

Aetiology and presentation of ankylosis of the temporomandibular joint: report of 23 cases from Abuja, Nigeria

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Abstract

Several studies have reviewed the management of ankylosis of the temporomandibular joint (TMJ), but only a few focused on the aetiology and clinical features. We retrospectively studied the aetiology and clinical features of patients with ankylosis of the TMJ who presented to the Maxillofacial Unit, National Hospital, Abuja, Nigeria, between 2004 and 2009. There were 13 male and 10 female patients, M:F ratio 1.3:1, age range 6–62, mean (SD) 20 (13) years. The aetiological factors were trauma ($n=11$) that comprised falls ($n=6$), untreated fractures of the zygomatic arch ($n=4$) and myositis ossificans ($n=1$); infection ($n=9$), that comprised cancrum oris ($n=3$) and ear infection ($n=6$); congenital or unknown ($n=2$), and coronoid hyperplasia ($n=1$). The maximum interincisal distance at presentation ranged from 0 to 25 mm (mean (SD) 6.7 (7.2) mm). Seventeen had facial deformities. The diagnoses recorded were as follows: left extracapsular ankylosis, ($n=8$); right intracapsular bony ankylosis, ($n=6$); left intracapsular bony ankylosis, ($n=4$); bilateral intracapsular bony ankylosis, ($n=4$), and bilateral intracapsular fibrous ankylosis ($n=1$). Extreme poverty was the main predisposing factor. There is a need for a concerted effort among healthcare providers, policy makers, and the world in general to eradicate poverty and improve healthcare to limit the incidence of ankylosis of the TMJ.

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Introduction

Ankylosis is the Greek for “stiff joint”.¹ The term “ankylosis of the temporomandibular joint” (TMJ) refers to bony or fibrous adhesion of the anatomical components of the joint and their ensuing loss of function.² The adhesion could be between the condylar head of the mandible and the glenoid

fossa of the temporal bone, or between any tissue component of the lower jaw (hard and soft) and the maxilla, zygoma, or the base of the skull. Ankylosis is a disabling condition that causes problems with mastication, digestion, speech, appearance, and oral hygiene.³

Ankyloses of the TMJ can be proper (true) or pseudo (false).⁴ In the first type the damage is caused intra-articularly by adhesion of the mandibular condyle to the glenoid fossa. In the second type, hypomobility of the joint results from extra-articular causes that could follow hyperplasia of the coronoid process; untreated fracture of the zygomatic arch or operation; radiation-induced fibrosis of the temporal muscle,

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or neurological or psychological disorders.^{2,4} Functionally, mandibular mobility (opening, lateral, and protrusive) is restricted in both types.⁴ Ankylosis of the TMJ may be classified by a combination of location (intra-articular or extra-articular), type of tissue involved (bony, fibrous, or fibro-osseous) extent of fusion (complete or incomplete), and side of location (unilateral, bilateral).³

It is most commonly associated with trauma (13–100%), local or systemic infection (0–53%), or systemic disease such as ankylosing spondylitis, rheumatoid arthritis, or psoriasis.^{3,5} Rarely is it congenital,^{6,7} and the cause could be unknown.⁸ As a result of better understanding of the management of condylar fractures, and also the reduced incidence of middle ear infections since the introduction of antibiotics, the incidence is decreasing in the west, but it is still high in Asia and Africa (Ajike S. Temporomandibular joint ankylosis: analysis of 65 cases. Paper presented at 17th International Conference on Oral and Maxillofacial Surgery, 2005).⁹

In Nigeria it has been identified as a complication of noma, which is a spreading gangrene that invades the facial tissues.¹⁰ Pathogenetically, noma is the result of a high bacterial load of normal micro-organisms in the mouth that breaks down the resistance of a failing immune system. This leads to a spreading gangrene that quickly invades the facial tissues and leads to severe facial disfigurement, ankylosis of the TMJ, oral incontinence, and problems with speech.¹¹ It is an escalating public health problem among impoverished children in the developing world, particularly in sub-Saharan Africa where the incidence in some countries is estimated to be 12/1000 cases in the most affected communities.^{12,13}

Ankylosis of the TMJ usually develops before the age of 10,¹ but could be found at any age, the usual range being 20–30 years.^{2,3} It is slightly commoner in boys than girls in a ratio of 1.4:1.2.⁹ Unilateral ankylosis has been reported to be commoner than bilateral, the ratio being 1.5:1.^{2,3,14} Patients present with limitation of mouth opening and a maximum interincisal distance of between 0 and 20 mm.³ It causes aesthetic defects to the face, malocclusion, inability to enjoy eating, and malnutrition, particularly when it develops in childhood.⁵

Many studies have reviewed various aspects of ankylosis of the TMJ, particularly the management, but few have focussed on the aetiology and clinical features. For the purposes of prevention and management we must examine these in our environment.

Patients and methods

We retrospectively studied patients with ankylosis of the TMJ who were treated at the Oral and Maxillofacial Surgery Unit, National Hospital, Abuja, Nigeria between 2004 and 2009. The national hospital is a new centre in the federal capital territory that caters for a population of about 15 million. Information collected from casenotes included patients' age,

Table 1
Details of 23 patients with ankylosis of the temporomandibular joint.

Variable	Number
Sex	
Male	13
Female	10
Mean (SD) age (years)	20 (13)
Range	6–62
Social class	
Low	19
Intermediate	2
High	2

Table 2
Aetiology of ankylosis of the temporomandibular joint in 23 patients.

Aetiological factor	Number
Fall	6*
Infection of the ear	6
Untreated maxillofacial injury	5**
Cancrum oris	3***
Congenital/unknown	2
Coronoid hyperplasia	1

* Outdoor activities ($n = 3$), accident at home ($n = 2$), and complication of a cerebrovascular accident ($n = 1$).

** Malunited fracture of zygomatic arch ($n = 4$) and myositis ossificans after haematoma ($n = 1$).

*** Although infected between the ages of 3 and 6 years, they presented between the ages of 26 and 28.

sex, specific aetiology, and clinical and radiological features. Data were analysed with the help of the Statistical Package for the Social Sciences (SPSS version 15.0, Chicago, Ill, USA). Data are given as incidence, and mean (SD).

Results

Twenty-three patients were seen between 2004 and 2009 and their details are shown in Table 1. The aetiological factors are shown in Table 2, the presenting complaints in Table 3, types of facial deformity in Table 4, and the types of ankylosis in Table 5.

The midline of the mandible was shifted towards the affected side in all 10 cases of unilateral intracapsular ankylosis and one case of left extracapsular ankylosis caused by cancrum oris. No shift was recorded in any of the 5 cases of bilateral ankylosis (Fig. 1) or in 7 cases of extracapsular ankylosis.

Table 3
Presenting complaints in 23 patients with ankylosis of the temporomandibular joint.

Complaint	Number
Inability to open mouth	15
Limited mouth opening	4
Snoring	2
Brought to hospital by staff	2

Table 4
Relation between age at exposure to injury and facial deformity in 23 patients with ankylosis of the temporomandibular joint.

Age (years) at exposure	Slight asymmetry (n=3)	Obvious asymmetry (n=7)	Asymmetry with scar (n=3)	Birdface deformity (n=4)
<1	2	6	0	2
3	1	0	3	2
5	0	1	0	0

Six patients had no signs of asymmetry.

Table 5
Types of ankylosis of the temporomandibular joint in 23 patients.

Type	Number
Left extracapsular	8
Right intracapsular bony	6
Left intracapsular bony	4
Bilateral intracapsular bony	4
Bilateral intracapsular fibrous	1

All 10 cases of unilateral intracapsular ankylosis had fullness of the cheek on the affected side and flattening on the normal side (Fig. 2). The 4 cases of bilateral intracapsular bony ankylosis presented with mandibular retrognathism described as “birdface” deformity (Fig. 3), and the 3 cases caused by cancrum oris presented with telltale scars (Fig. 4).

The maximum interincisal distance at presentation ranged from 0 to 25 mm (mean (SD) 7 (7) mm). Poor oral hygiene was recorded in 10 cases, fair in 9, and good in 4.

Orthopantomographic and transcranial views of the TMJ (open and closed) were used for diagnosis in 10 cases, and occipitontal and transcranial views (open and closed) in 5. Computed tomography was used in 2 cases and radiological records were missing in 2. Radiological reports described deformity of condylar heads and varying degrees of obliteration of the joint space in 15 cases, mandibulomaxillary fusion in 2, and displaced fractures of the zygomatic complex in 4.



Fig. 1. Bilateral intracapsular ankylosis: maximum interincisal distance of 0, no shift in the mandibular midline, and poor oral hygiene (published with the patient's permission).



Fig. 2. Left unilateral intracapsular ankylosis showing fullness on the left and flattening on the right (published with the patient's permission).



Fig. 3. “Bird face” deformity in bilateral intracapsular ankylosis (published with the patient's permission).

Discussion

Trauma is the commonest cause of ankylosis of the TMJ, the incidence of which ranges between 13% and 100%.^{2,15}



Fig. 4. Extracapsular ankylosis with tell-tale scar caused by cancrum oris (published with the patient's permission).

However, most reports lack details of what constitutes trauma as the cause of ankylosis. In the present study, it was the aetiological factor in 48% of cases, which comprised 26% of falls, and 22% of complications of untreated maxillofacial injuries. Miyamoto et al.¹⁶ concluded from an animal study that intracapsular condylar fracture increased the risk of ankylosis of the TMJ. Arthroscopic and magnetic resonance imaging studies of the effect of trauma on intracapsular structures have shown that damage to the articular surface, rupture of the disc, and intra-articular bone fragments, are factors that hasten ankylosis of the TMJ.^{17,18} However, intracapsular haematoma alone without intracapsular structural damage does not result in ankylosis.¹⁸ In this study, intra-articular structural damage as a result of indirect injury from impact to the symphysis was thought to be the cause of the ankylosis.

Untreated fractures of the zygomatic arch have been given little attention in scientific papers as a cause of extracapsular ankylosis. A displaced fracture impinges on the coronoid process, limiting its movement. The combination of poverty, ignorance, and sometimes lack of early access to treatment has resulted in delayed treatment of these fractures, with resulting extracapsular ankylosis. The health facilities available in the country are not adequate to cater for the population, which results in people having to travel some distance to them. Where the facilities are available, settlement of bills becomes a problem because of poverty. The country still operates to a large extent a “pay out of pocket” system to settle medical bills. A health insurance system was introduced recently but less than 10% of the population is covered. The resulting effect of these factors is delayed access to treatment that results in ankylosis of the TMJ.

Our report that 39% of ankylosis is the result of infection falls within the range of 0–53% reported earlier.³ Septic arthritis of the TMJ is rare but pathological microorganisms such as staphylococci can invade the joint from an adjacent middle ear infection or a distant nidus by the haematogenous route.^{19,20} An infective process, if undiagnosed or not adequately treated, may result in erosion of the articular surface and consequent fibrous or bony ankylosis.^{2,21}

The persistence of conditions such as ankylosis of the TMJ in Nigeria and other developing countries seems to be the result of many factors, all associated with poverty. Marck¹⁰ stated that noma had gradually disappeared in the western world by the end of nineteenth century as a result of economic progress, which gave the poorest in society the opportunity to feed their children adequately. However, noma and other childhood infections are still persistent in the developing countries as a result of extreme poverty.^{10,11,13,22} In a review of 69 Nigerian children who developed noma, there was a distinct history of recent debilitating infections, with measles being the most common, affecting 30 (43%) of the children.²³ Other infections reported in the 69 children included malaria (30%), chicken pox (9%), tuberculosis, and others (17%). The location of our study is a city centre, and may not be a true reflection of the situation in the country districts. Several

reports from Sokoto, a city in the north of the country, painted a picture of extreme poverty.^{10,11,22}

The diagnosis of congenital ankylosis is controversial. True congenital ankylosis of the TMJ is immobility of the joint from birth in the absence of other likely causes such as birth trauma.²³ The exact pathogenesis of congenital ankylosis is not known, but proposed hypotheses include anomalous fusion of embryonal mesenchyme in early development during embryogenesis, an abnormality of the stapedia artery in the fetus, hypervitaminosis A, and early loss of neural crest cells.^{6,7} In cases of ankylosis caused by obstetric trauma, mouth opening reduces gradually in the neonatal period as a result of fibrosis, and such cases when recognised later have been erroneously termed congenital ankylosis.⁶ It has been stated that ankylosis of unspecified origin, and some congenital forms of it, are to a great extent the result of unrecognised or undiagnosed septic arthritis.²¹ We were unable to find out whether our patients with congenital ankylosis (9%) who presented after 5 years of age truly had congenital ankylosis or whether the cause was septic arthritis, but they gave no history of obstetric trauma.

The age of the patient when exposed to injury seems to be a major factor in the assessment of maxillofacial deformities in ankylosis of the TMJ (Table 4). Childhood exposure results in defective osteogenesis of the cartilage damaged by the ankylosis process and also in loss of muscular guidance over the mandibular growth process,² consequently patients present with pervasive maxillofacial alterations such as unilateral intracapsular ankylosis (Fig. 2). In these cases, uninterrupted development of the unaffected side leads to lengthening and shifting of the mandibular midline to the affected side. In bilateral cases, defective osteogenesis leads to mandibular hypoplasia presenting as “bird face” deformity (Fig. 3). In the present study, all patients who presented with severe facial deformity were exposed to injury when they were less than 5 years old.

Ankylosis of the TMJ is distressing because it compromises function and the facial deformity has an adverse psychological effect on the child. There is a need for a concerted effort among healthcare providers to institute preventive measures including adequate treatment of infections and early treatment of maxillofacial injuries. The only truly effective way to prevent “the face of poverty” is to improve the economic position of the poorest of the poor throughout the world.

References

1. Malik NA. *Textbook of oral and maxillofacial surgery*. New Delhi: Jaypee Brothers Medical Publishers (P) Ltd.; 2002. pp. 207–218.
2. Valentini V, Vetrano S, Agrillo A, Torroni A, Fabriani F, Ianetti G. Surgical treatment of TMJ ankylosis: our experience (60 cases). *J Craniofac Surg* 2002;1:59–67.
3. Vasconcelos BC, Bessa-Nogueira RV, Cypriano RV. Treatment of temporomandibular joint ankylosis by gap arthroplasty. *Med Oral Patol Oral Cir Bucal* 2006;11:E66–9.

4. Spijkervet FKL, de Bont LGM, Boering G. Management of pseudoankylosis of the temporomandibular joint: report of cases. *J Oral Maxillofac Surg* 1994;**52**:1211–7.
5. McFadden LR, Rishiraj B. Treatment of temporomandibular joint ankylosis: a case report. *J Can Dent Assoc* 2001;**67**:659–63.
6. Ajike SO, Chom ND, Amanyeiwe UE, et al. Non-syndromal, true congenital ankylosis of the temporo-mandibular joint: a case report. *West Ind Med J* 2006;**55**:444–6.
7. Nwoku AL, Kekere-Ekun TA. Congenital ankylosis of the mandible. Report of a case noted at birth. *J Maxillofac Surg* 1986;**14**:150–2.
8. Manganello-Souza LC, Mariani PB. Temporomandibular joint ankylosis: report of 14 cases. *Int J Oral Maxillofac Surg* 2003;**32**:24–9.
9. Shashikiran ND, Reddy SV, Patil R, Yavagal G. Management of temporo-mandibular joint ankylosis in growing children. *J Ind Soc Pedod Prev Dent* 2005;**23**:35–7.
10. Marck KW. A history of noma, the “Face of Poverty”. *Plast Reconstr Surg* 2003;**111**:1702–7.
11. Enwonwu CO, Falkler Jr WA, Idigbe EO, et al. Pathogenesis of cancrum oris (noma): confounding interactions of malnutrition with infection. *Am J Trop Med Hyg* 1999;**60**:223–32.
12. Enwonwu CO. Noma: a neglected scourge of children in sub-Saharan Africa. *Bull World Health Organ* 1995;**73**:541–5.
13. Barmes DE, Enwonwu CO, Leclercq M-H, Bourgeois D, Falkler WA. The need for action against oro-facial gangrene (noma). *Trop Med Int Health* 1997;**2**:1111–4.
14. El-Hakim IE, Metwalli SA. Imaging of temporomandibular joint ankylosis. A new radiographic classification. *Dentomaxillofac Radiol* 2002;**31**:19–23.
15. Miyamoto H, Kurita K, Ishimaru JI, Goss AN. Ankylosis of the temporomandibular joint: literature review, case report and sheep model. *Aichi-Gakuin Dental Sci* 1999;**12**:53–64.
16. Miyamoto H, Kurita K, Ogi N, Ishimaru JI, Goss AN. The effect of an intra-articular bone fragment in the genesis of temporomandibular joint ankylosis. *Int J Oral Maxillofac Surg* 2000;**29**:290–5.
17. Goss AN, Bosanquet AG. The arthroscopic appearance of acute temporomandibular joint trauma. *J Oral Maxillofac Surg* 1990;**48**:780–4.
18. Sullivan SM, Banghart PR, Anderson Q. Magnetic resonance imaging assessment of acute soft tissue injuries to the temporomandibular joint. *J Oral Maxillofac Surg* 1995;**53**:763–7.
19. Markowitz HA, Gerry RG. Temporomandibular joint disease. *Oral Surg Oral Med Oral Pathol* 1950;**3**:75–117.
20. Wright V. Arthritis associated with venereal disease. A comparative study of gonococcal arthritis and Reiter’s syndrome. *Ann Rheum Dis* 1963;**22**:77–90.
21. Leighty M, Spach DH, Myall RWT, Burns JL. Septic arthritis of the temporomandibular joint: review of the literature and report of two cases in children. *Int J Oral Maxillofac Surg* 1993;**22**:292–7.
22. Enwonwu CO. Epidemiological and biochemical studies of necrotizing ulcerative gingivitis and noma (cancrum oris) in Nigerian children. *Arch Oral Biol* 1972;**17**:1357–71.
23. el-Mofty S. Ankylosis of the temporomandibular joint. *Oral Surg Oral Med Oral Pathol* 1972;**33**:650–60.