

Bilateral Transverse Facial Cleft with Unilateral Complete Cleft Lip and Palate: A Case Report.

*BARDI M, *ADEBOLA RA, **JATTO BK,
AFEBGUA D, *AHMADU R

*Department of Oral & Maxillofacial Surgery,
**Department of Plastic Surgery, Federal
University of Health Sciences/ Federal
University of Health Sciences Teaching
Hospital, Azare, Nigeria.

Department of Paediatrics, *Department
of Oral & Maxillofacial Surgery, Federal
University of Health Sciences Teaching
Hospital, Azare, Nigeria.

Correspondence

Dr. Jatto B. Kabir
Department of Plastic Surgery,
Federal University of Health Sciences/
Federal University of Health Sciences
Teaching Hospital,
Azare, Nigeria
E-mail: djteasy1@gmail.com

ABSTRACT

Background: The simultaneous occurrence of a bilateral transverse facial cleft with unilateral complete cleft lip and palate is extremely rare especially where no syndromic association can be established. This has hardly ever been reported in African population. The management thus, poses a challenge as the sequence and timing of different aspects of care is not fully established.

Case description: We report the clinical and radiographic (craniofacial computed tomographic scan) findings and surgical management of a rare case of bilateral transverse facial cleft with unilateral complete cleft lip and palate highlighting the challenges of providing care.

Treatment: patient had lip repair, lateral cleft repair and palatal repair in that sequence using Millard's rotational advancement, straight line and the Bardach's two flap palatoplasty techniques respectively.

Conclusion: Obvious challenges have been documented however; conscious efforts are required to document treatment protocols that can help in diagnosis and treatment of rare facial clefts presentations.

Keywords: Orofacial cleft, Tessier number 7 cleft, Cleft lip and palate, surgical repair.

INTRODUCTION

The prevalence of cleft of the lip and palate among Nigerian population is about 0.5 in 1000 with lateral facial reported to be rare (Eppley et al., 2005; Butali et al., 2014). However, the simultaneous occurrence of a unilateral complete cleft of the lip and palate and lateral facial cleft is reported to be universally rare especially where non-syndromic (Lee et al., 2021). This has made it difficult to establish a sequence, plan, or timing of surgeries for treating patients presenting with the above anomaly (Lee et al., 2021). The aim of this article was to report this rare case and describe the management with the patient outcome. Written informed consent was obtained from the patient's parents for publication of this article and accompanying images. Ethical approval for this study was obtained from the hospital research and ethics committee.

CASE DESCRIPTION

The patient was born at home in January 2023 at 38 weeks of gestation but there was no history of

ante natal attendance. There was no history of any relevant abnormalities in the family, and no teratogenicity were identified clinically through history. She presented with bilateral lateral facial cleft, left unilateral complete cleft lip and palate, (Figure 1). The lack of geneticist in our environment made it difficult to establish any association with a specific syndrome or genetic abnormality echocardiography and general physical examination did not reveal any other physical abnormality.

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Clinical and detailed radiological examination (Figure 2) of the maxillofacial region was performed to assess the morphology of the cleft and plan treatment. There was a complete cleft of the lip and palate on the left with lateral clefts bilaterally running from the presumed oral

commissure about 6cm and 5cm on the right and left respectively towards but short of the lateral canthus of both eyes. A computed tomography scan with three-dimensional (3D) rendition showed that the transverse facial clefts had bony

involvement. The cleft of the right involved the zygomatico-maxillary suture and the maxilla without involving the right infraorbital rim. On the left, this is less severe causing only a dent at zygomatico-maxillary suture.



Figure 1: Picture at presentation with bilateral transverse facial cleft, left unilateral complete cleft lip and palate



Figure 2: Computed tomography scan with three-dimensional (3D) rendition showing bony involvements



Figure 3: Picture showing nutritional rehabilitation using nasogastric tube feeding



Figure 4: Postoperative picture showing lip repair

DIAGNOSIS

The diagnosis made was a bilateral Tessier 7 facial cleft simultaneously occurring with a left unilateral complete cleft of the lip and palate. Patient was planned for nutritional support and optimization

for lip and facial cleft repair at 3months, palatal repair at 9-12months, nose and lip revision at 5years, alveolar bone grafting at 7years following the Centre's protocol for management of cleft lip and palate.



Figure 5: Intraoperative picture showing repair of deficient buccal mucosal using the buccal fat pad



Figure 6: Immediate postoperative picture showing location of corner of the mouth

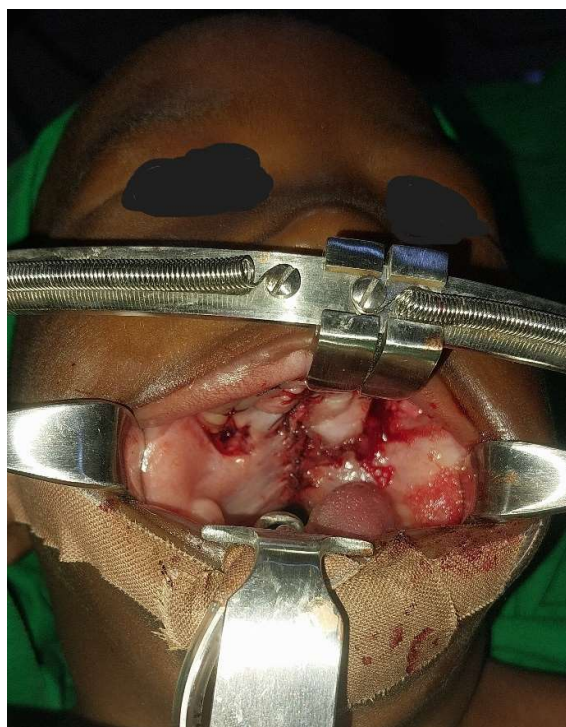


Figure 7: Immediate postoperative picture showing palatal repair using Bardach's two flap palatoplasty

TREATMENT

Although, the parents of the child were counselled on feeding and care for the child at presentation, at 2 months of age, the patient was admitted for malnutrition with a Z-score < 3 and chest infection as confirmed with pulmonary field consolidation on PA chest. Feeding optimization was done by the Paediatrician and Paediatrics nutritionist through tube feeding (Figure 3) with locally prepared F-75 and F-100 for two week and discharged with a Z-score <2. In addition, the mother was counselled on breastfeeding practice in addition to supplemental feeding using breast milk substitute. The chest infection was managed with parental antibiotics. Optimizing her greatly affected treatment timelines that she had lip repair at 5 months of age (Figure 4). Definite lip repair was performed using a modified Millard's rotational advancement flap. The bilateral facial cleft was corrected three months later using a straight-line technique. Closure was done in three layers (mucosal, muscle and skin). The attachment of the mucosal to the maxilla was carefully dissected to allow enough sulcus depth in the cheek area. Where the mucosal was deficient on the left, the buccal fat pad was utilized (Figure 5 & Figure 6). The commissure on both sides was determined using the point that was equidistant to the Cupid's bow peak and coincides

with the point where the oral mucosa begins to thin out. The palatal repair was furthered delayed because the mucoperiosteal dimensions were deficient in relation to the size of the defect. Patient had palatal repair at 15months using the Bardach 2-flap technique (Figure 7).

OUTCOMES

Craniofacial clefts are rare and due to their diverse presentation, their diagnosis and management protocol can be challenging and varied (Tessier, 1976; Paradowska-Stolarz et al., 2022). Most cleft centers in Nigeria repair the lip between 10-12weeks, (Efunkoya et al., 2015; Akinmoladun et al., 2017) but there are no clear recommendations on the appropriate timing of lateral cleft lip repair, even though an early correction will have huge psychological impact on patients and caregivers (Fletcher et al., 2012). Some studies have recommended that lateral cleft lip repairs should be performed on patients 3-4 months of age (Bordoloi & Saikia, 2014; Koh et al., 2019). In our case, early correction was marred by malnutrition which is not uncommon among children born with cleft (Delage et al., 2022). There was an instance where the child was admitted for parenteral nutrition. This seriously impacted the timing of surgical correction.

Our sequence of repair was lip at 5 months, bilateral lateral cleft at 8 months and palatal repair at 15 months. This sequence was followed because aside the fact that it is difficult to correct facial deformities in a single surgical procedure, depending on the age of presentation and the maturation or growth pattern of facial structures, it is important to consider the best possible outcome using developed landmarks and ease of correction (Alonso et al., 2008; Carstens, 2017).

The surgical correction of such composite defect is very challenging because just as the point of the oral commissure on both sides are needed for definite lateral cleft repair, so also an apex of the Cupid's bow on the lateral cleft segment is needed to correct the cleft lip (Lee et al., 2021). However, this patient did not have these reference points, thus, it was difficult to determine the order of surgical correction.

The apex of the Cupid's bow on the lateral cleft segment was determined using the point where the white row begins to disappear that coincides with the maximum bulbosity of the vermilion on that side (Tse, 2012) while symmetrical reference measurement of the horizontal lip length was used during lateral cleft correction (Adetayo et al., 2018). This did not exactly give symmetrical dimensions following repair but with healing, growth and development, it is expected to impact on minor differences (Cutting & Dayan, 2003). Minor irregularities observed included slight deviation of the columellar to the cleft side, slight asymmetry of nose and lip dimensions. Correction of these irregularities is planned for in a revision surgery in due course.

Although no complications other than minor scarring at the surgical site was observed, the patient was placed under general anaesthesia several times with the attendant risk of possible critical events (Habre et al., 2017). However, several procedures were required to correct these defects with no record of any critical event.

CONCLUSION

Here, we presented the case of a patient with a simultaneous presence of cleft lip and palate, bilateral transverse facial cleft. This rare case is difficult to repair due to the absence of anatomical landmarks on the lip on both sides, distortion of the nose and upper lip, distortion and deformity of the maxillary segments due to an alveolar cleft, and a wide cleft palate. Given that there have been no studies reporting details/protocols of surgeries for this rare phenomenon, this case may be helpful to surgeons working through a similar case in constructing a surgical plan, timing, and sequence.

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Conflicts of interest

The authors declare that they have no conflicts of interest.

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