





The Burden of Care in Nasoalveolar Molding Treatment in Cleft Patients

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Abstract

Objectives This study, first in Hungary, examined the success of presurgical nasoalveolar molding (NAM) therapy in cleft patients from a caregiver's perspective and revealed factors that can cause inconvenience.

Patients and Methods A survey-based study was performed using a 32-item questionnaire following NAM therapy. The survey was sent to families whose child underwent NAM therapy from 2010 until 2020 at the 1st Department of Paediatrics, Semmelweis University. The questions focused on four main parts: socioeconomic, origin of the cleft, difficulties of therapy, and self-assessment. Fifty-three families received the questionnaire, 17 of them completed it.

Results The mean age was 5 ± 3.7 weeks when NAM therapy started. Fifty-eight percent of the patients were male and 42% female. Patients are living more than 60 km from the cleft center (59%). Patients had to make the journey between their residence and the cleft center ~ 10 to 15 times. In most cases, NAM therapy was covered by health insurance (83%). The unilateral cleft and lip palate occurred 58%, while the bilateral were 42%. Thirty-five percent of the patients had an allergic reaction against the adhesive, and 35% were affected by wounds on their lips or noses. The way of feeding was variable. Seventeen percent of the parents were able to breastfeed. In all cases, parents were satisfied with the NAM therapy.

Conclusions The present study highlighted the value of caregivers' role in NAM therapy. The burden of care is acceptable, caregivers have high compliance, and are determined to help the effectiveness of therapy. Limitations of this study include a single-institute data with a small number of cases.

Keywords

- ▶ NAM
- ▶ cleft lip and palate
- ▶ quality of life
- ▶ treatment evaluation

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Introduction

Cleft lip and palate (CLP) are the most common facial congenital malformations that affect the soft and skeletal structures. For effective therapy, a multidisciplinary team that principally consists of a plastic surgeon, orthodontist, ENT specialist, and speech therapist is indispensable. To facilitate successful treatment, the cleft team should be aware of the economic and social situation of the parents that can impact the physician–patient relationship. Moreover, the following external factors can affect the efficiency of the therapy: travel distance, financial position of the caregiver, and level of parental engagement.¹

According to the surgical protocol currently followed at our institute, two surgical interventions—cheiloplasty and palatoplasty—are performed in the first year of a newborn's life. The cheiloplasty is preceded by nasoalveolar molding (NAM), a presurgical infant–orthopedic technique accurately described by Grayson et al.² The NAM treatment can enhance the surgical success of children with cleft lip by narrowing the cleft, molding the nasal cartilage, and passive control of the alveolar segments. Facial improvements due to NAM therapy and cheiloplasty can be observed in ►Figs. 1 and 2. Nevertheless, patients may face several problems, like difficulty in feeding, irritations of the skin, and the inability of the caregivers to precisely position and stabilize the NAM appliance. These effects can adversely impact the desired outcome of the therapy. Nevertheless, patients may face several problems, like difficulty in feeding, irritations of the skin, and the inability of the caregivers to precisely position and stabilize the NAM appliance. These effects can adversely impact the desired outcome of the therapy.

A better understanding of the complex social, geographical, and economic environment of the caregivers is essential in helping families with NAM.³ Understanding these difficulties may support the development of alternative treatment options in presurgical orthopedic techniques of cleft patients. The NAM device is made up of two parts. The oral palate plate can shape the alveoli, while the nasal support can make the nasal cartilages more symmetrical.^{4,5} Adhesive tape is used to approach the upper lip segments closer together and reduce the size of the fissure. This process starts during the first weeks of life and takes 3 to 4 months, for which the active participation of the caregivers is indispensable.⁶ Duties of the caregivers include changing the aforementioned adhesive tape and the repeated cleaning and repositioning the palatal plate.

Daily use of the NAM device and regular medical check-ups can represent a considerable burden on the patient's caregiver life. Moreover, the influence of this apparent burden of care can also affect the efficiency of the NAM treatment.⁷

Our study aims to examine the success and failure of the presurgical orthopedic technique not from a clinical point of view but from a caregiver's perspective and to try to determine factors that can reduce the effectiveness of the NAM

therapy and cause inconvenience both for the caregivers and the child.^{8,9} A further aim of the survey was to determine how caregivers were informed about the processes whether they would have needed more advice. This has been the first survey in Hungary since the NAM therapy was introduced in our country.

Patients and Methods

To identify those factors that can affect the regular use of oral and nasal palate, a survey-based study was performed using a 32-item questionnaire following NAM therapy. The therapy was performed at the 1st Department of Paediatrics at the Semmelweis University Budapest. The questions design was partly based on previous surveys by Dean et al.¹⁰

The survey was divided into four main parts. First, a socioeconomic part focused on financial, educational, and social factors that could affect the caregivers' chances of a successful NAM therapy. The second part dealt with the origin of the disease and any coexisting genetic disorders or malformations, like monogenic or chromosomal syndromes. The third part examined the possible hardships of NAM including difficulty breathing, feeding, wounds, and allergy. Finally, a fourth part of the survey was a self-assessment of the caregiver's overall satisfaction with the outcome of NAM.

The questionnaire included mostly multiple-choice or one-answer questions; however, some parts of the survey allowed caregivers to share their own experiences in a free format, using their own words. The survey was sent to families whose child underwent NAM therapy from 2010 until 2020 at Semmelweis University. Seventeen patients completed the survey. No objective evaluation of clinical success (physical, functional, or aesthetic) was included in the survey. Data were collected and presented as the mean \pm standard error of the mean. Statistical comparisons of data were performed with Fisher's exact probability test or a one-way analysis of variance with post hoc Tukey's multiple comparison test, as appropriate. A *p*-value of less than 0.05 was considered to be statistically significant. R Studio 3.4.2 was used for statistical analysis and diagnosis.

Results

During the study, 673 patients were treated with CLP from 2010 to 2020. We identified 53 patients of them who initiated NAM therapy. All families received the questionnaire, and 17 of them completed it.

Socioeconomic Factors

Most of the patients lived in a two-parent family with one sibling. In 47% of the families, the first child was affected by CLP. The mean age was 5 ± 3.7 weeks (range: 2–14 weeks) when the NAM therapy started. Fifty-eight percent patients were male, while 42% were female. Patients lived an average of 60 km from the cleft center (59%), with more than 60 minutes of traveling time (53%). In contrast, the duration of the hospital visit was less than 30 minutes (59%). It was observed that patients had to make the journey between their residence and the cleft center

Table 1 Demographic characteristic of participants

Variables	Summary of statistic (mean \pm SD)
Total participants (<i>n</i>)	17
Age (weeks)	4 \pm 8.7
Gender (<i>n</i>)	
Female	42% (<i>n</i> = 7)
Male	58% (<i>n</i> = 10)
Type of the dentofacial deficiency (<i>n</i>)	
Unilateral cleft and lip palate	58% (<i>n</i> = 10)
Bilateral cleft and lip palate	42% (<i>n</i> = 7)
Patient with cleft (<i>n</i>)	
First born	47% (<i>n</i> = 8)
Second born	29% (<i>n</i> = 5)
Third born	24% (<i>n</i> = 4)
Distance between the cleft center and residence (<i>n</i>)	
More than 60 km	59% (<i>n</i> = 10)
Less than 60 km	41% (<i>n</i> = 7)
Traveling time to the cleft center (min)	
More than 60 minutes	53% (<i>n</i> = 9)
Less than 60 minutes	47% (<i>n</i> = 8)
Number of the traveling (<i>n</i>)	
1–5 times	18% (<i>n</i> = 3)
6–10 times	54% (<i>n</i> = 9)
11–15 times	18% (<i>n</i> = 3)
15 times <	12% (<i>n</i> = 2)
Duration of the visit (min)	
Less than 30 minutes	59% (<i>n</i> = 10)
30–60 minutes	41% (<i>n</i> = 7)
Associated health problem (<i>n</i>)	29% (<i>n</i> = 5)
NAM treatment covered by health insurance (<i>n</i>)	83% (<i>n</i> = 14)
Receive paid or sick leave (<i>n</i>)	59% (<i>n</i> = 10)
Successfulness of the NAM therapy (<i>n</i>)	88% (<i>n</i> = 15)
Allergic reaction against the adhesive (<i>n</i>)	35% (<i>n</i> = 6)
Wounds on the lip or nose following the therapy (<i>n</i>)	35% (<i>n</i> = 6)
The way of feeding (<i>n</i>)	
Feeding bottle	54% (<i>n</i> = 9)
Haberman feeder	23% (<i>n</i> = 4)
Other special feeder	23% (<i>n</i> = 4)
Breastfeeding (<i>n</i>)	17% (<i>n</i> = 3)
Difficulty feeding (<i>n</i>)	23% (<i>n</i> = 4)
Difficulty breathing (<i>n</i>)	23% (<i>n</i> = 4)
Knowledge of the NAM therapy by the specialists (<i>n</i>)	82% (<i>n</i> = 14)
Usage of other source for advisements, like social media (<i>n</i>)	82% (<i>n</i> = 14)
Recommendation of surgery	100% (<i>n</i> = 17)

Abbreviations: NAM, nasoalveolar molding; SD, standard deviation.



Fig. 1 Patient with bilateral cleft lip and palate before nasolabial molding (NAM) treatment, after NAM treatment, and after cheiloplasty.

~10 to 15 times (54%). In most cases, the NAM therapy was covered under health insurance (83%). More than half of the patients (59%) could receive paid or sick leaves.

Cleft Types and Associated Malformations

The unilateral cleft and lip palate was present in 58%, while the bilateral was 42%. It was observed that the alveolar defect affected mainly the first born (47%). However, there were no correlations between the distribution of the dentofacial deficiency and its occurrence among the siblings ($p=0.3737$). Twenty-nine percent of the patients suffered from associated health problems like atrial septal defect, renal developmental abnormality, or corpus callosum agenesis.

Difficulties of the Treatment

The outcome of the therapy was largely compatible (83%). Nevertheless, 35% of the patients suffered from an allergic reaction against the adhesive. Furthermore, 35% of patients were affected by wounds on their lips or noses following the therapy. The way of feeding was variable. The feeding bottle was most commonly used (54%), but some patients also chose to use a Haberman feeder (23%) or other feeders (23%). Only 17% of the parents were able to breastfeed. According to the questionnaire, 23% of them experienced difficulty feeding or breathing. There was no correlation between the feeding or breathing difficulties on the feeding ($p=0.758$). Unfortunately, more than half of the caregivers (53%) did not receive any counseling or instructions on feeding from the cleft team.

Self-Assessment

Patients could receive information about the process from the treating specialists and also via social media (82%). In all

cases, parents were satisfied with the therapy, and they would recommend the NAM therapy to other caregivers of CLP patients. Excellent reliability was determined to assess the dependability of the survey results using the intraclass correlation coefficient (0.974). The results of the survey are summarized in **Table 1**.

Discussion

Patients with CLP undergo multiple surgical interventions to attain the appropriate form of the nose and the lip. Several presurgical treatments have emerged with the aim to diminish the severity of the labial and nasal deformity.^{11,12} However, the NAM technique seems to have proven to be one of the adjunct therapies.¹³

Besides the clinical achievements of NAM, there have been several studies addressing the sociographic, economic, and satisfaction aspects of the therapy from the caregivers' perspectives.^{7,10} It is mandatory to counsel caregivers about the NAM process and provide them with information regarding the chances of success or failure of the therapy and the possible complications.

Sischo et al presented how caregivers can cope and adapt to early cleft care using NAM. They found that caregivers often worry about the success of NAM (e.g., stress related to lip taping, appliance causing sores in their child's mouth, travel to weekly appointments).⁹ Thus, it is essential to establish and effectively communicate evidence-based guidelines to reduce barriers to care and optimize the chances of completing NAM treatment.¹⁰

Instead of presurgical NAM, early cleft lip repair (ECLR) provides another option in a protocol that decreases the burden of health costs.¹⁴ In our study, however, the NAM



Fig. 2 Patient with unilateral cleft lip and palate before nasolabial molding (NAM) treatment, after NAM treatment, and after cheiloplasty.

therapy was covered by state insurance. Travel costs of public transport were also covered by state insurance. Compared with ECLR, the NAM therapy became more economical in our patients by reducing expensive secondary surgical interventions in the nasal region afterwards.¹⁵

Compliance issues were of greater concern, with an estimated incidence of 30% for missed appointments and 26% for removal of the NAM appliance by the tongue movements.⁷ In our survey, we did not experience severe problems with the compliance of the caregivers regarding check-ups or application of the NAM plate; however, in one case, the baby was not able to wear the plate, and only the lip tape could be applied.

We were able to affirm the experience of a former project of Raina et al that there is a positive correlation between the quality of caregivers' social support system and their coping and psychosocial functioning during their infant's medical treatment.¹⁶ Our results are in substantial agreement with the findings of Sischo et al in which the caregivers could hardly cope with their leading role in the preventive NAM therapy without any social support or appropriate help. After all, the psychosocial well-being of caregivers is crucial for effective treatment and clinical outcomes.¹⁷

The limitations of our study were that although caregivers were well informed of the treatment process and possible difficulties of NAM therapy, they did not receive nutrition counseling. Others include a single institute data with a small number of cases. A multi-institutional study is needed to clarify the relationship between the caregivers' attitude and efficient NAM therapy.

We aim to introduce in the near future a fully digital workflow of NAM therapy in Hungary, and prefabricated palates can be easily used by caregivers in their home environment resulting in reduced travelling time and a calm atmosphere.

Conclusions

In conclusion, our work was based on the quality-of-life questionnaire to measure the effectiveness of the NAM therapy and the quality of caregivers' life. Present study highlights the value of caregivers' role in NAM therapy. The results may be summarized by pointing out the difficulties that caregivers face during NAM. Furthermore, our findings also suggest that the burden of care in NAM-treated patients is relatively high, but the caregivers are determined to help the effectiveness of therapy. Moreover, the aesthetic and functional outcomes of NAM are also of significant importance. Due to the limitations of this study, more research is needed to find a solution to minimize the number and the duration of regular medical check-ups.

Ethical Committee Information

Ethical approval for this study was obtained from our university ethics committee.

Conflict of Interest

There were no conflicts of interest. The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

References

- Alfonso AR, Ramly EP, Kantar RS, et al. What is the burden of care of nasolabial molding? *Cleft Palate Craniofac J* 2020;57(09):1078–1092
- Grayson BH, Santiago PE, Brecht LE, Cutting CB. Presurgical nasolabial molding in infants with cleft lip and palate. *Cleft Palate Craniofac J* 1999;36(06):486–498
- Abbott MM, Kokorowski PJ, Meara JG. Timeliness of surgical care in children with special health care needs: delayed palate repair for publicly insured and minority children with cleft palate. *J Pediatr Surg* 2011;46(07):1319–1324
- Barillas I, Dec W, Warren SM, Cutting CB, Grayson BH. Nasolabial molding improves long-term nasal symmetry in complete unilateral cleft lip-cleft palate patients. *Plast Reconstr Surg* 2009;123(03):1002–1006
- Grayson BH, Garfinkle JS. Early cleft management: the case for nasolabial molding. *Am J Orthod Dentofacial Orthop* 2014;145(02):134–142
- Hopkins EE, Gazza E, Marazita ML. Parental experience caring for cleft lip and palate infants with nasolabial moulding. *J Adv Nurs* 2016;72(10):2413–2422
- Levy-Bercowski D, Abreu A, DeLeon E, et al. Complications and solutions in presurgical nasolabial molding therapy. *Cleft Palate Craniofac J* 2009;46(05):521–528
- Sischo L, Chan JW, Stein M, Smith C, van Aalst J, Broder HL. Nasolabial molding: prevalence of cleft centers offering NAM and who seeks it. *Cleft Palate Craniofac J* 2012;49(03):270–275
- Sischo L, Broder HL, Phillips C. Coping with cleft: a conceptual framework of caregiver responses to nasolabial molding. *Cleft Palate Craniofac J* 2015;52(06):640–650
- Dean RA, Wainwright DJ, Doringo IL, Teichgraeber JF, Greives MR. Assessing burden of care in the patient with cleft lip and palate: factors influencing completion and noncompletion of nasolabial molding. *Cleft Palate Craniofac J* 2019;56(06):759–765
- McNEIL CK. Congenital cleft palate; a case of congenital cleft palate which required the fitting of a special appliance. *Br Dent J* 1948;84(07):137–141
- Latham RA, Kusy RP, Georgiade NG. An extraorally activated expansion appliance for cleft palate infants. *Cleft Palate J* 1976;13:253–261
- Shetye PK, Grayson BH. Nasolabial molding treatment protocol in patients with cleft lip and palate. *Semin Orthod* 2017;23(03):261–267
- Włodarczyk JR, Wolfswinkel EM, Fahradyan A, et al. Nasolabial molding: assessing the burden of care. *J Craniofac Surg* 2021;32(02):574–577
- Patel PA, Rubin MS, Clouston S, et al. Comparative study of early secondary nasal revisions and costs in patients with clefts treated with and without nasolabial molding. *J Craniofac Surg* 2015;26(04):1229–1233
- Raina P, O'Donnell M, Schweltnus H, et al. Caregiving process and caregiver burden: conceptual models to guide research and practice. *BMC Pediatr* 2004;4:1
- Sischo L, Clouston SA, Phillips C, Broder HL. Caregiver responses to early cleft palate care: a mixed method approach. *Health Psychol* 2016;35(05):474–482