

A Questionnaire-Based Survey Study to Assess the Burden of Care in Nasoalveolar Molding Treatment in Cleft Patients

Puja Priyadarshini

Senior Resident, Department of Plastic Surgery, Nalanda Medical College and Hospital, Patna, Bihar, India

Received: 01-12-2022 / Revised: 27-12-2022 / Accepted: 28-01-2023

Corresponding author: Dr. Puja Priyadarshini

Conflict of interest: Nil

Abstract

Aim: The study examined the success of presurgical nasoalveolar molding (NAM) therapy in cleft patients from a caregiver's perspective and revealed factors that can cause inconvenience.

Methods: A survey-based study was performed using a 32-item questionnaire following NAM therapy study was conducted at Nalanda Medical College and Hospital, Patna, Bihar, India. 600 patients were treated with CLP. We identified 50 patients of them who initiated NAM therapy. All families received the questionnaire, and 20 of them completed it.

Results: The mean age was 5 ± 3.7 weeks (range: 2–14 weeks) when the NAM therapy started. 60 percent patients were male, while 40% were female. Patients lived an average of 60 km from the cleft center (60%), with more than 60 minutes of traveling time (60%). The unilateral cleft and lip palate was present in 60%, while the bilateral was 40%. It was observed that the alveolar defect affected mainly the first born (45%). However, there were no correlations between the distribution of the dentofacial deficiency and its occurrence among the siblings ($p = 0.3737$).

Conclusion: 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.

Keywords: NAM, Cleft Lip And Palate, Quality of Life, Treatment Evaluation.

This is an Open Access article that uses a funding model which does not charge readers or their institutions for access and distributed under the terms of the Creative Commons Attribution License (<http://creativecommons.org/licenses/by/4.0>) and the Budapest Open Access Initiative (<http://www.budapestopenaccessinitiative.org/read>), which permit unrestricted use, distribution, and reproduction in any medium, provided original work is properly credited.

Introduction

Cleft lip and/or cleft palate affects approximately 1 in every 600 births worldwide with variation across geographic areas and ethnic groups. [1,2] The American Cleft Palate-Craniofacial Association has set parameters of care including the necessity of an interdisciplinary team, contemporary practices, and guidelines for long-term treatment and evaluation. Surgical repair of the cleft lip usually occurs within 12 months of life and cleft palate repair

within 18 months of life. [3] In appreciation for importance of optimal functional and aesthetic outcomes at the time of primary cleft lip and nose repair, nasoalveolar molding (NAM) was developed to reduce the severity of the cleft lip/nasal deformity and align the alveolar segments. [4]

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. [5] 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. [6]

A better understanding of the complex social, geographical, and economic environment of the caregivers is essential in helping families with NAM. [7] 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. [8,9] 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. [10] Duties of the caregivers includes 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. [11]

The study examined the success of presurgical nasoalveolar molding (NAM) therapy in cleft patients from a caregiver's

perspective and revealed factors that can cause inconvenience.

Materials and Methods

A survey-based study was performed using a 32-item questionnaire following NAM therapy study was conducted at Nalanda Medical College and Hospital, Patna, Bihar, India, 600 patients were treated with CLP. We identified 50 patients of them who initiated NAM therapy. All families received the questionnaire, and 20 of them completed it. The questions design was partly based on previous surveys by Dean et al. [12]

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. 20 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

Table 1: Demographic characteristic of participants

Variables	Mean \pm SD
Total	20
Age (weeks)	4 \pm 8.7
Gender	N
Female	8 (40)
Male	12 (60)
Type of the dentofacial deficiency	
Unilateral cleft and lip palate	12 (60)
Bilateral cleft and lip palate	8 (40)
Patient with cleft	
First born	9 (45)
Second born	6 (30)
Third born	5 (25)
Distance between the cleft center and residence	
More than 60 km	12 (60)
Less than 60 km	8 (40)
Duration of the visit (min)	
Less than 30 minutes	12 (60)
30–60 minutes	8 (40)

The mean age was 5 ± 3.7 weeks (range: 2–14 weeks) when the NAM therapy started. 60 percent patients were male, while 40% were female. Patients lived an average of 60 km from the cleft center (60%), with more than 60 minutes of traveling time (60%). The unilateral cleft

and lip palate was present in 60%, while the bilateral was 40%. It was observed that the alveolar defect affected mainly the first born (45%). However, there were no correlations between the distribution of the dentofacial deficiency and its occurrence among the siblings ($p = 0.3737$).

Table 2: Other features of patient

Variables	N
Associated health problem	6 (30)
NAM treatment covered by health insurance	17 (85)
Receive paid or sick leave	12 (60)
Successfulness of the NAM therapy	18 (90)
Allergic reaction against the adhesive	7 (35)
Wounds on the lip or nose following the therapy	7 (35)
The way of feeding	
Feeding bottle	11 (55)
Haberman feeder	5 (25)
Other special feeder	4 (20)
Breastfeeding	3 (15)
Difficulty feeding	4 (20)
Difficulty breathing	4 (20)
Knowledge of the NAM therapy by the specialists	16 (80)
Usage of other source for advisements, like social media	16 (80)
Recommendation of surgery	20 (100)

In most cases, the NAM therapy was covered under health insurance (85%). More than half of the patients (60%) could receive paid or sick leaves. 30% of the patients suffered from associated health problems like atrial septal defect, renal developmental abnormality, or corpus callosum agenesis. 35% of the patients suffered from an allergic reaction against the adhesive. The way of feeding was variable. The feeding bottle was most commonly used (55%), but some patients also chose to use a Haberman feeder (25%) or other feeders (20%). Only 15% of the parents were able to breastfeed. According to the questionnaire, 20% of them experienced difficulty feeding or breathing. There was no correlation between the feeding or breathing difficulties on the feeding ($p = 0.758$). Patients could receive information about the process from the treating specialists and also via social media (80%). 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).

Discussion

As the patient experiences the primary surgeries early in life, it is important that the treating team perform the best treatment in the first attempt; reduce the magnitude of the secondary deformities and finally the number of secondary procedures. Pre surgical nasoalveolar molding (NAM) [13] is one of the methods proposed for reducing the pre surgical cleft severity [14-16] and the future necessity for secondary surgeries. [17,18]

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. [11] 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). [18] 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. [19]

Instead of presurgical NAM, early cleft lip repair (ECLR) provides another option in a protocol that decreases the burden of health costs. [20] In our study, however, the NAM 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. [21] 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. [11] 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. [22] 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. [23]

Conclusion

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.

References

1. World Health Organization. Global strategies to reduce the healthcare burden of craniofacial anomalies: report of WHO meetings on international collaborative meetings on craniofacial anomalies. 2001.
2. Petersen PE. The World Oral Health Report 2003: continuous improvement of oral health in the 21st century—the approach of the WHO Global Oral Health Programme. Community Dentistry and oral epidemiology. 2003 Dec; 31:3-24.
3. American Cleft Palate-Craniofacial Association. Parameters of care. 2009.
4. Shetye PK, Grayson BH. Nasoalveolar molding treatment protocol in patients with cleft lip and palate. Semin Orthodont. 2017;23(3):261-267.
5. Alfonso A R, Ramly E P, Kantar R S. What is the burden of care of nasoalveolar molding? Cleft Palate Craniofac J. 2020;57(09):1078–1092.
6. Grayson B H, Santiago P E, Brecht L E, Cutting C B. Presurgical nasoalveolar molding in infants with cleft lip and palate. Cleft Palate Craniofac J. 1999;36(06):486–498.
7. Abbott M M, Kokorowski P J, Meara J G. 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.
8. Barillas I, Dec W, Warren S M, Cutting C B, Grayson B H. Nasoalveolar molding improves long-term nasal symmetry in complete unilateral cleft lip-cleft palate patients. Plast Reconstr Surg. 2009;123(03):1002–1006.
9. Grayson B H, Garfinkle J S. Early cleft management: the case for nasoalveolar molding. Am J Orthod Dentofacial Orthop. 2014;145(02):134–142.
10. Hopkins E E, Gazza E, Marazita M L. Parental experience caring for cleft lip and palate infants with nasoalveolar moulding. J Adv Nurs. 2016;72(10):2413–2422.
11. Levy-Bercowski D, Abreu A, DeLeon E. Complications and solutions in presurgical nasoalveolar molding therapy. Cleft Palate Craniofac J. 2009; 46(05):521–528.
12. Dean R A, Wainwright D J, Doringo I L, Teichgraeber J F, Greives M R. Assessing burden of care in the patient with cleft lip and palate: factors influencing completion and noncompletion of nasoalveolar molding. Cleft Palate Craniofac J. 2019;56(06):759–765.
13. Grayson BH, Cutting CB. Presurgical nasoalveolar orthopedic molding in primary correction of the nose, lip, and alveolus of infants born with unilateral and bilateral clefts. The Cleft palate-craniofacial journal. 2001 May;38(3):193-8.
14. Liou EJ, Subramanian M, Chen PK. Progressive changes of columella length and nasal growth after nasoalveolar molding in bilateral cleft

- patients: a 3-year follow-up study. Plastic and reconstructive surgery. 2007 Feb 1;119(2):642-8.
15. Lee CT, Garfinkle JS, Warren SM, Brecht LE, Grayson BH. Nasoalveolar molding improves appearance of children with bilateral cleft lip–cleft palate. Plastic and reconstructive surgery. 2008 Oct 1;122(4):1131-7.
 16. Barillas I, Dec W, Warren SM, Grayson BH. Nasoalveolar molding improves long-term nasal symmetry in complete unilateral cleft lip–cleft palate patients. Plastic and reconstructive surgery. 2009 Mar 1;123(3):1002-6.
 17. Pfeifer TM, Grayson BH, Cutting CB. Nasoalveolar molding and gingivoperiosteoplasty versus alveolar bone graft: an outcome analysis of costs in the treatment of unilateral cleft alveolus. The Cleft palate-craniofacial journal. 2002 Jan;39(1):26-9.
 18. Sischo L, Chan JW, Stein M, Smith C, Van Aalst J, Broder HL. Nasoalveolar molding: prevalence of cleft centers offering NAM and who seeks it. The Cleft palate-craniofacial journal. 2012 May;49(3):270-5.
 19. Dean R A, Wainwright D J, Doringo I L, Teichgraeber J F, Greives M R. Assessing burden of care in the patient with cleft lip and palate: factors influencing completion and noncompletion of nasoalveolar molding. Cleft Palate Craniofac J. 2019;56(06):759–765.
 20. Wlodarczyk J R, Wolfswinkel E M, Fahradyan A. Nasoalveolar molding: assessing the burden of care. J Craniofac Surg. 2021;32(02):574–577.
 21. Patel P A, Rubin M S, Clouston S. Comparative study of early secondary nasal revisions and costs in patients with clefts treated with and without nasoalveolar molding. J Craniofac Surg. 2015;26(04):1229–1233.
 22. Raina P, O'Donnell M, Schwellnus H, Rosenbaum P, King G, Brehaut J, Russell D, Swinton M, King S, Wong M, Walter SD. Caregiving process and caregiver burden: conceptual models to guide research and practice. BMC pediatrics. 2004 Dec;4(1):1-3.
 23. Aguilar R. Fatigue symptom and oximetry sign in a patient with a positive Covid-19 antigen test for Sars-Cov-2. Journal of Medical Research and Health Sciences. 2022; 5(8): 2165–2176.