

Unusual Complete Temporomandibular Joint Bone Ankylosis: a Case Report

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Abstract

Ankylosis of the temporomandibular joint is a clinical entity that not only alters the normal anatomy of the face but also severely hampers mastication. Early diagnosis and treatment are crucial to prevent functional impairments and psychosocial conditions.

Temporomandibular joint (TMJ) ankylosis is characterized by restrictions in movement due to intracapsular fibrous adhesions in capsular ligaments (fibrous ankylosis) and the formation of osseous masses resulting in articular component fusion (osseous ankylosis). Both types of ankylosis are mainly characterized by restrictions and/or limitations in mandibular movement and can originate from trauma, infection or systemic diseases.

A case of unilateral complete temporomandibular joint bone ankylosis with bilateral coronoid elongation was observed in a 26-year-old woman seeking treatment for a facial deformity. A panoramic radiograph revealed an evident morphological abnormality on the left mandibular ramus. Cone-beam CT (CBCT) with 3-D construction was performed to confirm the diagnosis.

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Introduction

Ankylosis is defined as abnormal joint mobility¹. The American Academy of Orofacial Pain (AAOP) defines temporomandibular joint (TMJ) ankylosis as the presence of restrictions in mandibular movement due to intracapsular fibrous adhesions or fibrous changes in the capsular ligaments (fibrous ankylosis) or the formation of osseous masses resulting in an articular component fusion (osseous ankylosis)². Long-standing fibrous ankylosis may progress to osseous ankylosis³. Additionally, ankylosis cases can be classified based on the site (intra articular versus extra articular), the degree of fusion (incomplete versus complete), number of joints affected (unilateral or bilateral), and involved tissues (fibrous or osseous)⁴.

Fibrous or osseous TMJ ankylosis can lead to various problems, such as a significant

reduction in the range of mouth opening, mastication difficulty, facial deformities, periodontal disease, growth disturbances in both jaws, difficulty breathing and sleep apnoea syndrome, especially during development⁵. All the conditions listed above may cause psychological distress^{2,6}. The leading cause of TMJ ankylosis is hemarthrosis secondary to a microtrauma, and the second leading cause is infection. Inflammation and systemic diseases can also be present. Osseous TMJ ankylosis is a rare, chronic and extensive disorder that can occur between the mandibular condyle and fossa, eliminating the available space for the articular disc⁷. Topazian⁸ proposed three stages of osseous ankylosis classification: 1) ankylotic bone limited to the condylar process, 2) ankylotic bone extending to the sigmoid notch and 3) ankylotic bone extending to the coronoid process.

The aim of this article was to present a case of unilateral complete ankylosis that was diagnosed and treated and some uncommon findings, such as a missing reminiscent disc and fibrous tissue between the mandibular condyle and the skull base. Additionally, abnormal transversal angulation of the external wing of the

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pterygoid process was observed on the affected side. Both of the aforementioned features have not been described in the literature before. The authors believe these features should be studied in more detail in cases with the same diagnosis.

Case Report

A 26-year-old woman visited the dental clinic in the Universidad Peruana Cayetano Heredia in the city of Lima-Peru for specialized treatment, and her chief complaints were poor facial aesthetics and the range of mouth opening gradually decreasing since childhood. She had no history of pain or trauma. However, the patient suffered a severe facial infection when she was fifteen years old and therefore underwent surgery at that time. Since then, this is the first time the patient claimed to have experienced a painless limitation in mouth opening. Therefore, she underwent surgery two more times to improve her condition, without successful outcomes. The patient could not recall the specific surgical procedures she underwent.

In the extraoral examination, the patient showed facial asymmetry mainly characterized by a flattening of the lower third of her face, her chin deviated to the right side, and her facial profile was convex. The patient reported that her facial asymmetry progressed over the years. TMJ movements were restricted on both sides, predominantly on the right side. For mouth opening, she could only reach an interincisal distance of 2 mm without pain. The intraoral examination was limited, but poor oral hygiene and dry mucosa were observed. Additionally, lip incompetence was present.

A panoramic X-ray (Fig. 1) showed an evident morphological abnormality on the left mandibular ramus. The mandibular sigmoid notch, glenoid cavity and articular eminence could not be clearly distinguished. A radiolucent band was present in the mandibular ramus, and accentuation of the antegonial notch ("trigger sign") was observed; the superior and inferior cortical walls of the mandibular canal could not be visualized. The presence of the radiolucent band in the middle of the mandibular ramus was considered a radiographic finding, and it was assumed that this feature resulted from one of the surgical procedures the patient underwent during her childhood; the presence of a notch suggested fragment displacement. An increased

osseous density was also observed around this area, which is consistent with the normal bone remodelling process.



Figure 1. Panoramic radiography showing a mandibular condylar morphology abnormality on the left side. The coronoid process is elongated (arrow), and mastoid cells are pneumatized (arrowheads). Additionally, a trigger sign was observed (discontinued arrow).

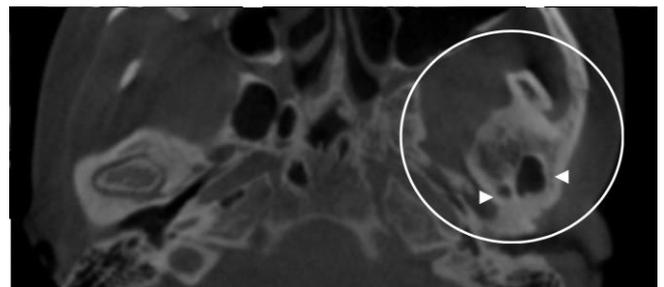


Figure 2. Axial view with CBCT showing no differentiation between the mandibular condyle, glenoid cavity and coronoid base. Pneumatization of the mastoid cells can also be observed.

Cone-beam CT (CBCT) confirmed total osseous fusion and no differentiation between the mandibular condyle, glenoid cavity or coronoid process, (Fig. 2) yielding a single mass. The external wing of the pterygoid process was elongated (Fig. 3) and had a different angulation than did that on the contralateral side.

Additionally, both coronoid processes were elongated (Fig. 4). With CBCT, the extent of the radiolucent band observed in the mandibular body was identified, (Fig. 5) and the presence of a chronic fracture was confirmed. Written informed consent was obtained to perform surgery and publish the results. After surgery, a panoramic radiographic scan was performed (Fig. 6), and the image showed a metal reconstruction plaque on the left mandible side.

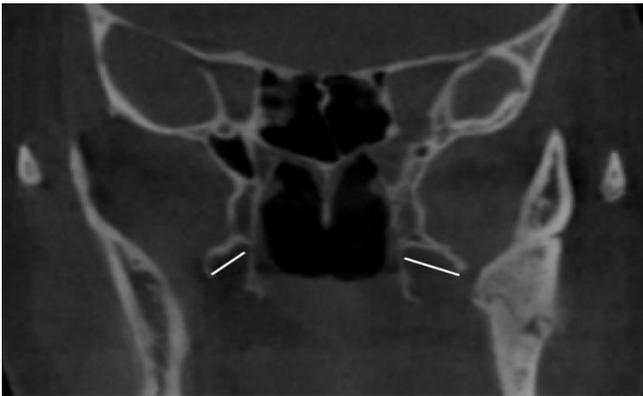


Figure 3. Coronal view with CBCT showing an elongated external wing of the pterygoid process on the left side with a different transversal angulation compared to that on the contralateral side.

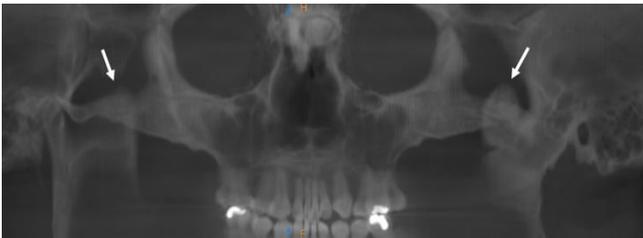


Figure 4. Panoramic reformation with CBCT showing both coronoid processes are elongated (arrows).

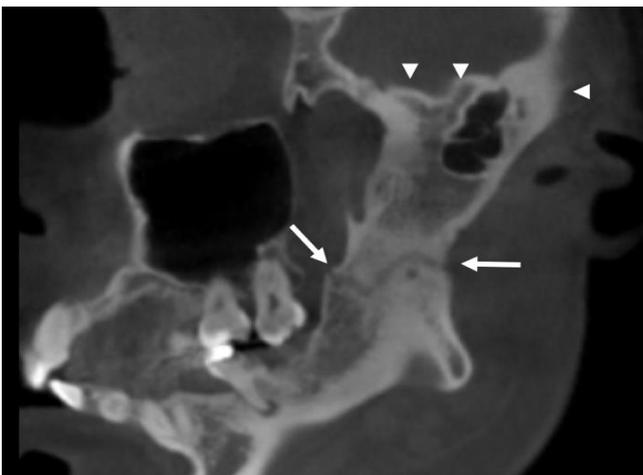


Figure 5. Sagittal view with CBCT demonstrating complete osseous ankylosis (arrowheads) and extension of the radiolucent band in the mandibular body between the posterior and anterior mandibular ramus edge (arrows).

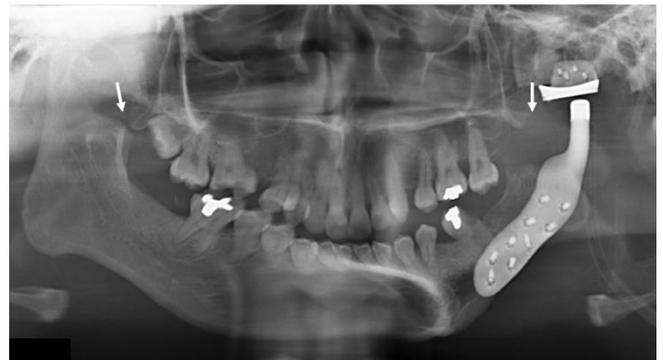


Figure 6. Postoperative panoramic radiograph showing the complete resection of both coronoid processes (arrows); the presence of a metal reconstruction plaque extending from the left mandibular body, angle and ramus; and a prosthetic mandibular condyle.

Discussion

TMJ osseous ankylosis is a rare condition. In the literature, few cases of this condition have been published. In the present case report, the main cause was a severe infection when the patient was a child. Currently, the most prevalent aetiological factor is trauma followed by infections. Poor jaw development can cause dental malocclusions and breathing problems. It can be speculated that the patient presented with breathing problems because of lip incompetence and dry mucosa, which were observed during the clinical examination. Similar to the patient in the present study, most patients who report limitations in mouth opening without pain are aware of this condition for a long time, and some even do not feel that it poses a significant problem¹. There is a direct correlation between severe cases of TMJ disorders and a lower quality of life⁶. However, only a few people experience stress, depression or decreased social quality of life.

In growing children with unilateral TMJ ankylosis associated with facial asymmetry, the hemi-mandible is shorter on the affected side; that on the contralateral side is longer and flattened, resulting in a deviated chin to the ankylosed side as well as retrognathia⁹. A variation of this asymmetry was presented; the affected side was longer, and the chin deviated toward the healthy side. However, this condition can be explained by the fact that the patient underwent surgery and presented a mandibular ramus chronic non-union fracture on the affected side.

Patients with TMJ osseous ankylosis can present pathognomonic radiographic features that facilitate the diagnosis. In general, the mandibular condyle can be fused to the temporal bone. Additionally, a large mass of bone can be observed, involving the mandibular condyle, the temporal bone and the zygomatic process (complete). In most cases, a radiolucent area is observed inside the bone mass, which indicates a reminiscent articular disc or fibrous tissue¹⁰. In the present case, complete fusion with the cranial base was observed without a radiolucent area inside. This feature has not been observed before in another case report.

The coronoid process is often elongated in patients with TMJ ankylosis. It is associated with reduced mouth opening, so it needs to be surgically resected and discharged.^[7] In the present case, complete osseous ankylosis was accompanied by bilateral coronoid elongation. This feature has been reported before in other studies and case reports. Guerrero *et al.*¹¹ showed that patients diagnosed with temporomandibular disorders (TMD) have a larger external wing of the pterygoid process than do patients without TMD disorders. The authors also found this feature on the affected side with the "concomitant angle variation" described before. When a restricted mandible condyle translation is present, as seen in cases of TMJ ankylosis, the temporalis muscle becomes hyperactive, and then the coronoid process elongates as a reactive response. TMJ-related muscles suffer from tensional changes that end in an osseous remodelling process, leading to structural changes at the insertion points¹². Articular disc displacement can cause long-term lateral pterygoid muscle spasm, especially on the superior fascicle, which inserts in the external wing of the pterygoid process and in the anteromedial cervical side of the mandibular condyle. Mandibular and maxillary bone growth can lead to hyperplasia, and hypoplasia occurs when the pterygoid process is the only altered part of the sphenoid bone¹³.

Osseous abnormalities can be observed on conventional radiographs. However, new techniques and devices have been developed to obtain better images, visualize overlapping anatomical structures and prevent image distortion. CBCT provides a large amount of information about TMJ osseous morphology, allowing the condylar surface and its volume to

be quantified exactly, with limited surrounding soft tissue information. Compared with magnetic resonance imaging (MRI), CBCT provides more information for the visualization of bone alterations present in TMD, such as flattening, osteophytes or erosion. CBCT has a lower spatial resolution and larger slice thickness (3 mm) than does MRI, allowing better visualization of osseous tissues. Additionally, MRI may include air spaces within the temporal bone, fibrous tissues within the TMJ and the surrounding areas of the lateral pterygoid muscle joined to the condylar articular surface¹⁴.

Other surgical treatment options include autogenous condylar grafts and personalized alloplastic prostheses for more severe cases⁵. The surgeons in this study used a metallic reconstruction plaque due to the financial limitations of the patient. Furthermore, current developments in navigation surgery can also deliver high-quality treatment outcomes, making surgeries more predictable and safer¹⁵.

In the present study, well-known radiographic features of osseous TMJ ankylosis and other atypical characteristics are presented to gain a better understanding of potential types of TMJ osseous ankylosis.

Conclusions

Since TMJ ankylosis can remain unnoticed for a long time primarily due to the absence of pain, an early diagnosis and intervention is mandatory to prevent major facial abnormalities. CBCT is a very important diagnostic tool for TMD diagnoses, mainly for those related to osseous pathologies, developmental disorders and trauma.

Declaration of Interest

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