|| ISSN(online): 2589-8698 || ISSN(print): 2589-868X ||

Available online on http://www.ijmbs.info

International Journal of Medical and Biomedical Studies; 2024; 8(5); 63-73

A Case Report

Surgical Assisted Rapid Maxillary Expansion in Hemifacial Syndrome Patient Post Primary Cleft Treatment: A Case Report

Dhia Thifal[1*], Melita Sylvyana[2], Abel Tasman[3], Saptiadi Oktora[3]

[1]Oral and Maxillofacial Surgery Resident, Faculty of Dentistry, Padjadjaran University, Bandung, Indonesia

[2]Oral and Maxillofacial Surgery Staff Department, RSUP Dr. Hasan Sadikin, Bandung, Indonesia

[3]Oral and Maxillofacial Surgery Staff Department, Faculty of Dentistry, Padjadjaran University, Bandung, Indonesia

Received: 11-09-2024 / Revised 14-10-2024 / Accepted 19-10-2024

Corresponding author: Dhia Thifal (dhia14002@mail.unpad.ac.id)

DOI: **https://doi.org/10.32553/ijmbs.v8i5.2878**

Conflict of interest: Nil

Abstract:

Introduction: Cleft lip and palate is a congenital malformation resulting from an incomplete or incorrect fusion during embryonic development. Hemifacial syndrome is one of rare orofacial anomalies with midface hypoplasia. Maxillomandibular deformity repair for maxillary hypoplasia can be achieved by repositioning the maxilla and mandible to correct occlusion, mastication, functional issues, cosmetic concerns, and facial symmetry. The main concerns in the case of maxillary hypoplasia are the repair of transverse (narrowing of the maxilla), sagittal (anterior posterior), and vertical (facial proportion) aspects. This study aimed to elucidate the treatment for patients with maxillary hypoplasia by utilizing the surgical assessment of rapid maxillary expansion (SARME) following cleft lip and palate surgery.

Case Report: A 21-year-old female patient had symptoms of a truncated maxilla, compromised speech, and challenges with mastication and ingestion. The patient's surgical history includes having undergone labioplasty, palatoplasty in 2002, and gnatoplasty in 2023. The recommended treatment for this situation involves SARME. The patient's speaking and feeding difficulties were caused by both transversal and vertical problems in the patient with maxillary hypoplasia. This is why the therapy was necessary.

Case Management: Implementation of surgical assessment rapid maxillary expansion (SARME).

Conclusions: This case report concludes a notable improvement in the upper and lower jaw's functional, masticatory, and aesthetic aspects in a patient with maxillary hypoplasia. This improvement was achieved with surgical assisted of rapid maxillary expansion (SARME) following primary cleft lip and palate surgery.

Keywords: *Cleft lip and palate, Hemifacial syndrome, SARME*

Introduction

Cleft lip and / or palate is the most prevalent craniofacial deformity resulting from inadequate fusion of the facial prominences. The incidence of Cleft lip and palate differs according on geographic origin, sex, and ethnicity[1]. In Europe, a

child with a cleft lip and palate is born about once every 700 babies1. Cleft lip and palate (CLP) has a complex aetiology. The specific processes underlying the contributions of genetic and environmental risk factors to etiopathogenesis remain unclear[2–4]. Furthermore, congenital abnormalities and syndromes are more common in patients with solitary cleft palates and more severe cleft lip and palate phenotypes[2]. In addition to severe malocclusions affecting the primary and permanent dentition, they frequently appear with developmental and congenital dental defects in tooth quantity (agenesis, supernumerary teeth), shape, and location4–6. As a result, cleft lip and palate patients need a sophisticated also early interdisciplinary treatment.[7]

After cleft lip and palate, hemifacial microsomia is the second most common type of isolated facial birth defect. A congenital skeletal abnormality known as hemifacial microsomia causes a deficiency on one side of the face, which is facial asymmetry and maxillomandibular hypoplasia are prominent traits. The underdevelopment of the ear, mandible, maxilla, zygoma, temporal bone, and related muscle and soft tissues is mainly a syndrome of the first brachial arch. Although the precise cause of hemifacial microsomia is unknown, research in the lab indicates that the clinical characteristics may result from an early loss of neural crest cell- related first brachial arch formation. Bimaxillary surgery has historically been used to treat modest skeletal deformities in individuals with hemifacial microsomia. Autogenous costochondral grafting is utilized to treat more severe defects.[8]

Maxillary hypoplasia are frequent in all age groups, with children with mixed dentition having an unusually high frequency of 18%9. Reduced posterior overjet and unilateral or bilateral posterior crossbite are common symptoms, and in later developmental stages, these may also result in anteroposterior discrepancy and facial asymmetry[10]. For the correction of maxillary hypoplasia, rapid maxillary expansion has been widely utilized. Rapid maxillary expansion may enhance nasal morphology and augment nasal cavity and maxillary sinus volume, according to recent studies[11–13]. Furthermore, it has been shown that maxillary transverse expansion may enhance oxygen saturation and have a favourable impact on respiratory performance[14].

For patients who are skeletally mature, a viable approach to accomplish effective maxillary expansion is surgical assessment of rapid maxillary expansion[15]. For adult patients, a combination of orthodontic and surgical procedures is frequently required to safely, effectively, and predictably repair hypoplasia maxilla. Due to increasing bone resistance, elements like the zygomatic buttress's structure and the ossification of the midpalatal suture inhibit extension. Surgically-assisted rapid maxillary expansion, or SARME, is the combination of orthodontics and surgery used to address maxillary atresia.[16]

This study aimed to elucidate the treatment for patients with maxillary hypoplasia by utilizing the surgical assessment of rapid maxillary expansion (SARME) following cleft lip and palate surgery.

Case

A 21-year-old female patient came to Oral and Maxillofacial Department of Hasan Sadikin Hospital with chief complaint having difficulty while eating or drinking, and a speech compromised (disartria). The patient was born from a 26-year-old mother with a history of P1A0 pregnancy. The patient was born with a caesarean delivery assisted by an obstetrician at a private hospital in the Cirebon area at 37 weeks' gestation and a birth weight of 2 kg. The patient’s mother has no history of falls, drug consumption, and herbal medicine consumption during pregnancy. Family history of cleft lip/palate, systemic disease, drug and food allergies

Text

were denied. The patient has completed immunization history. The patient had undergone labioplasty and palatoplasty at the regional clinic in Cirebon (2002).

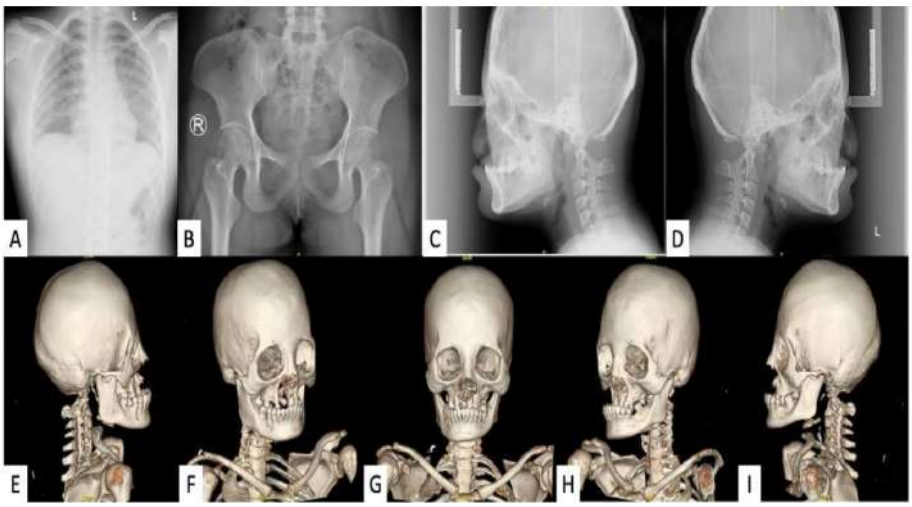

Figure 1: Supporting examination, (A) Chest X-ray, (B) Pelvic X-ray, (C-D) Cephalometry, (E-I) Head CT-Scan

From clinical examination, the patient was diagnosed with malocclusion class III Angle classification. From the head ct-scan, there was defects in the anterior midline of the left dentoalveolar bone to the nasal crest, hypoplasia of the maxillary bone, and defects in part of the hard palate which is form of palatognatoschizis. Also, there was hypertrophy of the right nasal concha accompanied by deviation of the nasal septum to the right, and bilateral maxillary and right ethmoidal sinusitis. From the chest x-ray, the patient was suspected with right bronchopneumonia, but no visible cardiomegaly. From digital pelvic x-ray, pelvic was within normal limits.

Figure 2: Orthopantomogram x-ray

From Orthopantomogram x-ray, there was a defect of midline maxilla which is form of gnatoschizis. The patient was diagnosed with gnatoschizis unilateral dextra post labioplasty
post palatoplasty and maxillary hypoplasia. We have obtained the patient's consent to this article.

Case Management

According to the clinical examination, head ct-scan, orthopantomogram x-ray, chest x-ray, and pelvic x-ray, the patient was decided to get gnatoplasty and grafting Illiaca to treat the gnatoschizis unilateral dextra, then anterior osteodistraction to treat the maxillary hypoplasia a year later.

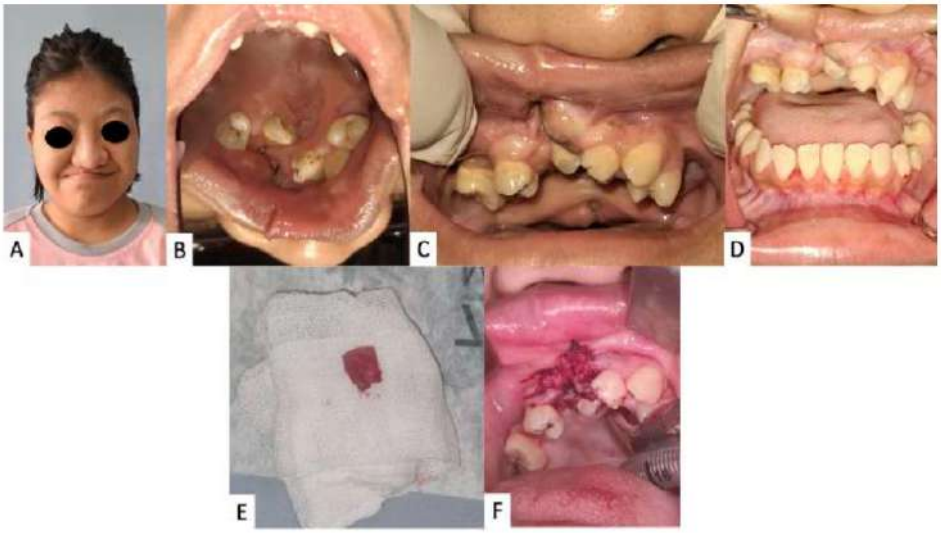

Figure 3: (A-D) Pre-operative, (E) Grafting Illiaca, (F) Gnatoplasty

Figure 4: One year Post Gnatoplasty, then patient was planned to performed osteodistraction

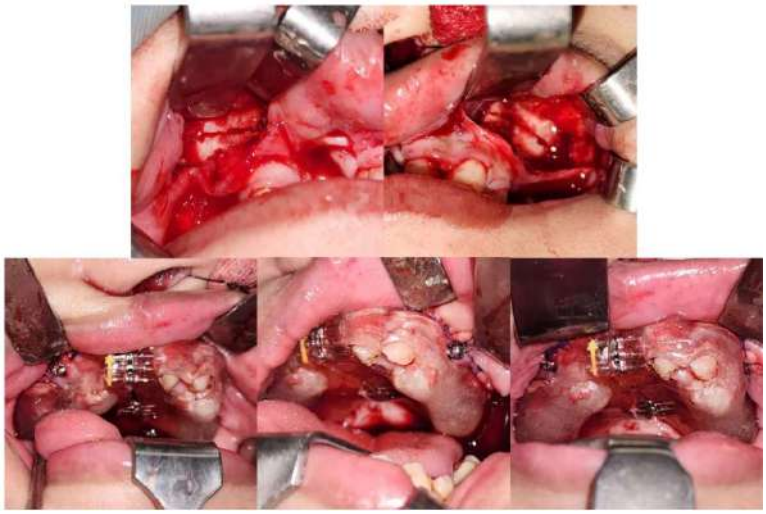

Figure 5: Anterior osteodistraction

We performed application of Surgically Assisted Rapid Maxillary Expansion (SARME) to treat this patient. The post-operative condition of this patient was within normal limits, there was no active bleeding, and has minimal swelling. But, the patient complained of a nasal congestion. The patient was given oral hygiene instruction, and planned for control at the outpatient clinic.

The patient was having a daily control for expansion screw activation at the outpatient clinic until post-operative day V. Then, at post-operative day VI the patient had IMF rubber installed from Temporary Anchorage Device (TAD) maxillary to mandibular bracket to increase the vertical dimension of the maxillary alveolar. After that the patient had expansion screw activation until post-operative day XXII. On post-operative day XXIII, the patient then consulted to Orthodontic Department to install mandibular bracket. And last, at post-operative day L, the patient came back to control and have no complaint.

Figure 6: POD I

Figure 7: POD III, Post-operative control and expansion screw activation

Figure 8: POD IV, expansion screw activation

Figure 9: POD V, expansion screw activation

Figure 10: POD VI, expansion screw activation and mandibular Temporary Anchorage Device (TAD) installed

Figure 11: POD XI, expansion screw activation

Figure 12: POD XII, expansion screw activation

Figure 13: POD XIV, expansion screw activation

Figure 14: POD XXII, expansion screw activation

Figure 15: POD XXIII, control and consulted to orthodontic department

Figure 16: POD L, patient have no complaint

In our case, we found an improvement in the upper and lower jaw's functional, masticatory, and aesthetic aspects.

Discussion

On our case, at first we performed gnatoplasty and and grafting illiaca. Scar tissue formation, palate muscle strain, and constriction of the maxillary complex are the results of the surgical procedures necessary for cleft lip and palate repair[17]. Additionally, canine impaction is frequently the result of maxillary constriction[18], which may also be caused by intrinsic factors affecting palatal growth19. Furthermore, Class III malocclusion and crossbite are caused by the lower tongue position stimulating mandibular growth[5]. Transverse skeletal disparity and alveolar integrity must be managed with maxillary expansion and secondary alveolar bone grafting (SABG). The palate volume is increased, normal growth and development is permitted, and adequate room for tongue position is provided by the maxillary expansion[20]. It improves the attractiveness of the face, promotes tooth eruption, allows orthodontic tooth movement, and stabilises and maintains the continuity of the maxillary complex[21]. For this technique, bone that is both autologous and allogenic can be utilized[22].

A secure and effective alternative for treating maxillary hypoplasia is maxillary expansion[23]. Orthopaedic expansion in children and teenagers has a positive prognosis since the midpalatal suture is either open or has very little interdigitation. Nevertheless, in adulthood, the skeletal consequences of rapid maxillary expansion may be limited by resistance resulting from midpalatal suture fusion[24,25] and the

structures around the maxilla, namely the zygomatic buttress and pterygopalatine structures.[16] Surgically-Assisted Rapid Maxillary Expansion (SARME) is a therapy option for these patients since it removes resistance in certain maxillary regions.[26]

In our case, expander activation started three days after surgery, however, some studies recommend immediate activation.[27,28] On the first activation protocol, we used it as a adaptation period, as gingival recession or dehiscence in the area required modification of the first activation protocol due to alterations in the gingiva between the maxillary central incisors. There was no radiographic control to support the theory that this was caused by an asymmetrical midpalatal suture (MPS) fracture in the space between the incisors[16].

Our patient also complained for having a nasal congestion after the osteodistraction anterior surgery. Previous study confirmed that surgically-assisted rapid maxillary expansion could improve patient's nasal breathing immediately after SARME.[16] While the reported morbidity of surgically assisted rapid maxillaryl expansion surgery is low, several problems have been documented. Previous publication have detailed the potential drawbacks of maxillary extension, such as severe bleeding and ensuing gingival recession. There have also been reports of root resorption. There have also been reports of devitalization of the teeth, altered pulpal blood flow, infections, and injuries to the maxillary nerve branches. However, peripheral blood flow is unaffected provided the procedure is carried out according to protocol.

There have also been reports of other problems, including unilateral enlargement, sinus infection, alar base flare, recurrence, and periodontal collapse.[15]

However, beside the risks and complications that potentially occur after the implementation of SARME, our patient has a notable improvement in the upper and lower jaw's functional, masticatory, and aesthetic aspects in a patient with maxillary hypoplasia. After fifty days post-operative, our patient didn't have any complaint.

Conclusions

This case report concludes a notable improvement in the upper and lower jaw's functional, masticatory, and aesthetic aspects in a patient with maxillary hypoplasia. This improvement was achieved with surgical assisted of rapid maxillary expansion (SARME) following primary cleft lip and palate surgery.

References

1. Mossey PA, Modell B. Epidemiology of oral clefts 2012: An international perspective. Cleft Lip Palate. 2012;16:1–18.
2. Jamilian A, Sarkarat F, Jafari M, Neshandar M, Amini E, Khosravi S, et al. Family history and risk factors for cleft lip and palate patients and their associated anomalies. Stomatologija. 2017;19(3):78–83.
3. Maili L, Letra A, Silva R, Buchanan EP, Mulliken JB, Greives MR, et al. PBX-WNT- P63-IRF6 pathway in nonsyndromic cleft lip and palate. Birth Defects Res [Internet]. 2019/12/11. 2020 Feb 1;112(3):234–44. Available from: https://pubmed.ncbi.nlm.nih.gov/31825181
4. Bartzela TN. Treatment approaches to syndromes affecting craniofacial and dental structures. J World Fed Orthod [Internet]. 2019;8(4):131–7. Available from: http://dx.doi.org/10.1016/j.ejwf.2019.10.002
5. Bartzela TN, Carels CEL, Bronkhorst EM, Jagtman AMK. Tooth agenesis patterns in unilateral cleft lip and palate in humans. Arch Oral Biol [Internet]. 2013;58(6):596–602. Available from:

http://dx.doi.org/10.1016/j.archoralbio.2012.12.007
6. Bartzela TN, Carels CEL, Bronkhorst EM, Rønning E, Rizell S, Kuijpers-Jagtman AM. Tooth agenesis patterns in bilateral cleft lip and palate. Eur J Oral Sci [Internet]. 2010;118(1):47–52. Available from: http://dx.doi.org/10.1111/j.1600-0722.2009.00698.x
7. Khdairi N, Halilah T, Khandakji M, Bartzela T. Rapid Maxillary Expansion Treatment in Patients with Cleft Lip and Palate: A Survey on Clinical Experience in the European Cleft Centers. J Clin Med. 2023 May 1;12(9).
8. Choi SH, Kang DY, Hwang CJ. Adult patient with hemifacial microsomia treated with combined orthodontics and distraction osteogenesis. American Journal of Orthodontics and Dentofacial Orthopedics. 2014 Jan;145(1):72–84.
9. Carlson C, Sung J, McComb RW, Machado AW, Moon W. Microimplant-assisted rapid palatal expansion appliance to orthopedically correct transverse maxillary deficiency in an adult. American Journal of Orthodontics and Dentofacial Orthopedics [Internet]. 2016;149(5):716–28. Available from: http://dx.doi.org/10.1016/j.ajodo.2015.04.043
10. Erdur EA, Yıldırım M, Karatas RMC, Akin M. Effects of symmetric and asymmetric rapid maxillary expansion treatments on pharyngeal airway and sinus volume. Angle Orthod [Internet]. 2020 May 1;90(3):425–31. Available from: https://pubmed.ncbi.nlm.nih.gov/33378426
11. Kavand G, Lagravère M, Kula K, Stewart K, Ghoneima A. Retrospective CBCT analysis of airway volume changes after bone-borne vs tooth-borne rapid maxillary expansion. Angle Orthod [Internet]. 2019/02/15. 2019 Jul;89(4):566–74. Available from:https://pubmed.ncbi.nlm.nih.gov/30768911
12. Truong CT, Jeon HH, Sripinun P, Tierney A, Boucher NS. Short-term and long-term effects of rapid maxillary expansion on the nasal soft and hard tissue. Angle Orthod [Internet]. 2021 Jan 1;91(1):46–53. Available from: https://pubmed.ncbi.nlm.nih.gov/33289784
13. Silva Filho OG da, Lara TS, Ayub PV, Ohashi ASC, Bertoz FA. Photographic assessment of nasal morphology following rapid maxillary expansion in children. J Appl Oral Sci [Internet]. 2011 Oct;19(5):535–43. Available from:https://pubmed.ncbi.nlm.nih.gov/21986660
14. Fastuca R, Perinetti G, Zecca PA, Nucera R, Caprioglio A. Airway compartments volume and oxygen saturation changes after rapid maxillary expansion: a longitudinal correlation study. Angle Orthod [Internet]. 2015 Nov;85(6):955–61. Available from: https://pubmed.ncbi.nlm.nih.gov/26516709
15. Melo K dos A, Costa ST, Stehling RSS, Urbano ES. Risks and complications in surgically assisted rapid maxillary expanison. Rev Gaucha Odontol. 2013;
16. Loriato L, Ferreira CE. Surgically-assisted rapid maxillary expansion (SARME): Indications, planning and treatment of severe maxillary deficiency in an adult patient. Dental Press J Orthod. 2020 May 1;25(3):73–84.
17. Townend PI. Technique of Rapid Expansion in Patients with Cleft Lip and Palate. Br J Orthod [Internet]. 1980;7(2):65–7. Available from: http://dx.doi.org/10.1179/bjo.7.2.65
18. Šmahel Z, Velemínská J, Trefný P, Müllerová Ž. Three-Dimensional Morphology of the Palate in Patients with Bilateral Complete Cleft Lip and Palate at the Stage of Permanent Dentition. The Cleft Palate-Craniofacial Journal [Internet].

2009;46(4):399–408. Available from: http://dx.doi.org/10.1597/07-199.1
19. Bizzarro M, Generali C, Maietta S, Martorelli M, Ferrillo M, Flores-Mir C, et al. Association between 3D palatal morphology and upper arch dimensions in buccally displaced maxillary canines early in mixed dentition. Eur J Orthod [Internet]. 2018;40(6):592–6. Available from: http://dx.doi.org/10.1093/ejo/cjy023
20. Generali C, Primozic J, Richmond S, Bizzarro M, Flores-Mir C, Ovsenik M, et al. Three- dimensional evaluation of the maxillary arch and palate in unilateral cleft lip and palate subjects using digital dental casts. Eur J Orthod [Internet]. 2017;39(6):641–5. Available from: http://dx.doi.org/10.1093/ejo/cjx019
21. Zhang W, Shen G, Wang X, Yu H, Fan L. Evaluation of alveolar bone grafting using limited cone beam computed tomography. Oral Surg Oral Med Oral Pathol Oral Radiol [Internet]. 2012;113(4):542–8. Available from: http://dx.doi.org/10.1016/j.oooo.2011.10.001
22. Scalzone A, Flores-Mir C, Carozza D, d'Apuzzo F, Grassia V, Perillo L. Secondary alveolar bone grafting using autologous versus alloplastic material in the treatment of cleft lip and palate patients: systematic review and meta-analysis. Prog Orthod [Internet]. 2019 Feb 11;20(1):6. Available from: https://pubmed.ncbi.nlm.nih.gov/30740615
23. McNamaraa JA. Maxillary transverse deficiency. American Journal of Orthodontics and Dentofacial Orthopedics [Internet]. 2000;117(5):567–70. Available from: http://dx.doi.org/10.1016/s0889-5406(00)70202-2
24. Persson M, Thilander B. Palatal suture closure in man from 15 to 35 years of age. Am J Orthod [Internet]. 1977;72(1):42–52. Available from: http://dx.doi.org/10.1016/0002-9416(77)90123-3
25. Wehrbein H. The mid-palatal suture in young adults. A radiological-histological investigation. The European Journal of Orthodontics [Internet]. 2001;23(2):105–14. Available from: http://dx.doi.org/10.1093/ejo/23.2.105
26. Vilani GNL, Mattos CT, de Oliveira Ruellas AC, Maia LC. Long-term dental and skeletal changes in patients submitted to surgically assisted rapid maxillary expansion: A meta-analysis. Oral Surg Oral Med Oral Pathol Oral Radiol [Internet]. 2012;114(6):689–97. Available from: http://dx.doi.org/10.1016/j.oooo.2012.01.040
27. Kurt G, Altug-Ataç AT, Ataç MS, Karasu HA. Stability of surgically assisted rapid maxillary expansion and orthopedic maxillary expansion after 3 years' follow-up. Angle Orthod [Internet]. 2010 Jul;80(4):425–31. Available from: https://pubmed.ncbi.nlm.nih.gov/20482344
28. Magnusson A, Bjerklin K, Nilsson P, Marcusson A. Surgically assisted rapid maxillary expansion: long-term stability. The European Journal of Orthodontics. 2009 Apr 1;31(2):142–9.