Temporomandibular Joint Ankylosis with Maxillary Extension: Proposal for Modification of Sawhney's Classification

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Abstract

Temporomandibular joint (TMJ) is a unique joint in which both jaws must open synchronously for function. Any pathology in one or both joints results in functional problems with associated poor quality of life. TMJ ankylosis (TMJA) is a joint pathology as a result of bony and/or fibrous adhesion of the joint apparatus, resulting in partial or total loss of function. This is a retrospective study from two tertiary referral centers in northwest region of Nigeria from 2012 to 2016. Data retrieved include gender, age, etiology of ankylosis, duration of ankylosis, laterality of ankylosis, type of imaging technique, type of airway management, types of incision, surgical procedure, interpositional materials used, and complications. Data were analyzed using SPSS for Window version 20.0 (IBM Corp.). Results were presented as simple frequencies and descriptive statistics. A total of 36 patients with TMJA were seen during the study period; out of which 7 (19.4%) patients had maxillary extension of the ankylotic mass. There was a male: female ratio of 1.3:1. Four (57.1%) patients were within the age group between 5 and 10 years, two (28.6%) within the age group between 11 and 15 years, while only one (14.3%) was within the age group between 31 and 35 years. All the cases (7 [100%]) of maxillary extension were secondary to cancrum oris (noma). Cheek scarring as a result of management of cancrum oris was observed. In addition, intraoral fibrosis eliminating the upper and lower buccal sulci extending to the molar regions was also noted. With the involvement of the maxilla in the ankylotic mass, the authors have proposed modification of Sawhney's classification by the addition of Class V. The authors have suggested a name for the new classification to be "Modified Sawhney's Classification of Temporomandibular Joint Ankylosis". Aggressive postoperative physiotherapy for a sufficient period of time (minimum of 6 months) is paramount.

Keywords

- ankylosis
- Bramley–Alkayat
- condylectomy
- gap arthroplasty
- postrami
- temporomandibular joint

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The temporomandibular joint (TMJ) is a unique joint in the body because it is the only joint where both the right and left sides must move synchronously. It is also the only joint that is lined by fibrocartilage. Similarly, it is also the only joint in which factors remote to the joint can affect the joint function, e.g., the teeth in occlusion, and it is the only joint in which emotional status of the patient affects leading to TMJ pain dysfunction syndrome. TMJ ankylosis is a joint pathology as a result of bony and/or fibrous adhesion of the joint apparatus, resulting in partial or total loss of function.[1,2] Since the TMJ is a unique joint in which both jaws must open synchronously for function, any pathology in one or both joints results in functional problems with associated poor quality of life. Etiologies of TMJ ankolysis range from trauma (e.g., forceps delivery and[3] fall on the chin[4]), infection (e.g., middle ear infection and cancrum oris [noma]), and ankylosing spondylitis.[5] Several classifications have been proposed in the classification of TMJ ankylosis in order to have a picture of surgical difficulty.[6] It has been classified simply as true or false ankylosis. The true ankylosis is further classified by Sawhney[7] into types I, II, III, and IV.

Type I occurs where the condyle is medially angulated and associated with a deformed articular fossa together with a mild-to-moderate amount of new bone formation. Type II is found where there is no recognizable condyle or fossa, but instead a large mass of new bone extending from the ramus to the base of the skull. Type III ankylosis usually results from a medially displaced fracture dislocation with bone bridging the mandibular ramus to the zygomatic arch, while type IV is found when the joint architecture is replaced completely by bone with fusion of the condyle, sigmoid notch, and coronoid process to the zygomatic arch and glenoid fossa (►**Fig. 1**).[8] The authors have discovered this classification to be inadequate based on our series with the ankylotic mass involving the maxilla.

The main aim of this study, therefore, was to present case series of TMJ ankylosis with maxillary involvement in a resource scarce population and to propose a modification of Sawhney's classification by the addition of type V (when the joint architecture is replaced completely by bone with fusion of the condyle, sigmoid notch, and coronoid process to the zygomatic arch, glenoid fossa, and maxilla).

Case Series

This is a retrospective study from two tertiary referral centers (Usmanu Danfodiyo University Teaching Hospital, Sokoto, Sokoto State and National Orthopaedic Hospital Dalla, Kano State) in northwest region of Nigeria from 2012 to 2016. Data retrieved include gender, age, etiology of ankylosis, duration of ankylosis, type of imaging technique, type of airway management, types of incision, surgical procedure, interpositional materials used, and complications. Ethical approval was obtained from the ethics and research committee of Usmanu Danfodiyo University Teaching Hospital.

A total of 36 patients with temporomandibular joint ankylosis (TMJA) were seen during the study period, out of which 7 (19.4%) patients had maxillary extension of the ankylotic mass. A male: female ratio of 1.3:1 was observed. Four (57.1%) patients were within the age group of 5 to 10 years, two (28.6%) in the age group of 11 to 15 years, while only one (14.3%) patient was within the age group of 31 to 35 years (►**Table 1**). ►**Figs. 2** and **3** show the preoperative and postoperative clinical photograph of one of the patients. All seven cases (100%) of maxillary extension were secondary to cancrum oris (noma) and preoperative interincisal distance was between 0 and 5 mm. Apart from the clinical features of ankylosis, we also noticed cheek scarring as a result of management of cancrum oris (►**Fig. 4**). In addition, intraoral fibrosis eliminating the upper and lower buccal sulci extending to the molar regions was also noted (►**Fig. 5**).

None of the patients in this series could afford a computed tomographic (CT) scan because of financial reasons; hence, they were only able to afford plain radiographs (transcranioblique with mouth opened and closed). Although these radiographs could not depict the maxillary fusion, total obliteration of the

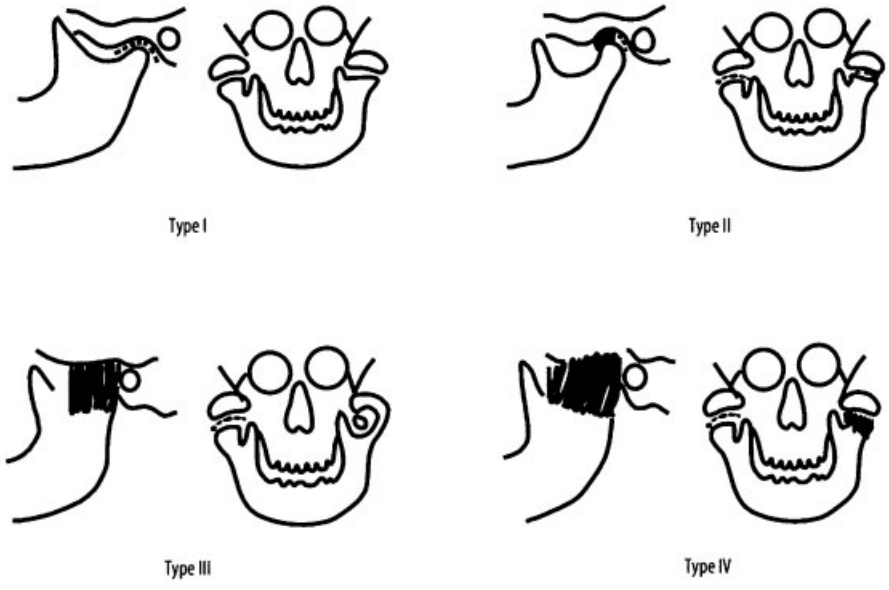

Fig. 1 Schematic diagram of Sawhney's classification of temporomandibular joint ankylosis.

Table 1 Summary of clinical parameters of cases of TMJA with maxillary extension

Age (years)	Sex	Etiology	EOA	TOS	IM	PoC	PoMO (cm)	PoF	ASN
5	M	CO	BIL C + R + M + Z	R + M	TM	FN palsy	3.0	6	Yes
9	F	CO	LF C + R + M + Z	R + M	TF	None	3.5	5	Yes
9	M	CO	RT C + R + M + Co	R + M	Ma	Scar	2.8	5	Yes
10	M	CO	BIL C + R + M + Z	R + M	TM	AOB	3.4	5	Yes
13	F	CO	RT C + R + M + Co	R + M	Ma	FN palsy	4.0	6	Yes
15	M	CO	RT C + R + M + Co	R + M	Ma	None	3.7	5	Yes
33	M	CO	LF C + R + M + Co	R + M	Ma	FN palsy	4.0	6	Yes

Abbreviations: AOB, anterior open bite; ASN, additional surgery needed; BIL, bilateral; C, condyle; CO, cancrum oris; Co, coronoid; EOA, extent of ankylosis; FN, facial nerve; IM, interpositional material; LF, left; M, maxilla; Ma, masseter; PoC, postoperative complication; PoF, postoperative follow-up; PoMO, postoperative mouth opening; R, ramus; RT, right; TF, temporalis fascia; TM, temporalis muscle; TMJA, Temporomandibular joint ankylosis; TOS, type of surgery; Z, zygoma.

joint space with shortened ramus was noted on the plain radiographs. Four (57.1%) patients had tracheostomy, while three (42.9%) had fiberoptic intubation. Bramley–Al-kayat incision was utilized in three (42.8%) cases, while postrami incision was performed in two (28.6%) patients. Two (28.6%) patients had combined Bramley–Al-kayat and postrami incisions. All the patients had ramus procedure with separation of the bony mass from the maxillary tuberosity via intraoral upper buccal sulcus incision. The fusion to the maxillary tuberosity was on the medial aspect of the extremely shortened, but broadened mandibular ramus, thus visibility of the fusion on exposure of the ramus laterally was not feasible (►**Fig. 6**). We also noticed that this group of patients had very short ramus with the angle of the mandible very close to the ankylotic mass. Furthermore, the styloid process was palpable close to the angle of the mandible medially indicating low level of the base of the skull. Based on this, the ostectomy made was just above the angle of the mandible in six patients to avoid accidental puncture of the sigmoid sinus that resulted into severe hemorrhage in one of the cases when the ostectomy was made high up through the ankylotic mass. The outcome of the ostectomy close to the angle resulted in little or no ramus (►**Fig. 7**) with prominent anterior open bite postoperatively as compared with other classifications. Also, all the patients had anesthesia of the cranial nerve V3 because of the mandibular resection at the angle of the mandible. However, this was not of concern to patients.

Temporalis fascia/muscle (►**Fig. 8**) and masseter muscle (►**Fig. 9**) were utilized as interpositional material. The patients had been followed up for a maximum of 6 months

Fig. 2 Preoperative photograph of temporomandibular joint ankylosed patient.

Fig. 3 Postoperative photograph of temporomandibular joint ankylosed patient.

Fig. 4 Cheek scarring as a result of management of cancrum oris.

Fig. 5 Intraoral fibrosis eliminating the upper and lower buccal sulci extending to the molar regions.

Fig. 7 Ostectomy close to the angle resulting in little or no ramus (white vertical arrow).

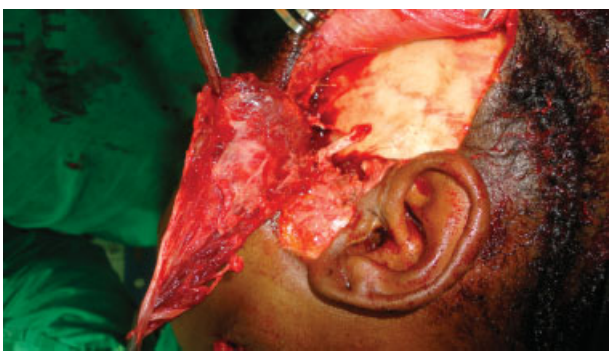

Fig. 8 Temporalis muscle/fascia for gap arthroplasty.

Fig. 9 Masseter muscle for gap arthroplasty.

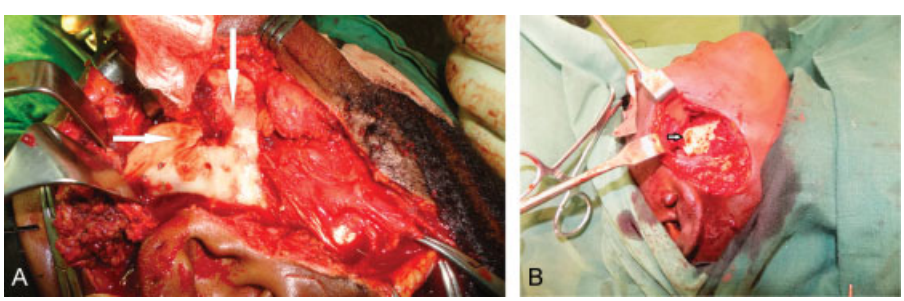

Fig. 6 (**A**) Intraoperative exposure of ankylotic mass involving the glenoid fossa, condylar head, zygomatic arch (vertical arrow), and maxilla (mesial to the horizontal arrow). (**B**) Intraoperative exposure of ankylotic mass extending to the maxilla (horizontal arrow).

after which they failed to report for routine check-up. Postoperative interincisal distance records were taken at the last follow-up period. All the patients required secondary surgeries (distraction osteogenesis and joint replacement) for correction of residual jaw deformities; however, all were satisfied with the opening of the mouth as aesthetics was not of their primary concern.

Discussion

Temporomandibular joint ankylosis has been described as the most common cause of acquired mandibular deformity in both children and adults if it is not managed in early age.[9] The etiopathogenesis of TMJA consist of previous trauma (road traffic accident [RTA] and fall), previous TMJ surgery, arthritis, and infection. Etiopathogenesis can also be congenital or idiopathic. It has been reported from the literature that the most common etiopathogenesis of TMJA is preceding trauma (RTA and fall) and closely followed by infection.[10–12] This position is in tandem with our findings as all the seven cases were due to cancrum oris (noma). Noma which is an infectious lesion destroys the facial and oral tissues and surrounding structures which can include the TMJ.[13] Cancrum oris in underdeveloped nations especially sub-Saharan Africa have been associated with poverty where there is high rate of chronic malnutrition, lack of portable drinking water, poor environmental and personal sanitation, and high contact with viral and bacterial pathogens[13–15] In northwest Nigeria, this condition has been reported to be common.[16,17]

The authors have reported pattern of tissue destruction in noma cases in northwest Nigeria with destruction of orofacial soft tissues, mandible, and maxilla[17]; however bony fusion of the mandible and maxilla alone was neither observed nor reported. On the contrary, Shivanand et al[18] have reported bony fusion as a sequelae of noma without the TMJ involvement. This sequelae of noma have been reported in various case series.[19] To the best of our knowledge, no report of TMJA existed together with mandibular and maxillary fusion in noma patient, thus necessitating this current case series. In addition, this new clinical entity did not fit into the classification of TMJA by Sawhney.

The successful treatment of TMJA requires detailed preoperative assessment of the type and degree of the deformity.[20] This information is important for accurate surgical treatment planning. The most important imaging technique reported in the literature for the preoperative assessment of TMJA is CT.[21,22] However, this mode of imaging is inadequate in resource-scarce countries, and if available, it is out of financial reach of citizens due to out-of-pocket payment system in the healthcare delivery. Sanders et al[23] have reported that conventional radiographs underestimated the extent of the bony ankylosis that is found at the time of operation. None of the proposed Class V patients in our series were able to afford a CT due to financial constraints. Because of this, the extent of the ankylotic mass was discovered intraoperatively.

This intraoperative discovery have led the authors to propose a modification of Sawhley's classification by the addition of Class V (when the joint architecture is replaced completely by bone with fusion of the condyle, sigmoid notch and coronoid process to the zygomatic arch, glenoid fossa, and maxilla). The modification will allow such an extent to be incorporated into the previous classification for proper patient management. This is critical because the plain radiographs (transcranioblique view of the skull), which the patients could afford, revealed only the shortened mandibular ramus with obliteration of the joint spaces. Since this maxillary fusion was noticed intraoperatively, we opined that perhaps if an occipitomental view of the skull was taken; it could have shown this fusion as radio-opacity in the ramus-maxillary tuberosity region. In view of the fact that occipitomental view of the skull is not required in accessing TMJ pathologies, it was not requested for in any of these patients. We thus propose inclusion of this radiographic view in TMJA where the history and clinical features may be pointing toward cancrum oris as the etiological factor and patient cannot afford a CT scan.

When TMJA occurs during growth of a child, it results in narrow oropharyngeal airway as a result of shortening of mandibular rami and narrowing of space between the mandibular angle.[24] All these structural abnormalities with limited or no mouth opening results in difficulty in securing airway.[24] Different techniques for airway management in TMJA include awake nasal intubation either blind or fiberoptic guided, retrograde intubation, and tracheostomy.[24,25] Tracheostomy has generally being regarded as the last option due to severe morbidity, long-term side effects, and mortality; however, this method has been our airway management in four (57.1%) patients, while three (42.9%) patients had fiberoptic intubations. The side effects of tracheostomy have been reduced in our series by early decannulation usually immediately following surgery and mostly within 24 hours postoperatively. This early decannulation has been reported by Braimah et al.[26]

Bramley–Al-kayat incision was utilized in three (42.8%) cases because it has been reported to provide outstanding visibility due to a larger flap with intact temporal fascia that is not reflected with it.[27] Additionally, it has been shown to have less chances of sensory damage,[27] simplicity of handling, closeness to the temporomandibular joint, good functional results, and successful clinical results.[28,29] Postrami incision was also utilized in two (28.6%) cases when the ankylotic mass has extended into the ramus for easy access. This approach into the ankylotic mass has been reported in the literature.[26] Two (28.6%) patients had combined Bramley–Al-kayat and postrami incisions when the ankylotic mass could not be approached via only one incision. In all the seven cases, intraoral incision was added to expose the ankylosis particularly to osteotomize the ankylotic mass from the maxilla. This combination approach may also be an arsenal in the management of the proposed Class V patients.

The basic principle in the management of TMJA is surgical release of ankylosis mass. This technique is largely termed gap arthroplasty in which the use of an interpositioning material to prevent reankylosis is frequently performed.[30] The gap arthroplasty could be performed on the condyle (Condylar procedures) or the ramus (ramus procedures).

Total joint replacement using costochondral graft from the ribs has also been reported.[31] Abbe[32] in 1880 was the first to report gap arthroplasty, which involved the removal of block of bone and leaving a gap between the ascending ramus and the temporal bone. Brisement force under general anesthesia has also been used in the management of fibrous ankylosis. The authors made ostectomy cuts in six (85.7%) cases very close to the mandibular angle to avoid the sigmoid sinus. This surgical maneuver was adopted when in one (14.3%) of the cases, severe hemorrhage occurred because the ostectomy cut was made high up the ramus through the ankylotic mass.

Several types of grafts (biologic and nonbiologic) have been utilized as interpositional materials to prevent TMJ reankylosis. The use of autogenous temporalis muscle as a myofascial flap has gained recognition due to its closeness to the surgical site. Other types of graft materials include masseter muscle, fascia lata, and auricular cartilage.[33] Silastic, Proplast/Teflon, metallic fossa implants, and acrylic marbles have also been used as alloplastic interpositional materials; however, they are fraught with high failure rates.[34] In our series, when the Bramley–Al-kayat incision was utilized, the temporalis fascia or temporalis muscle was utilized as the interpositional material. Similarly, when the postrami incision was used, the masseter muscle was the choice as interpositional material. Temporalis muscle has been favored as interpositional material in preventing reankylosis; however, temporalis muscle has been reported to fail due to contraction of the muscle postoperatively. To overcome this challenge, Balaji[29] has modified the temporalis muscle flap by anchoring the flap to the submandibular region to inhibit the contraction of the muscle postoperatively. No alloplastic material was utilized in our series.

Secondary surgery required to correct residual jaw deformities (distraction osteogenesis) was not performed as all of the patients were satisfied with jaw opening alone.

Another possible option apart from distraction osteogenesis for secondary surgery especially in noma-induced ankylosis is joint reconstruction with a customized total joint prosthesis, involving both the upper and lower joint elements. However, affordability of this prosthesis in a resource-scarce environment will be another challenge.

Conclusion

With the involvement of the maxilla in the ankylotic mass, the authors have proposed a modification of Sawhney's classification by the addition of Class V (when the joint architecture is replaced completely by bone with fusion of the condyle, sigmoid notch, and coronoid process to the zygomatic arch, glenoid fossa, and maxilla). The authors have suggested a name for the new classification to be "Modified Sawhney's Classification of temporomandibular joint ankylosis." Although computed tomography scan is the standard investigative procedure for TMJA, however, in resource-scarce environment, plain radiographs with their shortcomings still have their roles. In addition, aggressive postoperative physiotherapy for a sufficient period of time (minimum of 6 months) is vital.

Conflicts of Interest
None.

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