



Zygomatico-coronoid Bony Ankylosis Co-existing with Temporo-mandibular Joint Ankylosis: Unusual Radiological Findings in a Post-traumatic Maxillofacial Patient

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Authors' contributions

This work was carried out in collaboration among all authors. Authors JSD, IA and ROB designed the study, performed the statistical analysis, wrote the protocol and wrote the first draft of the manuscript.

Author MIS managed the analyses of the study and the literature searches. All authors read and approved the final manuscript.

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Case Report

ABSTRACT

Zygomatico-coronoid ankylosis is an unusual event. Twenty-four cases have been reported so far in the English literature but the exact incidence might be more. In this case report, the management of a 30-year-old male patient with zygomatico-coronoid bony ankylosis is presented following road traffic accident.

Examination revealed moderate head injury with basal skull fracture, fractured mandible and dislocated right temporomandibular joint (TMJ). There were also fractured ribs, right femur and ankle fractures. Craniofacial CT scan showed compound fracture of the left parasymphysis of the mandible with supero-lateral fracture dislocation of the right mandibular condyle. He had mandibular fracture fixation with metal plates and screws and closed reduction of right TMJ. He

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was subsequently discharged with strict instructions on active jaw exercise. He was lost to follow up for 3 months and on re-presentation in the hospital, mouth opening was 1.4 cm. Repeated CT scan showed bony fusion involving the right zygomatic arch and the coronoid process. Regarding treatment, surgical approach to the coronoid process was performed intraorally, however, because of persistent restricted mouth opening intraoperatively, a pre-auricular approach using the Bramley-Alkayat incision was used to assess the condyle in order to perform high condylectomy and utilize temporal muscle as inter-positional material.

Keywords: Ankylosis; coronoid; coronoidectomy; zygoma.

1. INTRODUCTION

The temporomandibular joint is the most advanced joint in the body because of unique features which include; closest joint to the brain, both joints must open synchronously, it has two joint spaces with two different movements occurring in them, it is the only joint lined with fibrocartilage and it is the only joint that factors remote to the joint, such as the teeth in occlusion, have influence on [1,2]. Furthermore, several studies have reported the link between emotional and psychological status of individuals with temporomandibular joint diseases [3-5].

Extracapsular ankylosis of the temporomandibular joint is a very serious condition that causes either limitation or inability to open the mouth. It is an extrinsic condition characterized by mandibular immobility as a result of fusion between the coronoid process of the mandible with the maxilla, temporal or zygomatic bones [6]. Aetiology of such extrinsic conditions include; gunshot injuries, zygomatic complex fractures with/without coronoid fracture, infratemporal space infections, chemical burns, mandibular fractures and extension of intra-articular ankylosis [1,7].

The main symptom of this condition is limited mandibular movements caused by the union of the zygoma and the coronoid process of the mandible [8]. The disorder also has an adverse effect on the psychology as well as social life of the individual that may lead to overall poor quality of life [8].

Literature search has reported only 24 cases of zygomatico-coronoid bony ankylosis (ZMCA) [9]. Most cases resulted from trauma [8]. The aim of this case report is to add to the available data on extra-articular temporomandibular joint ankylosis affecting the zygomatic bone and the coronoid processes of the mandible in a post-traumatic maxillofacial patient.

2. CASE REPORT

A 30-year-old male patient presented in the accident and emergency department of our hospital with multiple injuries following a road traffic accident. On examination, he had moderate head injury with basal skull fracture, mandible and rib fractures, fractured right femur and ankle.

Craniofacial computed tomographic scan showed compound fracture of the left parasymphysis of the mandible with supero-lateral fracture dislocation of the right mandibular condyle (Fig. 1a, 1b and 1c). After stabilization in intensive care unit and clearance from neurosurgeons and cardiothoracic surgeons, he underwent closed reduction of the superior-lateral fracture dislocation of the right condyle and open reduction and fixation of the left parasymphyseal fracture of the mandible with miniplate osteosynthesis under general anaesthesia (Fig. 2a, 2b, 2c and 2d).

Post-operative recovery was uneventful. He was discharged to outpatient clinic where regular jaw exercise was done. Mouth opening achieved during clinic visit was above 3.5 cm.

He was lost to follow up for 3 months but later presented back to the clinic with limitation in mouth opening. The inter-incisal distance at presentation was 1.4 cm. Attempt to proceed with jaw exercise proved difficult. A new CT scan was done which showed an unusually excessive bone formation in the superior aspect of the right ramus near the mandibular notch and coronoid process extending to and uniting with the zygomatic arch (Fig. 3a, 3b, 3c and 3d). There was also reduced joint space in the right temporomandibular joint.

Another surgery was conducted to release the extra –and intra-articular joint ankylosis. After releasing the zygomatico-coronoid bony ankylosis via intraoral approach, the mouth

opening did not improve. It was then decided to carry out high right condylectomy via Bramley-Alkayat incision. After the condylectomy, adequate mouth opening of 3.5 cm was achieved intraoperatively. Active jaw exercise was commenced early postoperatively. He was discharged to out-patient department with strict instruction on jaw opening exercises. His last clinic visit was 2 months after the second surgery with 3.5 cm inter-incisal distance. The patient relocated to another part of the country and asked for a medical report so as to continue with the postoperative rehabilitation in the new place of abode.

3. DISCUSSION

Several complications such as effusion, internal joint derangement, haemarthrosis, dislocation, fibrous adhesion, temporomandibular joint ankylosis and extra-articular temporomandibular joint ankylosis can arise following direct and/or indirect trauma to the temporomandibular joint leading to limitation or deviation of the jaw opening [10]. Extra-articular bone formation in this location is unusual following trauma, however, in other parts of the body such as the hip, shoulder, elbow and knee joints, they are frequently affected by extra-articular ossification [10].

Since extra-articular bone formation is rare in the maxillofacial region, the mechanisms by which the ankylosis develops remain

ambiguous. However, one popular theory was that metaplastic changes occur in connective tissue elements that normally do not have osteogenic potential following trauma, infection or surgery [8]. We believe that the traumatic dislocation of the right temporomandibular joint in our case might have caused shearing of bone in the region causing connective tissue to have osteogenic potential and thereafter leading to bony ankylosis. It has been reported that over 88% of cases of pseudo-ankylosis are associated with a history of trauma [11]. Traumas, with or without fracture, including surgery, can induce the formation of fibrous adhesion in the temporal and masseter muscles [11].

Management of extra-articular temporomandibular joint ankylosis is excision of the extra-articular bony mass and/or coronoidectomy, however, there is debate as to whether the approach should be intraoral or extraoral [12]. Intraoral approach leaves no scar mark on face and facial nerve injury is totally avoided, however, access is difficult and limited. Extra-orally, Bramley-Alkayat [13] approach gives good access, but approach may be associated with facial nerve injury in inexperienced hands [12]. Additional benefits of the Bramley-Al-Kayat incision include: reduced likelihood of sensory damage, simplicity of administration, proximity to the temporal joint, and successful functional and clinical result [14,15].

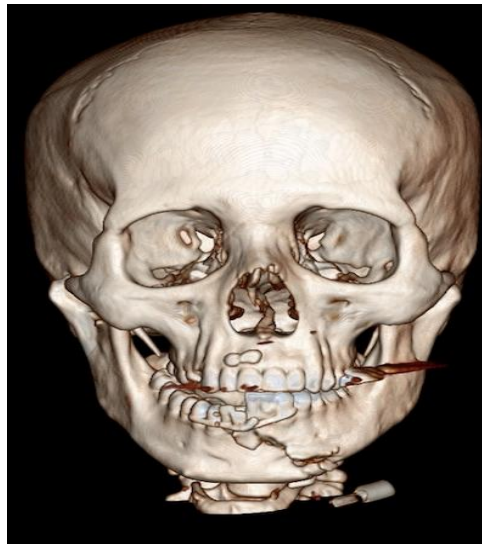


Fig. 1a. 3D reconstructive CT scan showing the left mandibular parasymphyseal fracture of the mandible

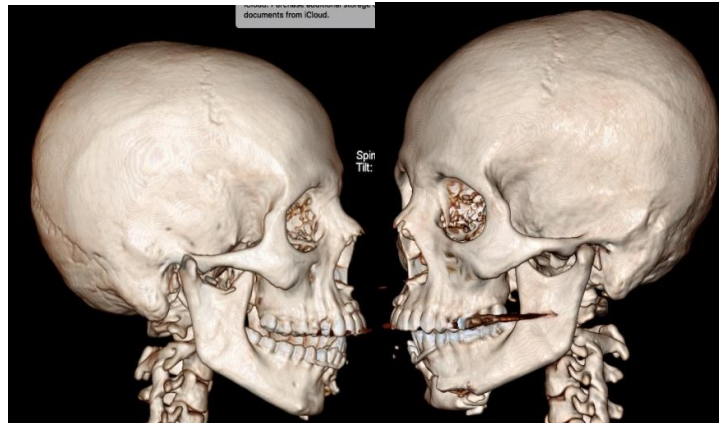


Fig. 1b and 1c. 3D reconstructive CT scan showing the right superolateral dislocation of the condyle and left parasymphyseal fracture of the mandible

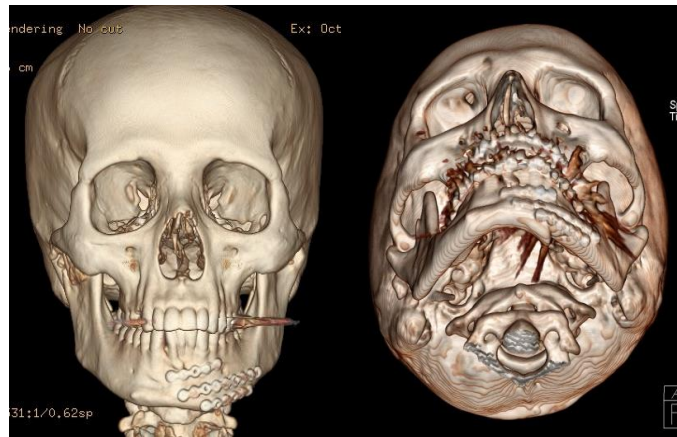


Fig. 2a and 2b. Postoperative CT scan with 3-D reconstruction showing fixation of the left mandibular parasymphyseal fracture with three miniplates and screws. Also both condyles are sitting in the glenoid fossae

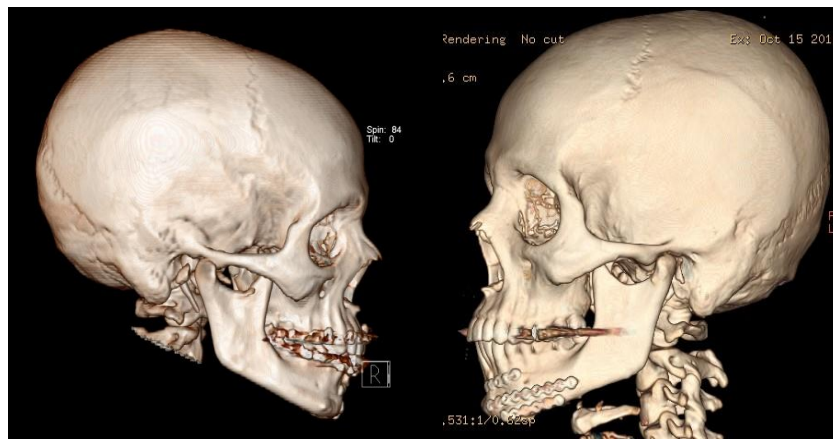


Fig. 2c and 2d. Postoperative CT scan with 3-D reconstruction showing satisfactory reduction of dislocated right condyle and also well positioned left condyle

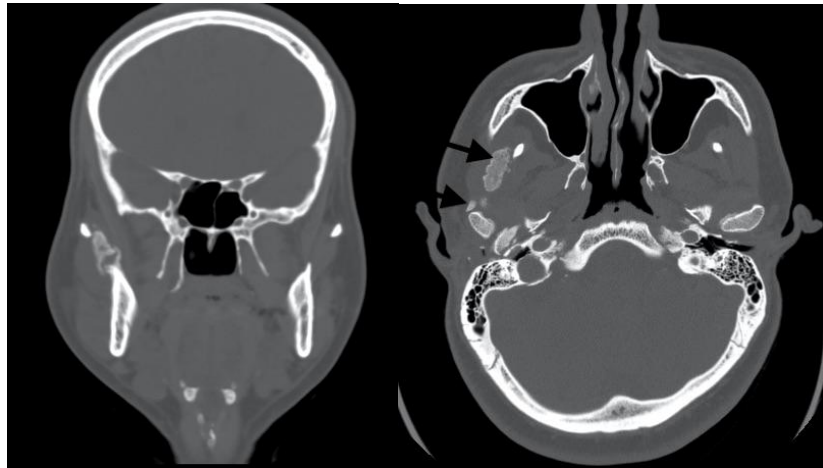


Fig. 3a and 3b. 3a showing coronal section of CT scan showing the right zygomatico-coronoid bony ankylosis. 3b showing axial section of CT scan showing both the right zygomatico-coronoid bony ankylosis and condylar-glenoid ankylosis separately (black arrows)

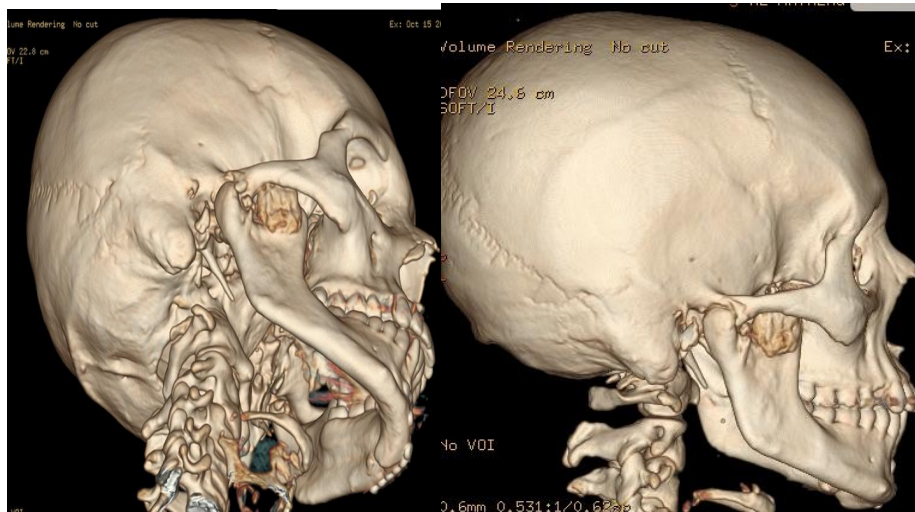


Fig. 3c, 3d. CT scan with 3-D reconstruction depicting the zygomatico-coronoid bony ankylosis and condylar-glenoid ankylosis separately

Initial approach in our case was intraoral to remove the extra-articular bony mass, however, when the mouth opening was inadequate, it was decided to open up the ankylotic right temporomandibular joint via preauricular approach using the Bramley-Alkayat incision through which high condylectomy and replacement of the meniscus with temporalis muscle flap as inter-positional material was performed. Access to the affected site was excellent via this approach and sufficient removal of ankylotic osseous mass was made possible. We did not perform coronoidectomy in our case as the bony mass was removed preserving the coronoid process.

Early postoperative mouth opening exercises, strict follow-up, and even forceful opening are necessary to overcome postoperative adhesions and or re-ankylosis that may develop [16,17]. With bleeding and lack of adequate physiotherapy together with the young age of the patient where there is high osteogenic potential, ankylosis is more likely to occur. To be more specific, in our patient coupled with trauma as etiological factor, there was lack of adequate physiotherapy following the closed reduction of the supero-laterally dislocated right temporomandibular joint. Patient's compliance to aggressive physiotherapy is essential to prevent re-ankylosis [18].

4. CONCLUSION

Zygomatoco-coronoid ankylosis is a very atypical condition therefore oral and maxillofacial surgeon should have a thorough knowledge of the clinical and radiographic features of this rare clinical entity. Most extra-articular ankylosis involving the coronoid process results from coronoid hypertrophy in long-standing cases of ankylosis. Fusion is uncommon especially in an intact coronoid process. The exceptional piece of our case is that the fusion between coronoid process and zygomatic arch was caused by an island of bone which could be a result of extrarticular ossification due to inadequate jaw physiotherapy after reduction of the traumatic fracture dislocation of the right temporomandibular joint. In cases of ankylosis, the coronoid process should also be examined carefully. Furthermore, in other to prevent recurrence, aggressive postoperative physiotherapy is required.

CONSENT AND ETHICAL APPROVAL

As per university standard guideline, participant consent and ethical approval have been collected and preserved by the authors.

COMPETING INTERESTS

Authors have declared that no competing interests exist.

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