

Congenital cheek teratoma with temporo-mandibular joint ankylosis managed with ultra-thin silicone sheet interpositional arthroplasty

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ABSTRACT

Primary cheek teratomas are rare with < 5 reported cases. None had associated temporo mandibular joint ankylosis (TMJA). The fundamental aim in the treatment of TMJA is the successful surgical resection of ankylotic bone, prevention of recurrence, and aesthetic improvement by ensuring functional occlusion. Early treatment is necessary to promote proper growth and function of mandible and to facilitate the positive psychological development of child. Inter-positional arthroplasty with ultra-thin silicone sheet was performed. Advantages include short operative time, less foreign material in the joint space leading to negligible foreign body reactions and least chances of implant extrusion. Instead of excising a large bony segment, a thin silicone sheet was interposed and then sutured ensuring preservation of mandibular height. Aggressive post-operative physiotherapy with custom made dynamic jaw exerciser was used to prevent recurrence.

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INTRODUCTION

Facial teratoma or dermoid is a rare, generally benign tumor. Less than 5% of those appearing in the head and neck of children are estimated to be malignant. Primary cheek teratomas are rare with ≤ 5 cases reported in the literature. None of these had associated temporo mandibular joint ankylosis (TMJA).

The fundamental aim in the treatment of TMJA is the successful surgical resection of ankylotic bone, prevention


of recurrence, and esthetic improvement by ensuring functional occlusion. Early treatment is necessary in children to promote proper growth and function of mandible and to facilitate the positive psychological development of child. Aggressive post-operative physiotherapy is essential to prevent recurrence.

CASE REPORT

A 4-year-old girl presented with progressively decreasing mouth opening since birth and facial deformity. According to the parents, at birth there was a large soft-tissue mass involving the entire right cheek with restricted mouth opening, which was excised at 6 months of age when biopsy confirmed it to be a mature teratoma. Decrease in mouth opening continued even after surgery, with hypertrophy of the right infraorbital region.

On examination, a large cheek scar with loss of subcutaneous tissue severely restricted mouth opening (inter incisor distance < 3 mm) and maxillary hypertrophy

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in the infra orbital region were observed [Figure 1]. Oral hygiene was poor with absence of anterior teeth in both upper and lower jaw.

Computed tomography face revealed loss of joint space, bony ankylosis between the coronoid process and zygomatic process of the maxilla with hypertrophy of infraorbital rim and the adjoining zygomatic arch [Figure 1].

Child underwent exploration of the facial skeleton using the previous preauricular scar. All soft-tissue adhesions between the cheek skin and the mandible were excised. Articular ankylosis had a fibrous component. Coronoidectomy was done to release the bony block between the infra orbital rim, zygomatic arch and the mandible with dramatic improvement in mouth opening [Figure 2]. After ensuring adequate mouth opening the hypertrophied malar prominence was shaved to ensure facial symmetry. As there was extensive scar tissue around the malar region most probably due to previous surgery, ultra-thin silicone sheet was used as an inter-positional material instead of standard temporalis muscle flap.

After complete transaction of the ankylotic bone, the gap was kept bare enough so as to pass 0.2 mm ultra-thin silicon sheet of size 5 × 5 cm [Figure 3]. Medially, it was anchored with 3-0 nylon to medial pterygoid muscle and laterally to periosteum of zygomatic arch so as to fix the sheet securely and also to cover the two transacted bone ends completely.

Post-operatively child was put on extensive physiotherapy using an acrylic dynamic jaw exerciser [Figure 4 and Video 1]. After 6 months of follow-up child has painless jaw movement with 2.5 cm inter incisor distance. Child is now planned for further refinement of facial contour.

DISCUSSION

Congenital cheek teratomas are rare with ≤ 5 cases reported in literature.^[1-3] They grossly appear as heterogeneous masses with solid and cystic components. They are believed to arise from migration and entrapment of mesoderm with ectoderm during embryogenesis of the oral cavity.^[3]

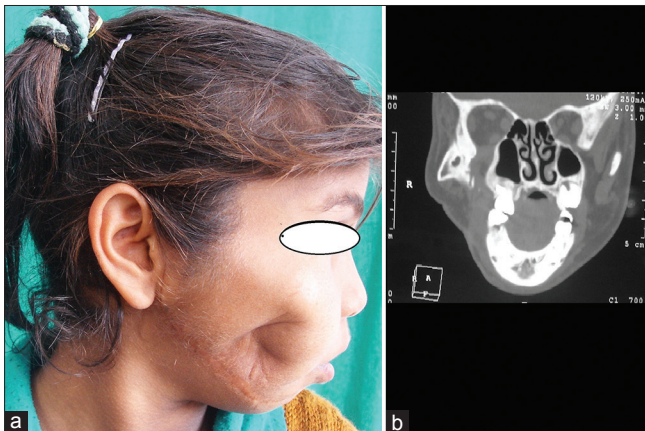


Figure 1 (a and b): Scar in the center of cheek with restricted mouth opening and computed tomography showing bone involvement

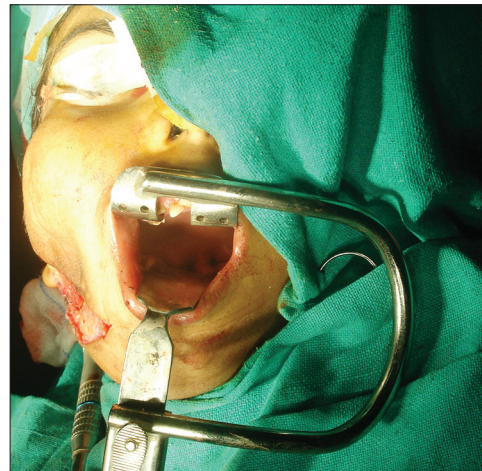


Figure 2: Restoration of mouth opening

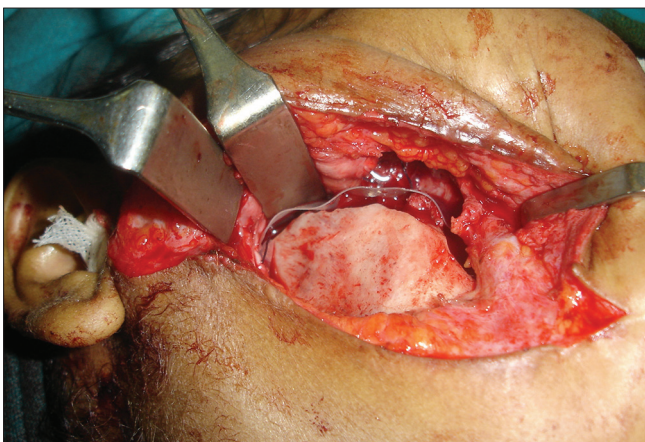


Figure 3: Silicone sheet placed between bone segment

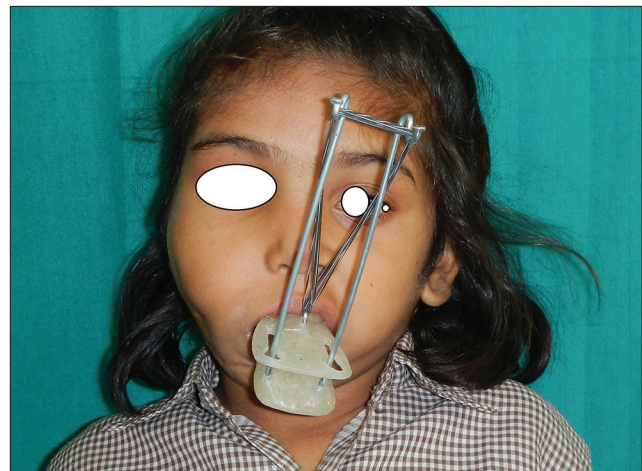


Figure 4: Dynamic jaw exerciser in place

Cheek dermoids do not cause respiratory difficulty.^[2,3] Feeding was a problem, not only due to the mechanical obstruction but also due to TMJA. Excision done at 6 months of age corrected the cosmetic deformity but the ankylosis continued to increase with age.

TMJA is most commonly associated with trauma.^[4] Congenital cases have been reported. They are rare and should be described in detail for better understanding of the disease. None of the cases of congenital TMJA previously described had any association with congenital cheek teratoma or dermoid.^[5]

All TMJA presenting at birth are not truly congenital. The etiology of this disorder is heterogeneous and should be differentiated from the acquired forms, such as traumatic forceps delivery and infections.^[6] Congenital TMJA is a malformative sequence. Joint deformity creates a cascade of events leading to facial deformity, occlusion abnormality and poor nutritional status. It is a pattern of multiple anomalies derived from a single known or presumed previous anomaly or mechanical factor like a condylar region malformation.^[5]

Clinical presentation is characterized by significant reduction in mouth opening and variable degrees of facial disfigurement such as micrognathia, asymmetry, jaw deviation and malnutrition. The cause of ankylosis in our patient was both articular and extra articular. The malformation of the facial skeleton was part of the teratoma complex leading to congenital trismus.

At birth some mobility is present due to lack of ossification of the cranial sutures and trismus is apparent by 6th month of age as in our case.^[5]

The fundamental aim in the treatment of TMJA is the successful surgical resection of ankylotic bone, prevention of recurrence, and esthetic improvement by ensuring functional occlusion. Early treatment is necessary to promote proper growth and function of mandible and to facilitate the positive psychological development of child.^[7-9]

To minimize the chances of re ankylosis and to conserve as much height as possible it was decided to perform an interpositional arthroplasty.

We used a newer technique for joint reconstruction, to prevent recurrence of ankylosis. We used ultra-thin silicon sheet of thickness of 0.2 mm replicating the normal intra-articular disc. The fixation of the sheet reduces the chances of re-ankylosis by preventing displacement or extrusion of the implant.^[10] The advantages were short operative time (1.5 h), minimal foreign body load in the joint space hence least chance of extrusion and no loss

of mandibular height. Post-operative physiotherapy for about 1 year is as important as surgery if not more, in the overall outcome for patient. Custom made dynamic acrylic jaw exerciser is easy to assemble and can be used in children as small as 3-4 years of age.

CONCLUSION

Cheek dermoid is a rare entity with no reports of its association with TMJA. Congenital TMJA becomes apparent at about 6 months of age. Prompt surgical treatment is required for the treatment of both teratoma and ankylosis. Inter-positional arthroplasty is the treatment of choice. Early surgery prevents growth abnormalities of the facial skeleton. Ultra-thin silicon sheet has been rarely used in children. Post-operative physiotherapy is an important part of the rehabilitation process. Dynamic jaw exerciser allows painless physiotherapy even in young children.

Our technique is a good alternative to conventional techniques but more randomized trials are required to assess its effectiveness for universal adoption.

REFERENCES

1. Sreetharan V, Kangesu L, Sommerlad BC. Atypical congenital dermoids of the face: A 25-year experience. *J Plast Reconstr Aesthet Surg* 2007;60:1025-9.
2. Demirtas I, Kutluhan A, Ugras S, Beckerecioglu M, Akpoltan N. Primary immature teratoma of the cheek: 2 cases. *East J Med* 1997;1:47-8.
3. Morof D, Levine D, Grable I, Barnewolt C, Estroff J, Fishman S, *et al.* Oropharyngeal teratoma: Prenatal diagnosis and assessment using sonography, MRI, and CT with management by ex utero intrapartum treatment procedure. *AJR Am J Roentgenol* 2004;183:493-6.
4. Kaban LB, Perrott DH, Fisher K. A protocol for management of temporomandibular joint ankylosis. *J Oral Maxillofac Surg* 1990;48:1145-51.
5. Gil-da-Silva-Lopes VL, Luquetti DV. Congenital temporomandibular joint ankylosis: Clinical characterization and natural history of four unrelated affected individuals. *Cleft Palate Craniofac J* 2005;42:694-8.
6. Baraldi CE, Martins GL, Puricelli E. Pseudoankylosis of the temporomandibular joint caused by zygomatic malformation. *Int J Oral Maxillofac Surg* 2010;39:729-32.
7. Chidzonga MM. Temporomandibular joint ankylosis: Review of thirty-two cases. *Br J Oral Maxillofac Surg* 1999;37:123-6.
8. Shashikiran ND, Reddy SV, Patil R, Yavagal C. Management of temporo-mandibular joint ankylosis in growing children. *J Indian Soc Pedod Prev Dent* 2005;23:35-7.
9. Topazian RG. Etiology of ankylosis of temporomandibular joint: Analysis of 44 cases. *J Oral Surg Anesth Hosp Dent Serv* 1964;22:227-33.
10. Kalra GS, Kakkar V. Temporomandibular joint ankylosis fixation technique with ultra thin silicon sheet. *Indian J Plast Surg* 2011;44:432-8.

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