at the beginning of IO treatment, halfway through IO treatment, and after IO treatment had ended. Questionnaires (including written instructions on the first page) were handed out by the orthodontists of the cleft teams during routine checkups planned at the ages of 6 (T1), 24 (T2), and 58 (T3) weeks. The mothers were asked to complete and return the questionnaire at around the previously mentioned ages.

Statistical Analysis

The data from the returned questionnaires were evaluated and were analyzed statistically. A factor analysis was performed within the domains. The reliability of the questionnaire was determined with the Cronbach alpha as well as for each domain separately. Differences between the mean values of the two treatment groups, IO+ and IO–, of each domain as well as for the questionnaire, were tested with *t* tests at T1, T2, and T3. In order to check for center effects and for interactions between center and IO, a two-way analysis of variance was performed for the questionnaire at T1, T2, and T3 with IO and center as the dependent variables. Finally, the results from both treatment groups were pooled in order to identify the factors containing the items with the highest and lowest mean scores.

RESULTS

Questionnaires were available from 49 mothers. The number of the included questionnaires fluctuated due to

TABLE 2 Domain 2 (Comings and Goings of the Baby), Mean Satisfaction Scores for Both Groups IO– and IO+, Number of Participants (n), Standard Deviation (SD), 95% Confidence Interval (95% CI), and *p* Value (*p*)* at T1, T2, and T3*

Age	IO	n	Mean	SD	95% CI	<i>p</i>
T1	IO–	26	1.45	0.31	–0.38, 0.07	.142
	IO+	22	1.61	0.45		
T2	IO–	24	1.29	0.23	–0.20, 0.10	.678
	IO+	23	1.33	0.27		
T3	IO–	19	1.27	0.22	–0.29, 0.04	.521
	IO+	18	1.40	0.28		

* IO– = treatment group without infant orthopedics (IO); IO+ = treatment group with IO; T1 = 6 weeks of age; T2 = 24 weeks of age; T3 = 58 weeks of age.

questionnaires not being returned or not being completed around the target ages. The age range for the completed questionnaires was T1: 4 to 9 weeks, T2: 22 to 28 weeks, and T3: 55 to 66 weeks. The reliability (Cronbach alpha) of the four domains ranged between .66 and .85, whereas the reliability of the total questionnaire (all four domains together) was .88. The reliability of the domains could not be improved by leaving out any items. The results for all four domains and for the questionnaire as a whole are presented in Tables 1 through 5. No significant differences (*t* tests) were found between the groups IO+ and IO– at T1, T2, and T3 (Tables 1 through 5). Mothers from both groups appeared to be satisfied to the same degree, and the mean level of satisfaction fell between very satisfactory and satisfactory in both groups.

Factor analysis within the domains resulted in 10 factors (Table 6) and showed high correlation between the items within the resulting 10 factors. The contents of the items within each factor were explicable and logical. All items were included within the 10 factors and were distributed evenly among the factors. Because the reliability of the domains could not be improved by leaving out one or more of the items, no attempt was made to further analyze the factors.

The pooled results from both treatment groups IO+ and IO– showed highest mean satisfaction scores for the item from factor 2 (domain 1): “do you like to hug your baby” (mean score = 1.06, SD = 0.32), from factor 3 (domain 1): “do you like to take your baby for a walk” (mean score = 1.11, SD = 0.32), and for the item from factor 5 (domain 2): “do you feel satisfied with the baby’s reaction when hugging” (mean score = 1.14, SD = 0.37). The lowest mean satisfaction scores were found for the item from factor 7 (domain 3): “do you feel you have sufficient time for yourself” (mean score = 2.26, SD = 0.71) and for the item from factor 9 (domain 4): “do you feel you have sufficient contact with other parents of children with clefts” (mean score = 2.24, SD = 0.81).

DISCUSSION

It is the challenge and aim of cleft palate teams to render optimal care and to guard or to improve the quality of life

TABLE 3 Domain 3 (Motherhood and Life Outside), Mean Satisfaction Scores for Both Groups IO– and IO+, Number of Participants (n), Standard Deviation (SD), 95% Confidence Interval (95% CI), and *p* Value (*p*)* at T1, T2, and T3*

Age	IO	n	Mean	SD	95% CI	<i>p</i>
T1	IO–	26	1.79	0.37	–0.30, 0.16	.507
	IO+	22	1.85	0.42		
T2	IO–	24	1.68	0.38	–0.26, 0.23	.512
	IO+	23	1.69	0.44		
T3	IO–	19	1.56	0.34	–0.41, 0.08	.491
	IO+	18	1.72	0.40		

* IO– = treatment group without infant orthopedics (IO); IO+ = treatment group with IO; T1 = 6 weeks of age; T2 = 24 weeks of age; T3 = 58 weeks of age.

of their patients. Because resources and professional help are limited, treatment has to be cost effective or in severe, rare cases efficacious and worth the possible (worst) side effects. The proper attitude of the professionals toward care, good quality management of the CLP team, and its organization is the only way to establish, assure, and improve the level of care delivered by the team. Assessment of patient satisfaction and quality of life is an essential part of treatment outcome and quality management and therefore should be included in the outcome measures.

There is information available about the psychological well-being of CLP patients and their parents (Strauss and Broder, 1991; Broder, 2001; Hunt et al., 2005; Pope et al., 2005), but the interaction with treatment is an area that warrants further exploration. It was concluded in the report of recent World Health Organization (WHO) meetings on international collaborative research on craniofacial anomalies (WHO, 2002) that more and better clinical research is needed. Of equal importance, consensus has to be reached about outcome measures, especially for psychological variables and quality of life. This part of the clinical trial (Dutchcleft) assessing the effects of IO contributes to the knowledge of early psychosocial factors regarding the interaction of treatment and satisfaction in motherhood. Although one would like to measure the quality of life of the patient, it is not measurable early in life. Alternatively, the satisfaction in motherhood was measured, because the bigger personal investment in care in case of IO and the increased psychological support by the team may introduce differences in the appreciation of different aspects of motherhood regarding the experience with IO and the perceived support. It was decided to evaluate satisfaction in motherhood, because the mother is usually the primary caretaker in Dutch society, at least during the first year of life. The questionnaire in this study was administered during the treatment period with IO and several weeks after its termination (6 to 58 weeks). The reliability was good and the factor analysis, which included all items, was logical and explicable. Differences in the appreciation of motherhood between mothers with or without the experience with IO in time and between centers ranged between .03 and .16 and were never statistically significant. The power of the trial was sufficient to demonstrate significant

TABLE 4 Domain 4 (Support), Mean Satisfaction Scores for Both Groups IO– and IO+, Number of Participants (n), Standard Deviation (SD), 95% Confidence Interval (95% CI), and *p* Value (*p*)* at T1, T2, and T3*

Age	IO	n	Mean	SD	95% CI	<i>p</i>
T1	IO–	26	1.48	0.28	–0.31, 0.04	.653
	IO+	22	1.61	0.31		
T2	IO–	24	1.56	0.34	–0.21, 0.19	.893
	IO+	23	1.57	0.33		
T3	IO–	19	1.48	0.25	–0.26, 0.14	.117
	IO+	18	1.54	0.35		

* IO– = treatment group without infant orthopedics (IO); IO+ = treatment group with IO; T1 = 6 weeks of age; T2 = 24 weeks of age; T3 = 58 weeks of age.

differences when larger than .2 to .25. If differences larger than .2 were found between groups, these would have been significant. This was not the case. Therefore, it is not plausible that differences in satisfaction in motherhood will develop in the long run as a result of IO within a CLP population. For this reason, no future measurements are intended during the follow-up of this clinical trial.

Recently, but unfortunately after the onset of this clinical trial, new evidence regarding the negative effect of parental stress was reported by Pope et al. (2005). They reported higher levels of parental stress in infancy and toddlerhood for a CLP population. When consistent over time, this may lead to higher levels of adjustment problems when children reach toddlerhood. It is, however, unclear to what extent satisfaction in motherhood is associated with this emotional state, and thus it remains unclear if IO has any (lasting) influence on parental stress, for better or for worse. It has, however, been shown from the present study that there was no difference between groups in the perceived support from partners, family, or professionals, which according to Bradbury (1994) is an important variable related to parental adjustment.

All in all, the mothers responded positively to the questions indicating satisfaction in motherhood during the first year of life. Mothers were most satisfied with hugging and walking their babies and were less satisfied with the time for other occupations or pursuits, the perceived support, and the information and advice obtained from the cleft team, the general physician, and other medical institutions. This is in accordance with the

TABLE 5 Mean Satisfaction Scores of the Questionnaire as a Whole (Four Domains) for Both Groups IO– and IO+, Number of Participants (n), Standard Deviation (SD), 95% Confidence Interval (95% CI), and *p* Value (*p*)* at T1, T2, and T3*

Age	IO	n	Mean	SD	95% CI	<i>p</i>
T1	IO–	26	1.50	0.22	–0.25, 0.05	.199
	IO+	23	1.60	0.29		
T2	IO–	24	1.45	0.19	–0.15, 0.10	.299
	IO+	23	1.48	0.24		
T3	IO–	19	1.43	0.20	–0.24, 0.05	.352
	IO+	18	1.53	0.24		

* IO– = treatment group without infant orthopedics (IO); IO+ = treatment group with IO; T1 = 6 weeks of age; T2 = 24 weeks of age; T3 = 58 weeks of age.

TABLE 6 Results of Within-Domain Factor Analysis

Domain 1
 Factor 1: Satisfaction with taking care of the baby (4 items)
 Factor 2: Fulfillment experienced when interacting with the baby like hugging and consoling (4 items)
 Factor 3: Fulfillment experienced when interacting with the baby like playing, walking and visiting (4 items)

Domain 2
 Factor 4: Satisfaction with functioning of the baby like sleeping and eating (6 items)
 Factor 5: Mother's judgment of the baby's reaction towards comforting, hugging, and playing (4 items)

Domain 3
 Factor 6: Satisfaction in motherhood in general, relationship, family spirit, and overall satisfaction since the birth of baby (6 items)
 Factor 7: Satisfaction in life outside of motherhood, sufficient time for other occupations or pursuits (4 items)

Domain 4
 Factor 8: Satisfaction with support of partner (3 items)
 Factor 9: Satisfaction with support of professionals and other parents (4 items)
 Factor 10: Satisfaction with support from friends and family (3 items)

findings of Speltz et al. (1993), who found that mothers of children with CLP reported less favorable social support than mothers of children with less visible anomalies. Last, but not least, mothers experienced inadequate contact with other parents, especially parents with children with clefts. Professionals and parent support associations should use these findings to improve the quality of care.

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REFERENCES

- Bradbury ET, Hewison J. Early parental adjustment to visible congenital disfigurement. *Child Care Health Dev.* 1994;20:251–266.
- Broder H. Using psychological assessment and therapeutic strategies to enhance well-being. *Cleft Palate Craniofac J.* 2001;38:248–254.
- Gnoinski WM. Infant orthopedics and later orthodontic monitoring for unilateral cleft lip and palate patients in Zurich. In: Bardach J, Morris HL, eds. *Multidisciplinary Management of Cleft Lip and Palate.* Philadelphia: WB Saunders; 1990:578–585.
- Hoeksma JB, Koomen HMY. *Development of Early Mother-Child Interaction and Attachment.* Amsterdam, The Netherlands: Vrije Universiteit, Pro Lingua, Dissertation. 1991.
- Hoeksma JB, Koomen HMY. Responsiviteit en hechting: Een enquête bij ouders van kinderen met en kinderen zonder een schisis. *Nederlands Tijdschrift voor de Psychologie.* 1987;42:282–290.
- Huddart AG. The effect of form and dimension on the management of the maxillary arch in unilateral cleft lip and palate conditions. *Scand J Plast Reconstr Surg.* 1987;21:53–56.
- Huddart AG. An evaluation of presurgical treatment. *Br J Orthod.* 1973;1:21–25.
- Hunt O, Burden D, Hepper P, Johnston C. The psychological effects of cleft lip and palate: a systematic review. *Eur J Orthod.* 2005;27:274–285.
- Isarin J. *De eigen ander.* Amsterdam: Vrije Universiteit; 2002. Thesis.
- Kuijpers-Jagtman AM, PrahL-Andersen B. History of neonatal maxillary orthopedics: past to present. In: Berkowitz S., ed. *Cleft Lip and Palate: Diagnosis and Management.* 2nd ed. Berlin, Germany: Springer-Verlag; 2006:395–404.
- Larson M, Sällström KO, Larson O, McWilliam J, Ideberg M. Morphologic effect of preoperative maxillofacial orthopedics (T-traction) on the maxilla in unilateral cleft lip and palate patients. *Cleft Palate Craniofac J.* 1993;30:29–34.
- Maris CL, Endriga MC, Speltz ML, Jones K, DeKleyen M. Are infants with orofacial clefts at risk for insecure mother-child attachments? *Cleft Palate Craniofac J.* 2000;37:257–265.
- Pope AW, Tillman K, Snyder HT. Parenting stress in infancy and psychological adjustment in toddlerhood: a longitudinal study of children with craniofacial anomalies. *Cleft Palate Craniofac J.* 2005;42:556–559.
- PrahL C, Kuijpers-Jagtman AM, Van't Hof MA, PrahL-Andersen B. Infant orthopedics and facial appearance: a randomized clinical trial (Dutchcleft). *Cleft Palate Craniofac J.* 2006;43:659–664.
- PrahL C, Kuijpers-Jagtman AM, Van't Hof MA, PrahL-Andersen B. Infant orthopedics in UCLP: effect on feeding, weight, and length: a randomized clinical trial (Dutchcleft). *Cleft Palate Craniofac J.* 2005;42:171–177.
- PrahL C, Kuijpers-Jagtman AM, Van't Hof MA, PrahL-Andersen B. A randomized prospective clinical trial into the effects of infant orthopedics on maxillary arch dimensions in unilateral cleft lip and palate (Dutchcleft). *Eur J Oral Sci.* 2001;109:297–305.
- PrahL C, Kuijpers-Jagtman AM, Van't Hof MA, PrahL-Andersen B. A randomized prospective clinical trial of the effect of infant orthopedics in unilateral cleft lip and palate: prevention of collapse of the alveolar segments (Dutchcleft). *Cleft Palate Craniofac J.* 2003;40:337–342.
- Shaw WC, Semb G, Nelson P, Brattström V, Mølsted K, PrahL-Andersen B. *The Eurocleft Project 1996–2000.* Amsterdam, The Netherlands: IOS Press; 2000.
- Speltz ML, Morton K, Goodell EW, Clarren SK. Psychological functioning of children with craniofacial anomalies and their mothers: follow-up from late infancy to school entry. *Cleft Palate J.* 1993;30:482–489.
- Strauss RP, Broder H. Directions and issues in psychological research and methods as applied to cleft lip and palate and craniofacial anomalies. *Cleft Palate Craniofac J.* 1991;28:150–156.
- Winters JC, Hurwitz DJ. Presurgical orthopedics in the surgical management of unilateral cleft lip and palate. *Plast Reconstr Surg.* 1995;95:755–764.
- World Health Organization (WHO). *Global Strategies to Reduce the Health Care Burden of Craniofacial Anomalies.* Geneva, Switzerland: WHO; 2002.
- Zielhuis GA, Straatman H, Van't Hof-Grootenboer AE, van Lier HJ, Rach GH, van den Broek P. The choice of a balanced allocation method for a clinical trial in otitis media with effusion. *Stat Med.* 1990;9:237–246.