

Congenital Temporomandibular Joint Ankylosis: Case Report and Literature Review

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Abstract

Congenital temporomandibular joint (TMJ) ankylosis is a rare craniofacial malformation which involves the fusion of the mandibular condyle to the cranial base without any form of trauma or infection. It is severely disabling, both functional and aesthetic (with limited mouth opening, feeding difficulties, and progressive facial imbalance at birth). The scenario in this report is an 8-year-old girl who presented with the complaint of left-sided bony ankylosis of the TMJ. The affected side was completely fused with the bony mandible, and the mandible was hypoplastic in the affected side, which was proven by the use of computed tomography with 3D reconstruction. The operation performed on the patient was the gap arthroplasty by the preauricular method using the insertion of a temporomyofascial flap, and the next step includes bilateral coronoidectomies. Physiotherapy was also initiated on the third day of operation to ensure that the jaws moved. At 3 months follow-up, the patient had no recurrence or complications of the mouth opening of 29 mm. The case has pointed out the necessity to diagnose early, perform appropriate surgical procedures, and provide consistent physiotherapy to achieve the best outcomes in congenital TMJ ankylosis.

Keywords

Congenital Temporomandibular Joint Ankylosis, Gap Arthroplasty, Temporomyofascial Flap, Coronoidectomies

1. Introduction

Congenital temporomandibular joint (CTMJ) ankylosis is a rare form of develop-

mental anomaly, where the mandible condyle ankylosises with the temporal bone either at birth or shortly after. Ankylosis congenita, unlike acquired ankylosis, occurs without trauma, infection, or other systemic inflammatory pathologies, and in such situations, no specific external cause can usually be identified [1]. The severe functional and aesthetic outcomes of the condition are decreased motility of the jaw, feeding issues, speech disabilities, and aggravated facial asymmetry during the development stage.

The temporomandibular joint is formed by the independent mesenchymal condensations of the mandibular condyle and temporal glenoid fossa in embryogenesis [2]. Failure of this process (as a result of vascular compromise, intrauterine infection, or genetic mutations) may lead to abnormal ossification of the joint surfaces and bony fusion of the joint surfaces [3]. Its aetiology is not yet clear, and the majority of them are termed idiopathic. Though congenital ankylosis is usually unilateral, bilateral cases have been reported, and the ankylosis may be of a fibrous or bony nature depending on the extent of the fusion [4].

The early diagnosis is very important, because in the growing years, ankylosis may lead to the impairment of the normal development of the mandible and, as a result, cause the secondary defects of micrognathia and occlusal cant [5]. Imaging modalities, especially computed tomography with 3D reconstruction, are crucial in making the diagnosis and also establishing the extent of bony fusion.

CTMJ ankylosis management should be multidisciplinary, with the participation of maxillofacial surgeons, otolaryngologists, and physiotherapists. Surgery has been the foundation of treatment [6] [7]. The main issue with paediatrics is the low compliance rates, and this poses a challenge because postoperative physiotherapy is a mandatory requirement to keep the joints moving and prevent recurrence [8].

In the present report, a somewhat rare case of congenital unilateral bony TMJ ankylosis on the left side in an 8-year-old female, who successfully underwent gap arthroplasty, interpositional, left temporomyofascial flap, and organised postoperative physiotherapy to be able to open her mouth and achieve functional facial appearance, is presented [9].

2. Case Presentation

An 8-year-old girl presented to the Otolaryngology outpatient clinic together with her mother and mentioned that she had not open her mouth since she was five years old. It was associated with feeding issues, including difficulty during breastfeeding and a progressive facial asymmetry increasing with the age of the child. The patient had normal developmental milestones. There was no history of trauma, infection, surgery, family history of similar conditions, congenital disorders, craniofacial abnormalities, or musculoskeletal disorders.

Clinical evaluation revealed the patient had a severe restriction in opening the mouth, with the greatest incisal opening estimated at only a few millimetres. The chin was in an anti-clockwise position, and the lower facial outline was asymmet-

rical, which was consistent with unilateral mandibular hypoplasia. The occlusal plane was tilted, there was absence of cranial nerve loss, there was no syndromic defect such as ear deformities, cleft palate and other systemic defects.

The left temporomandibular joint (TMJ) showed complete bony fusion with a three-dimensional (3D) reconstruction on a computed tomography (CT) scan, which was a true case of bony ankylosis. CT demonstrated complete bony fusion of the left TMJ, enlargement of the mandibular condyle, pseudoarthrosis, and hypoplasia of the left mandibular ramus and condyle. The imaging also revealed that it was shortening the mandible on the same side, thus causing deviation of the chin on the affected side, the opposite joint was unaffected. Taken together, the results formed the diagnosis of unilateral left-sided congenital bony TMJ ankylosis. The preoperative clinical appearance and radiological findings are shown in **Figures 1-4**.



Figure 1. Frontal view in open-mouth position reveals severe restriction of mouth opening secondary to long-standing temporomandibular joint ankylosis.

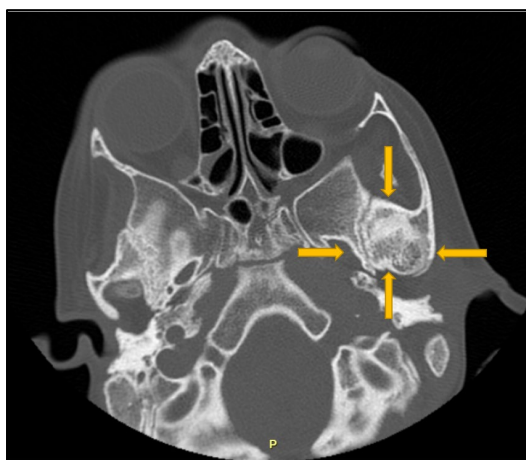


Figure 2. Axial view showing left TMJ bony fusion.

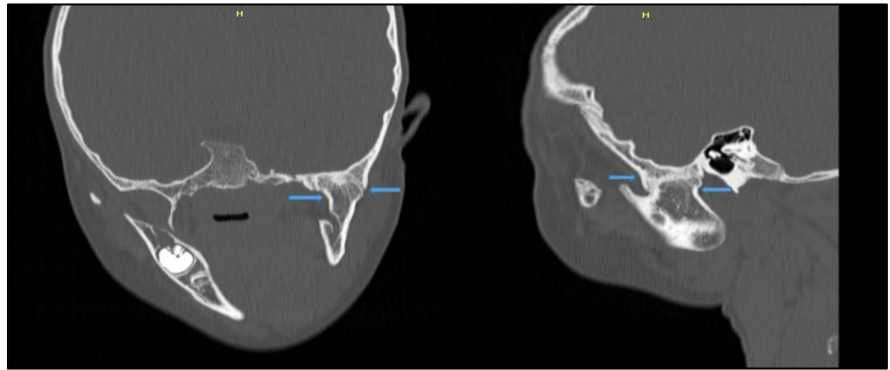


Figure 3. Coronal and sagittal views demonstrating pseudoarthrosis formation and hypoplastic left mandibular condyle.

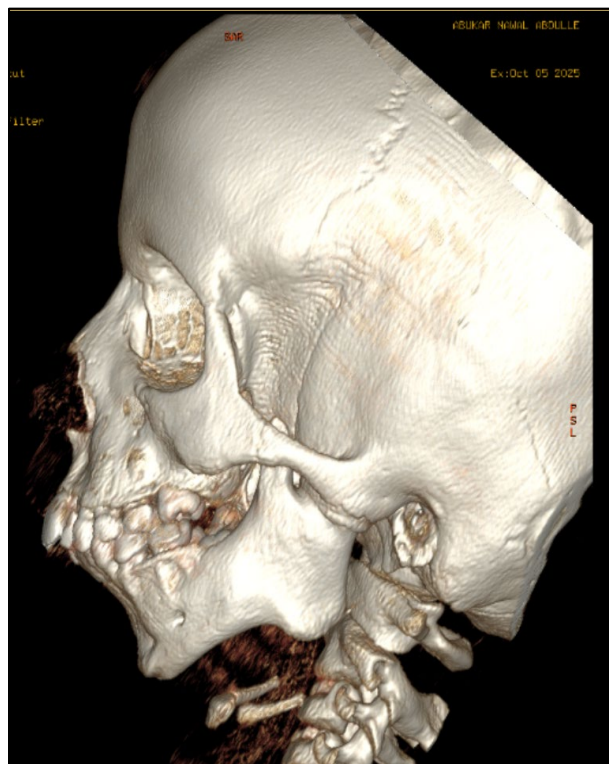


Figure 4. 3D CT image showing chin deviation toward the affected side.

The patient was scheduled to be operated on under general anaesthesia to have gap arthroplasty. An incision was made before the ear, approaching the ankylosing joint and the bony fusion was resected. A temporomyofascial flap was then lifted and turned over into the surgical area to act as an interpositional barrier between the bone surfaces, which were resected in an attempt to prevent reankylosis. Bilateral intraoral coronoidectomies were further done to liberate more muscle restrictions to improve jaw mobility. A maximal incisal opening (MIO) of 29 mm was recorded intraoperatively, which ensured that the joint was moving well. Representative intraoperative photographs illustrating the surgical approach of the ankylotic mass are shown in **Figure 5** and **Figure 6**.



Figure 5. Intraoperative view of the preauricular incision during gap arthroplasty.

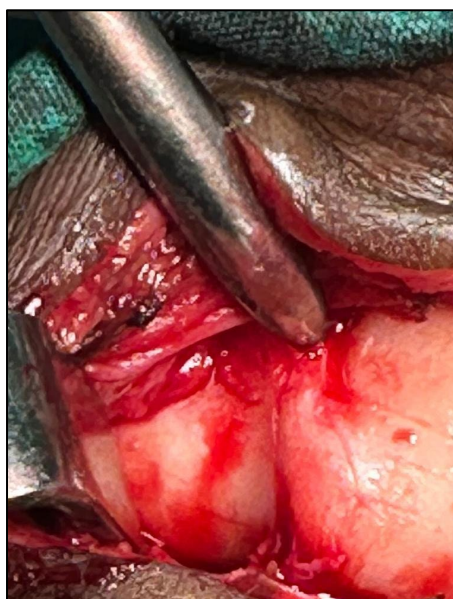


Figure 6. Intraoperative view showing the surgical field of the ankylotic mass during gap arthroplasty.

During the initial day of the postoperative period, the patient had a stable maximal incisal opening (MIO) of 29 mm without any neurovascular compromise or infection. Physiotherapy was initiated with the help of controlled mouth-opening and active-assisted movements with the jaw on the third day to maintain the range achieved. The patient and her mother were informed about the need for regular exercise to prevent relapse, and follow-ups were used to monitor adherence. The postoperative lateral view demonstrating the healed preauricular surgical incision and restoration of facial contour after gap arthroplasty is shown in **Figure 7**.



Figure 7. Immediate postoperative lateral view demonstrating the healed preauricular surgical incision and restoration of facial contour after gap arthroplasty.

In a three-month follow-up period after surgery, the patient was found to have continued improvement of the mouth opening, with an MIO of 29 mm with no discomfort, pain, or ankylosis recurrence. The degree of mastication and speech was good, and positive changes in facial symmetry were observed. The surgery had a positive outcome, as the postoperative functional and aesthetic stability signifies a functional and rehabilitative success in the treatment of congenital ankle-ankylosis of the TMJ. The postoperative appearance and improvement in mouth opening are demonstrated in **Figure 8**.



Figure 8. Postoperative clinical photograph demonstrating improved maximal mouth opening following gap arthroplasty and bilateral coronoidectomies.

3. Discussion

Congenital temporomandibular joint (TMJ) ankylosis is an infrequent but important clinical phenomenon which presents distinct diagnostic and treatment problems [10]. It is defined as a bony or fibrous union of the condyle of the mandible to the temporal bone, occurring at birth or in the near future and not due to infection or trauma [11]. The fact that this patient did not have such acquired causes helps to assert the diagnosis of a true congenital origin [12]. The lack of facial trauma history, local, or systemic infection, prior surgery, or other known etiologic factors aided in the diagnosis of congenital TMJ ankylosis in this patient. Early ankylosis of the joints disrupts the development of the mandibular growth, leading to an increasing face asymmetry and limited mouth opening, diminished mastication and speech [13]. The severity of deformity is strongly dependent on the timing of joint fusion; earlier fusion results in greater structural and functional impairment [14].

TMJ ankylosis is a complicated issue that should be handled carefully in pre-operative planning [11]. The surgical treatment objectives include the restoration of mandibular movement, re-establishment of joint functions, and reduction of recurrence [5]. Gap arthroplasty using interpositional material is also one of the surest methods in surgery among the available alternatives in the pediatric age group. It allows full resection of the ankylotic mass and the provision of a sufficient distance between the temporal bone and mandibular ramus [14]. The key to the success of this procedure is largely the maintenance of this space at the post-operative stage in order to avoid refusion. In the case in point, the temporomyofascial flap was used as an interpositional material due to its good vascularity, position in the operating field, and ability to prevent re-ankylosis [2].

Additional treatment measures mentioned in the literature are costochondral graft reconstruction, and alloplastic joint replacement. In this pediatric patient, however, the choice of gap arthroplasty with an interpositional temporomyofascial flap was favored due to its lack of donor-site morbidity (related to graft harvesting), maintains growth potential, and offers a well-vascularized local tissue barrier to minimize the risk of re-ankylosis.

Bilateral coronoidectomy was also done to overcome the possible muscular limitation that could have caused an inhibition of postoperative mobility. This may be a critical step in children, where hyperactivity and fibrosis of the masticatory muscles may further restrict the movement of the mouth. Adequate intraoperative maximal incisal opening (MIO) is a vital indicator of success in the long run [10]. An intraoperative MIO of 29 mm was obtained in this patient, which indicates adequate relaxation of the ankylotic and muscular restraints [5].

Physiotherapy after surgery is one of the pillars of ankylosis management because recurrence of the condition is directly attributed to poor movements in the jaw after the surgery. This is because early introduction of exercises remodels the neocondylar joint and maintains mobility of the joint [7]. The level of compliance

may, however, be difficult among the children, and the parental involvement would prove essential. Here, physiotherapy was started on the third day of surgery, and a set of exercises involving mouth opening was gradually undertaken [8]. The patient reported no discomfort, pain, or recurrence during the three-month follow-up period, where she had a stable maximal incisal opening (MIO) of 29 mm.

Even though the risk of recurrence remains real as reported in the literature, it is possible to obtain positive results due to the early detection of the condition, an accurate surgical procedure, and regular physiotherapy. The case highlights the need to incorporate multidisciplinary intervention where surgical skills and rehabilitative treatment are incorporated to replace the lost functionality as well as the facial unity in congenital TMJ ankylosis [9].

The follow-up period is a limitation in this case as it was relatively short, three months. This period showed that the patient had shown good mouth opening and functional improvement but still, there is a possibility of recurrence of TMJ ankylosis especially in children who are growing. Follow-up will thus be scheduled on a long-term basis to assess jaw functionality, mandibular growth, facial symmetry and to detect recurrence at an early stage.

Acknowledgements

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Consent

Written informed consent was obtained from the patient's guardian for publication of this case and accompanying images. Please see the Appendix section.

Author Contributions

Mohamed Nor Abdi: Conceived the study, managed the patient, performed the surgery, conducted the literature review, drafted the manuscript, and critically revised the final version.

Mohamed Osman Dahir Alasow: Interpreted the radiological images and contributed to the clinical diagnosis.

Abdulkadir Ahmed Mohamed: Assisted in patient management and clinical data collection.

Abdirahman Ahmed Ali: Prepared the clinical photographs and surgical figures, designed the figure layouts, assisted with clinical data collection, and critically reviewed the manuscript.

Abdikarim Mohamed Ali: Supervised the work, reviewed and edited the manuscript, and approved the final version.

All authors read and approved the final manuscript and agree to be accountable for all aspects of the work.

Conflicts of Interest

The authors declare that there is no conflict of interest regarding the publication of this paper.

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Appendix

Patient Consent Form for Case Report and Use of Clinical Images

I, Aisha Ibrahim Hussein (parent/guardian name), being the legal guardian of Amal Abdelhadi Abukar (child's name), hereby give my consent for the use of my child's clinical information and images (including facial photographs, radiographs, CT scans, and intraoral pictures) for educational and scientific publication purposes, including case reports, journal articles, or conference presentations. I understand that: - My child's name and any identifiable information will NOT be published or revealed. - The images will be used only for medical and educational purposes. - Providing this consent is entirely voluntary, and refusal will not affect my child's treatment or care in any way. - I may withdraw my consent at any time before publication, without any consequences to my child's medical care. **Guardian Details:**
 Name: Aisha Ibrahim Hussein
 Relationship to Patient: Mother
 Signature: [Signature] Date: 07-11-2023

Witness:
 Name: Dr. Abdulkadir Ahmed
 Signature: [Signature] Date: 07-11-2023

Treating Physician:
 Name: Dr. Mohammed Nor Abdi
 Signature: [Signature] Date: 07-11-2023

Thank you for your contribution to medical education and scientific advancement.