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Temporomandibular Joint Ankylosis: An Epidemiological Study in Marrakesh

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Abstract Temporomandibular joint (TMJ) ankylosis is a disabling disease where the main clinical feature is limited mouth opening. Aim: Our study's aim is to draw epidemiological, clinical and imaging aspects of this disease in Marrakesh. Patients and methods: retrospective study gathering all TMJ ankylosis cases operated between 2008 and 2016 at maxillo-facial surgery department in University Hospital Mohamed VI of Marrakech. Results: a total of 27 cases were included, the mean age was 22,9 years (3-55 years). The sexe-ratio was 0,5. Unilateral cases (81,5%) were more common than bilateral. The most frequent aetiology was trauma (85,2%) followed by infection (7,4%) and one case of ankylosing spondylitis (3,7%). Mouth opening less than 10mm was observed in 67%. According to Topazian Classification, Stage I was predominant (59%). Conclusion: TMJ ankylosis is frequent in our context, especially due to neglected condylar trauma so the best treatment remains the prevention based on early diagnosis and proper management of TMJ trauma.

Keywords: *temporomandibular joint, ankylosis, epidemiology*

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1. Introduction

Ankylosis is a Greek terminology meaning "stiff joint". Temporomandibular joint ankylosis refers to an obliteration of the joint space with abnormal bone morphology leading to fusion with opposite joint components. [1]

It results in limited mouth opening and can be an extremely disabling condition from the view point of function, hygiene and cosmetic appearance. [2]

When it occurs in a child, it can have devastating effects on the future growth and development of the jaws and teeth. Furthermore, in many cases, it has a profoundly negative influence on the psychosocial development of the patient because of the obvious facial deformity. [3]

TMJ ankylosis is most commonly linked to trauma (13-100%), infection (10-49%), and systemic diseases like anklosing spondylitis, rheumatoid arthritis and psoriasis (10%). [4]

The incidence of TMJ ankylosis is declining in Europe and North America, partly as a result of better, earlier management of condylar fractures and partly because of the use of antibiotics which have reduced persistence and recurrence of infection. TMJ ankylosis is relatively common in developing countries mainly resulting from childhood trauma. [5] A condylar trauma is either ignored by the parents or mismanaged in hospitals.

The diagnosis of temporomandibular ankylosis is made on the history of long-standing inability to open the mouth following injury or an inflammatory process in the joint [6], and the preoperative evaluation usually consists of physical and radiographic examination.[7] Current methods include panoramic radio-

graphy, and Computed tomography (CT).[8] Recently, the value of three-dimensional CT (3D-CT) prior to surgery has been advocated. [7,9,10]

The treatment of TMJ ankylosis, based on surgery along with physiotherapy, is a significant challenge because of technical difficulties and high incidence of recurrence. [11]

The purpose of this study was to draw epidemiological (frequency, age, sex, etiology) clinical and imaging aspects of TMJ ankylosis in Marrakesh.

2. Patients and Methods

This retrospective descriptive study was carried out at the department of Oral & Maxillofacial Surgery at the university hospital of Marrakesh, between 2008 and 2016. Twenty seven patients with limited mouth opening and radiographically proven TMJ ankylosis were included in this study.

Demographic information like name, age and gender were recorded. Patients were questioned concerning any history of fall, road traffic accident, birth trauma, local or systemic infection, degenerative joint diseases and joint surgery. Type of symptoms as well as duration of the joint ankylosis was also noted. Both intra oral and extra oral

examination of the patient was performed and relative findings regarding patient general nutrition, mouth opening, chin deviation, facial deformity, and occlusal relationship were recorded.

Radiographic examination of the temporomandibular joint was done in all patients including an orthopanthomogram and CT scan to aid reaching a definite diagnosis and determining the severity according to Topazian classification as follows:

- Stage I: Ankylotic bone limited to the condylar process.
- Stage II: Ankylotic bone extending to the sigmoid notch.
- Stage III: Ankylotic bone extending to the coronoid process.

The patient was diagnosed with either Unilateral or Bilateral TMJ ankylosis.

3. Results

We gathered 27 cases over 9 years. Females were predominant with 66,6% (18 cases); the male to female ratio was 1:2. The mean age was 22,9 years [range 3-55 years] and the most frequent group was from 21 to 30 years as shown on (Figure 1). Etiologically, trauma was reported in 88,8% (24 cases) essentially resulting from chin trauma during falls (15 cases) while 8 cases were due to road traffic accidents and 1 case suffering from epilepsy with recurrent injuries during seizures. Infection was found in 2 cases (1 otitis media and 1 septicemic state). One case was related to ankylosing spondylitis. The etiological factor majorly occurred in the first decade (48%) (Figure 2), and the duration of symptoms ranged from 1 to 16 years with a mean of 5,7 years.

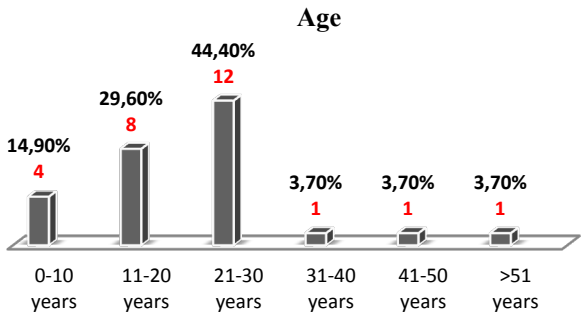

Figure 1. Distribution of temporomandibular joint ankylosis among different age groups

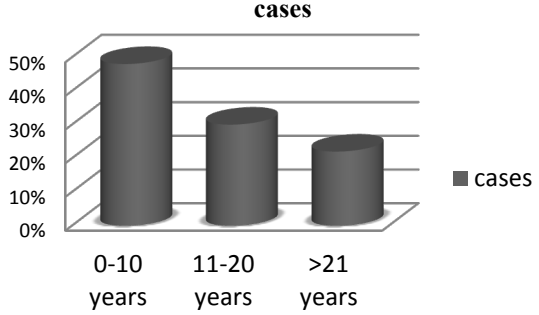

Figure 2. Age at occurrence of etiological factor

26% of cases showed malnutrition signs. Deformity concerned 63% (17 cases) including 9 cases of chin deviation, 5 cases of retrognathism and 3 cases had the typical bird face and pre-auricular bulging was found in 26% of cases. (Figure 3) Severe forms were predominant as a restricted mouth opening less than 10mm was reported in 67%, Figure 4 shows patients' distribution following the mouth opening degree. (Figure 5). 85% of patients had poor oral hygiene with multiple caries. (Figure 6)

Figure 3. Bird face deformity in bilateral TMJ ankylosis

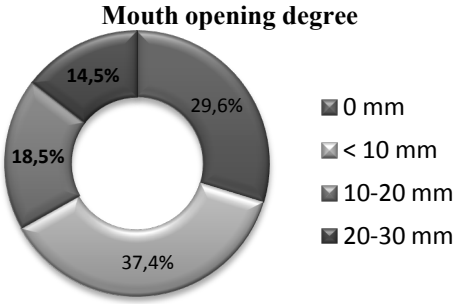

Figure 4. Patient's distribution following mouth opening degree

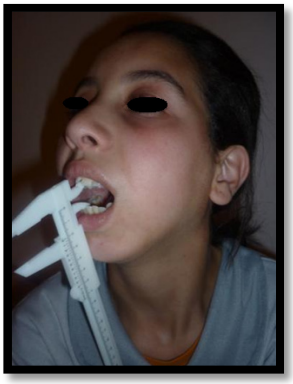

Figure 5. Interincisal distance measured by a caliper showing a restricted mouth opening

Figure 6. Unilateral severe TMJ ankylosis associated to poor mouth hygiene with multiple caries

Among the study unilateral cases were predominant with 81,5% (22 cases). Based on CT-scan, stage I ankylosis comprised the highest proportion (59%) followed by stage II (26%) then stage III (15%) (Figure 7).

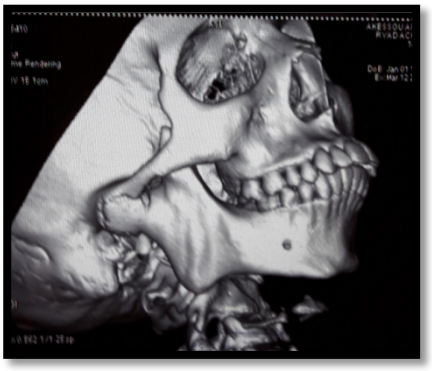

Figure 7. 3D CT-scan showing a unilateral stage I ankylosis associated to hypertrophic coronoid process

4. Discussion

This affliction was known since ancient Greece, at the time of Hippocrates who proposed teeth extraction to facilitate feeding [12], and up to now, it's management remains challenging for the surgeon.

TMJ ankylosis can be classified according to:

Location: Kazanjian [13] divided it into two groups, intra articular or "true" ankylosis in which the ankylosing factor lies in the joint proper resulting in an adhesion of mandibular condyle to glenoid fossa, and extra articular or false ankylosis that is due to lesions involving extra articular structures such as hypertrophic coronoid process, Jacob's disease, depressed fractures of zygomatic arch, myositis ossificans, postirridiation and burn scaring.

- Site of involvement: unilateral or bilateral forms.
- Extent of fusion: we distinguish complete and incomplete ankylosis. In the first a continuous bony bloc unites the ramus to glenoid fossa, its thickness frequently reaches 3cm according to Dufourmentel. [12,14] While in the second, a part of the articular surface remains undamaged.
- Type of tissue involved: fibrous, fibro-osseous or osseous [5].
- Radiographic findings: many classifications have been proposed over the decades: Topazian [15] (1966), largely accepted, including 3 stages; Sawhney [16] (1986) involving IV classes; recently ElHakima [17] (2002) described a 4 stages classification following the relation of the ankylotic mass to the surrounding vital structures especially the base of the skull.

Our study confirms that TMJ ankylosis is a frequent condition in developing countries, and the largest samples have been recorded in Egypt and India. [18]

It's found a variability concerning sex ratio from a study to an other, but majorly a female predominance is noticed [5,19,20], which could be due to hormonal differentiation between boys and girls or could be related to the differentiation in the anatomy of the neck of the condyle [21].

Studies stated that TMJ ankylosis is commonly seen in children and young adults [22], the rich vascularized lamellar bony structures in adolescents and young adults with greater growth and reparative potential are more prone to developing ankylosis after TMJ trauma. [23]

Among our study, the highest proportion was in the second decade, this delay is especially due to difficulties to access hospitals and to lack of awareness about this pathology.

Frequently, because of the slow evolution of ankylosis, the direct connection between cause and effect is unknown to the patient and his family [13].

Even thought, we could find that trauma was the leading cause with 88,8% , followed by infection either local or systemic, then rheumatic disease. Our results concord with a great proportion of studies all over the world [5,18,19,20,21,22]. In the pre-antibiotic era, infection was the most common cause of ankylosis 18, this remains true for some developing countries mostly because of Noma. [23]

The high incidence of post-traumatic TMJ ankylosis in our context is explained by neglected condylar fractures, fear of pain and consequent prolonged immobilization, or mismanaged condylar trauma.

For Hegab [24], trauma leads to three main events:

1. Disc displacement with severe injury to the articular surfaces with formation of intra-articular hematoma rich in osteoprogenitor cells
2. Change in local tissue environment includes disturbances involving the microvasculature and changes in oxygen tension, pH (Alkaline) and blood flow. These local changes of the tissue produce a milieu that supports osteogenesis.
3. Trauma signal lead to local release of osteoinductive cytokines especially bone morphogenic proteins (BMP) that synthesize osteoid and chondroid. These lead to differentiation of the osteoprogenitor cells into osteoblasts or chondroblasts start lay down of new bone.

Currently, new hypothesis have been advanced such as extracapsular hematoma, distraction osteogenesis, genetic predisposition, hypertrophic nonunion and hypercoagulable state of blood. [25]

During infection or rheumatic disease, TMJ ankylosis occurs following damages to articular surfaces and lesions of the disc which make bony edges in contact. [12,26]

It's found in the literature that the etiological factor usually occurs in children under 10 years [5,13,27,28] which concord with our study's results.

TMJ ankylosis is a severely debilitating condition. It effects mastication, digestion, speech and facial appearance and can cause long-term psychological problems. It's characterized by a progressive mouth opening restriction which was mostly severe in our sample.

The degree of deformity depends upon two factors [29]:

- Age of onset: it's more marked when it occurs during 15 first years of life. [13]
- Unilateral or bilateral involvement. For the unilateral forms, chin is deviated to the effected side and posteriorly displaced. Body and ramus of the mandible is short and there is prominent and high antegonial notch on the effected side.Intra orally dento-alveolar segment is adapted to bring teeth in

functional position, however, teeth usually remain in cross bite on the effected side. In bilateral forms, the chin is posteriorly displaced and under developed, which produces double chin effect (birds deformity), while mandible, incisors and floor of mouth are anteriorly inclined.

Once the diagnosis is suspected, a radiographic examination is needed to confirm and classify the ankylosis in order to propose and adequate management. Nowadays, Ct-Scan is the best imaging modality for planning surgery as it displays the anatomical relationship between the ankylosed segment and the surrounding vital structures [17], permit the measurement of coronoid length helping in the diagnosis of coronoid elongation, measurement of ankylosis mass helping in estimation of amounts of autogenous and alloplastic material required for gap arthroplasty, gives a detailed knowledge of the deformity [30].

Furthermore, according to Majidi it indicates, in some cases, the etiology of the ankylosis.

5. Conclusion

TMJ ankylosis is frequent in our context, especially due to neglected condylar trauma so the best treatment remains the prevention based on early diagnosis and proper management of TMJ trauma. Besides, an Early detection and early intervention to release the ankylosed joint minimizes the severity of the restriction of facial growth and reassure a quasi normal life to the patient.

Conflicts of Interest

No conflict of interest.

Contributions of the Authors

All the authors contributed to the medical care of the patient, as well as the writing this article they approved.

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