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Use of Customized Marpe in an Infant with Single Central Incisor Syndrome: A Case Study

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Abstract

Background: The solitary median maxillary central incisor is a rare anomaly associated with numerous craniofacial and oral developmental disorders, such as a single central incisor located in the midline of the maxilla and abnormalities such as facial dysmorphia and congenital stenosis.

Objective: To present a clinical case report of an infant patient with solitary median maxillary central incisor who underwent surgery to correct pyriform aperture stenosis and subsequent rapid maxillary expansion using a microimplant-assisted rapid palatal expander.

Results: The early diagnosis and interventions performed on the patient allowed an increase in nasal airflow with a positive response in the rehabilitation of respiratory and swallowing functionality.

Conclusion: The microimplant-assisted rapid palatal expander device use in an infant proved to be a safe and effective procedure.

Keywords: Solitary Median Maxillary Central Incisor Syndrome; Syndrome; Palatal Expansion Technique; Orthodontic Anchorage Procedures

Introduction

The solitary median maxillary central incisor (SMMCI) is a rare anomaly associated with numerous craniofacial and oral developmental abnormalities, such as the presence of a solitary incisor located in the midline of the maxilla, both in the primary and permanent dentition, with a higher prevalence in the maxillary arch; it can rarely occur in the mandible [1-6].

Anomalies such as facial dysmorphia and congenital stenosis are often associated with SMMCI, whose etiology can be explained by the absence or reduction of lateral growth of the maxillary midline around days 37 and 38 of embryonic development. Congenital pyriform stenosis is a rare cause of nasal obstruction that can occur in newborns. It is caused by the overgrowth of the medial nasal process of the maxilla, causing a narrowing of the anterior third of the nasal cavity. There is often an association with other malformations, most notably a triangular-shaped palate and a single central megaincisor, which can be found in up to 75% of these patients [7-10].

The diagnosis of SMMCI can be made in the prenatal or neonatal period. Prenatal diagnosis can be made at 18-22 weeks of gestational age by ultrasonography, which analyzes features of the head, face, nose, and palate [6]. The pediatrician's initial clinical examination of the newborn may reveal severe respiratory distress with nasal obstruction and may alert to the possible association with the presence of a single central incisor, as supported by the scientific literature [11,12]. Nasal endoscopy and/or computed tomography (CT) exams are extremely useful for diagnosing nasal obstruction, with CT being the chosen exam to detect SICSU soon after birth [11,12]. Cases of SMMCI without severe respiratory compromise may be diagnosed later, during the transition from primary to permanent dentition, around six years of age. Late diagnosis can affect the patient's quality of life, resulting in learning disabilities, poor respiratory efficiency, obstructive sleep apnea, and dental changes consistent with a hypoplastic (atrophic) palate [13].

The treatment is multiprofessional and multidisciplinary, involving several areas of health care. In the presence of respiratory failure and the child's inability to feed properly, surgery to expand the pyriform aperture through sublabial access is the procedure of choice [9]. In the presence of transverse maxillary deficiency, rapid maxillary expansion (RME) procedures can provide gains in upper airway dimensions [14,15]. This maxillary expansion can be successfully performed using expander appliances [14,15]. However, there is no established treatment protocol for neonates who do not have dental support for such appliances. This paper aims to present a clinical case report of an infant patient with SMMCI who underwent surgery for pyriform aperture stenosis and subsequent RME using a MARPE-c appliance.

Diagnosis and Etiology

A boy, born on April, 2020, by cesarean section, did not allow the probe passage in the nasal cavities during the first care, with Down Syndrome. He developed early respiratory distress, worsening his breathing pattern in the first 12 hours of life, requiring intubation and mechanical ventilation. The severe respiratory distress and nasal obstruction complicated several extubation attempts. During this period, the patient did not accept oral feeding, presented a narrowing of the nasal cavities, and was referred to the ORL (Otorhinolaryngology) Department of University for evaluation.

The ORL team requested a CT of the paranasal sinuses on May, 2020 (1 month 19 days), to diagnose airway obstruction, and it was possible to visualize the presence of a single central incisor (Figure 1). On June, 2020 (2 months 4 days), the patient was evaluated with a bronchoscopy, which showed no changes. At that time, an attempt was made to pass the 3 mm nasofibroscope through the nostrils. There was no progression and significant respiratory discomfort during the test collection (effort), leading to the hypothesis of pyriform aperture stenosis and the indication for surgical intervention. On July, 2020 (3 months 1 day), the patient underwent pyriform aperture repair, without intercurrence, with mechanical ventilation and occlusive dressing. On postoperative day 1 (PO1), on July, 2020 (3 months 2 days), a new CT was performed on the paranasal sinuses, and the patient was discharged on July, 2020 (3 months 4 days) (Figure 1).

After the surgical phase, the expected improvement in the patient's breathing did not occur, given the presence of snoring episodes suggestive of obstructive sleep apnea which required evaluation by the orthodontic team, who proposed RME to promote an increase in the air volume of the complex and possible improvement of the patient's respiratory function, with a MARPE-c appliance, individually manufactured for the patient in this study.

The Digital Imaging and Communications in Medicine (DICOM) (CT scan) and Standard Triangle Language (STL) (intraoral scan and microimplant) files were superimposed.

The planning of the appropriate mini-implant location was defined by CT, considering bone thickness, soft tissue thickness, and the distance of the mini-implants to the adjacent oral tissues without injuring any precious structures or tooth germs. The paramedian region in zone T was chosen, 3 mm from the midpalatal suture, where it was observed that there was sufficient osseous volume in all three planes of the CT [16] (Figure 2).

The virtually selected mini-implants were suitable for the MARPE technique, model HS MARPE by PecLab® (Belo Horizonte - Brazil). Mini-implants with a thickness of 1.8 mm, a thread of 7 mm, and a transmucosal profile of 4 mm were used for both sides.

The palate was scanned on July, 2020 (3 months 8 days), with an intraoral scanner (3Shape A/S - Copenhagen - Denmark) to fabricate the expander appliance customized for the small palate. After the virtual installation of the mini-implants, this positioning was grouped with the STL of the intraoral scan to create a single file that maintains their placement. Next, the file was 3D printed (Form2 printer and Grey Pro resin - Somerville - USA). To attach the device to the small anchors and keep them stable, support rings were modeled in CAD (Computer Aided Designer) and machined in CNC (Computer Numeric Control) to serve as a guide for installing the mini-implants in the mouth, determining both the axis and the insertion limit. Since the patient did not have any erupted teeth that would allow the bands to be installed, acrylic was used in the palatal region to adjust the 3D orientation of the device. Subsequently, a guide was created to replicate the precise placement of the device during fabrication when these implants were fixed (Figure 3). The development and virtual installation were performed by Kika Orthodontics (Sorocaba, Brazil).

The appliance was installed in the Surgical Center on September, 2020 (4 months 28 days) and activated according to the maxillary expansion (ME) protocol of four activations, corresponding to one complete rotation of the expander lathe. The placement procedure went smoothly, and the patient was discharged the day after surgery. At the first follow-up visit, one week postoperatively, it was indicated to start the activations of the MARPE-c, following the protocol of 2/4 turn per month, for three months, with a total expander screw opening of 2.5 mm, from the transoperative to the postoperative period. After the last activation, the appliance was left in place for two months for bone formation in the region of the midpalatal suture. The device was removed in an ambulatory setting on February, 2021 (10 months 13 days), without the need for anesthesia; the patient underwent a new CT scan on November, 2021 (1 year 7 months).

For this study, the Informed Consent Form and the Minor's Consent Form were signed by the patient's legal guardian (mother).

Treatment Objectives

The treatment objectives were to (1) improve the patient's quality of life (2) enable clinical interventions that facilitate proper rehabilitation of respiratory and swallowing functionality. The treatment planning was (1) maxillary expansion using a microimplant-assisted rapid palatal expander device (2) to correct pyriform aperture stenosis.

Treatment Alternatives

When dealing with patients who suffer from severe medical conditions, it was necessary to adopt a surgical intervention that takes into account the stomatognathic functions, such as breathing and swallowing. In the case of the child under consideration, a tracheostomy was performed to ensure the necessary respiratory support, while an oxygen tube was used to provide supplemental oxygen. This approach aimed to maintain the balance between the patient's medical needs and their stomatognathic functions.

Treatment Progress

Two CT scans were performed, an initial pre-RME (T1) and a final post-RME (T2), to investigate changes in the nasomaxillary complex. In evaluating the CT scans using the image manipulation software Dolphin Imaging (Dolphin Imaging & Management Solutions, Chatsworth, California Version 11.7 Premium), the patient's head was repositioned on the CT images according to the methodology proposed by Cevidanes et al. (2009) [17].

The changes in the nasomaxillary complex were observed in the maxilla, the nasal cavity, their areas, and the nasomaxillary complex airway volume. The measurement of the nasomaxillary complex airway volume began with the definition of the area in the midsagittal plane formed by its anatomical limits: posterior (defined by points C4, Ba, and PCP); anterior (defined by points Na, Rhi, ANS, PNS); superior (defined by anatomical points Na and PCP); and inferior (defined by point C4), as shown in Table 1.

Landmark	Definition
Na	Intersection of the frontonasal sutures in the median plane
Rhi	End point of the internasal suture
ANS	Projection formed by the fusion of the two maxillary bones at the intermaxillary suture
PNS	Most posterior point of the palatine bone and floor of the nasal cavities in the midsagittal plane
C4	Most anterior and superior point of the fourth cervical vertebra
Ba	Most anterior and inferior point on the border of the foramen magnum in the sagittal plane
PCP	Most posterior and superior point of the sella turcica

Table 1: Landmarks of the nasomaxillary complex.

(N) Nasion; (Rhi) Rhinio; (ANS) Anterior nasal spine; (PNS) Posterior nasal spine; (C4) Cervical; (Ba) Basicium; (PCP) Posterior clinoid process. Sources: Caple & Stephan (2016) and Hassan (2013).

After delineating the nasomaxillary complex area, this region was segmented. The air space filling was obtained in all sections - axial, sagittal, and coronal (Figure 4), allowing the air volume of the nasomaxillary complex to be measured.

The nasomaxillary complex changes observed in the maxilla and nasal cavity were measured in the axial section. The software's vertical reference line was positioned in the patient's midsagittal plane, and the horizontal line was placed at the right and left jugal cephalometric points.

The following measurements were made (Figure 5): Anterior maxillary width (AMW) - linear measurement of the buccal bone cortices in the right and left canine region; Posterior maxillary width (PMW) - linear measurement of the buccal bone cortices from the area of the right deciduous maxillary first molar to the area of the left deciduous first molar; Anteroposterior distance between the anterior and posterior maxillary widths (APMW) (Figure 5A). Anterior nasal width (NW) - linear measurement between the upper limits of the right and left infraorbital foramen; Posterior nasal width (PNW) - linear measurement between the internal bony lamina of the right and left nasal cavities at the level of the posterior nasal spine; Anteroposterior distance between the widths of the nasal cavity in the anterior and posterior regions (APNW) (Figure 5B); Nasal cavity area (NCA) - area bounded by the anterior and posterior regions of the nasal cavity (Figure 5C); Maxillary area (MA) - area bounded by the anterior and posterior regions of the maxilla (Figure 5D).

Results

The use of the microimplant-assisted rapid palatal expander device (MARPE-c) promoted improvement in snoring episodes and significant changes in the nasomaxillary complex observed in the maxilla, nasal cavity, and air volume of the nasomaxillary complex, as described in table 2.

Variables	T1	T2
NAV (mm^3)	1058.0	5735.0
AMW (mm)	20.0	23.5
PMW (mm)	29.1	43.5
APMW (mm)	11.00	15.7
NW (mm)	4.6	9.2
PNW (mm)	7.3	14.4
APNW (mm)	17.3	32.1
MA (mm^2)	263.6	484.4
NCA (mm^2)	125.3	393.0

Table 2: Pre-expansion (T1) and post-expansion (T2) nasomaxillary complex measurements.

NAV = nasal airway volume; AMW = anterior maxillary width; PMW = posterior maxillary width; APMW = anteroposterior distance between anterior and posterior maxillary widths; NW = nasal cavity width; PNW = posterior nasal cavity width; APNW = anteroposterior distance between anterior and posterior nasal cavity widths; MA = maxillary area; NCA = nasal cavity area; mm = millimeters; mm2 = square millimeters; mm3 = cubic millimeters.

The evaluation of T1 (pre-MRE) and T2 (post-MRE) CT scans, through 3D reconstructions, showed gain in air volume of the nasomaxillary complex (Figure 6) and satisfactory skeletal changes (Figure 7).

Discussion

SMMCI may be associated with several congenital nasal anomalies such as choanal atresia, intranasal stenosis, and congenital stenosis of pyriform aperture (CSPA) [18-20]. Choanal atresia is a bone or membranous obstruction of the posterior nasal aperture caused by a failure of oronasal disintegration. Intranasal stenosis is a bone narrowing of the nasal cavity between the pyriform aperture and the posterior choana, while CSPA is an anterior obstruction secondary to bony growth of the maxillary nasal processes [21-23].

Prenatal diagnosis of SMMCI can be performed by magnetic resonance imaging [24] and, after birth, since the clinical aspects among the possible alterations are similar, CT scan presents importance to distinguish which nasal anomaly is present [1,18,21].

Another form of diagnosis is an examination of the nose, which can be very difficult because of the small size of the nasal cavities [18,25]. Anterior rhinoscopy shows narrow nasal cavities at the expense of osseous submocosal prominence. The light of the cavities may be less than 2 mm [25]. In the case presented in this study, the patient had a diagnostic hypothesis of pyriform aperture stenosis, and during the initial evaluation by the pediatrician, it was not possible to pass a tube for nasal aspiration, as is routinely done in the nursery. At birth, he presented with dyspnea and cyanosis, demonstrating the presence of severe nasal obstruction [18,26].

The American Academy of Pediatric Dentistry (AAPD) recognizes the functional severity of this type of situation based on the fact that the neonate is a preferential nasal breather, and any cause of severe nasal obstruction can lead to respiratory failure, asphyxia, obstructive sleep apnea [18,21,26], as well as alteration of occlusion and craniofacial development [14,15]. Patients with obstructive sleep apnea have decreased upper airway volume and increased airflow resistance, leading to mouth breathing and craniofacial, muscular, and dental anomalies, such as transverse maxillary deficiency [14,15].

This case was challenging because the patient combined two syndromes, Down syndrome and SMMCI, which required a multiprofessional team [27], involving a pediatrician, an otorhinolaryngologist, and an orthodontist. The otorhinolaryngologist performed the surgical phase (correction of the pyriform aperture stenosis), and the orthodontist (MC Jr) installed the maxillary expansion appliance [28]. This treatment approach is chosen because the appliance can be used in periodontal patients where dental elements could not serve as an anchorage or in edentulous patients. This applicability enabled the appliance installation in the infant referred to in this study. The indication for maxillary expansion was based on the patient's severe respiratory difficulties and the presence of snoring, suggestive of episodes of obstructive sleep apnea, a serious condition for pediatric patients. It is known that the RME directly affects airway expansion and thus improves breathing [29-31]. Another advantage of this approach at such a young age of the child is the prevention of future oral sequelae (malocclusions) that could be presented due to the maxillary deficiency, such as lack of space for the maxillary dental alignment, occlusal disharmony between the dental arches, presence of a crossbite, and inadequate positioning of the tongue, which may assume a lower position (on the mandibular floor) and backward, and may interfere between the incisors, creating an anterior open bite [32,33].

In this patient, many factors are involved in the nasomaxillary complex changes, such as surgery to correct stenosis of the pyriform aperture, use of the MARPE-c appliance, which allowed transverse maxillary enlargement and adjacent areas, and skeletal growth inherent to the patient's age. The long interval between CT scans was due to the radiation doses during the examinations, which could affect the patient's growth and development. The tomographic images showed a significant difference in airway volume between the examinations, especially in the obstruction that the patient still had, despite having undergone correction of the stenosis of the pyriform aperture. In this situation, RME was a key element that allowed the development of the middle third of the face. This orthodontic procedure was a pioneer, as it had never been performed on an infant patient before. In the presented clinical case, ERM promoted the development of the middle third of the face and enlargement of the upper airways with improvement of respiratory function and action on the patient's snoring. This orthodontic procedure was a pioneer, as it had never been performed on an infant patient and proved to be safe and a good treatment option for increasing the volume of the nasomaxillary complex against possible obstructive sleep apnea.

Conclusion

Early diagnosis of SMMCI is of great importance to improve the patient's quality of life and to allow clinical interventions that can provide adequate rehabilitation of masticatory and phonetic functions, as well as deglutition and aesthetics. Using the MARPE appliance in infants proved to be a safe and effective procedure.

Highlights

- Infant patient, solitary median maxillary central incisor and palatal expansion technique
- Digital imaging planning of anchorage control using a mini-implant-assisted rapid palatal expander.
- Early diagnosis and intervention were key factors in improving nasal respiratory airflow and swallowing functionality.

CRediT Author Statement

Mario Cappellette Jr: Conceptualization, Methodology, Validation, Formal analysis, Investigation, Writing - Original Draft, Writing - Review & Editing, Visualization, Supervision, Project administration. Silvia Negrisoli: Writing - Original Draft, Visualization. Daniela Pimentel Machado Renofio Hoppe: Data Curation. Cristiane Barros André: Software, Data Curation. Lucas Fiorante Cappellette: Data Curation. Lucia Hatsue Yamamoto: Validation, Formal analysis, Writing - Original Draft, Writing - Review & Editing, Visualization.

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Availability of Data and Materials

The datasets used and/or analyzed during the current study are available from the corresponding author on reasonable request.

The authors declare that they have no competing interests.

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