

Bilateral Temporomandibular Joint Ankylosis Caused by Rheumatoid Arthritis: A Case Report

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ABSTRACT----

Background: We present a case of bilateral Temporomandibular joint (TMJ) ankylosis due to Rheumatoid arthritis (RA), the first case in our centre and indeed in Nigeria to the best of our knowledge. The ankylosis was successfully released by bilateral condylectomy.

Methodology: The case records of a 60 year old woman with an inability to open the mouth over a period of 13 years following a previously rheumatoid arthritis of peripheral joints was reviewed. The details of clinical and radiological findings were recorded in addition to the definitive management.

Result: Bilateral condylectomy was performed and the ankylosis released. Vigorous jaw exercise was commenced on her and she had an uneventful post-operative stay. She was discharged home with moderate mouth opening. Early mobilization and aggressive physiotherapy was instituted in order to prevent re-ankylosis. She had not experienced relapse since a year after the procedure.

Conclusion: A rare case of bilateral TMJ ankylosis resulting from long standing RA hereby presented, this case was successfully managed by bilateral condylectomy. It is, of paramount importance, that dental health care providers have a proper knowledge of systemic diseases affecting the function of orofacial structures

Keywords--- TMJ ankylosis, Rheumatoid arthritis, Condylectomy

1. INTRODUCTION

Temporo-mandibular joint ankylosis can be defined as inability to open the mouth due to either a fibrous or bony union between the head of the condyle and glenoid fossa[1]. This immobility of the joint invariably results in difficulty of mastication, poor oral hygiene, impairment of speech, facial asymmetry and mandibular micrognathia particularly in young children with complete inability to open the mouth[2].

The principal causes of ankylosis of TMJ are trauma, infection and other inflammatory conditions[3]. The primary inflammatory condition that commonly affects TMJ is seen in cases of RA [4] which is thought to be autoimmune in nature.

RA is a multi-systemic chronic symmetrical polyarthritis of unexplained cause that is characterised by chronic inflammatory synovitis of mainly peripheral joints[5-7]. One theory is that an autoantibody to abnormal immunoglobulin in joint tissues leads to the formation of an antigen-antibody complex which activates complement causing inflammation and synovial damage[8]. The mean age of onset is between 30 and 40 years with female predominance.

TMJ is affected in half of all cases but it is usually among last joints to be affected[4] and the involvement is often bilateral and of fibrous variety.

The frequency of temporomandibular joint involvement based on clinical and radiological findings is rather diverse and involvement may manifest as pain, restricted range of movement and locking of the joint[9]. TMJ ankylosis is however a rare complication. The aim of this article is to present a case of bilateral bony TMJ ankylosis caused by RA which to the best of our knowledge is the first case to be reported in Nigeria.

2. CASE REPORT

A 60-year old female Nigerian was referred to the Oral and Maxillofacial clinic, with a complaint of 13 years inability to open the mouth. She gave a history of progressive limitation of mouth opening with an associated pain at both TMJ which grew worse over a period of time. No known history of crushing blows or transmitted force to both joints. This was her first dental visit.

However, eight years before the complete stiffness of joint, she had intermittent generalised painful swelling of wrist, knee, ankle and TMJs which was associated with fever. The TMJs were the last joints to be affected by the generalised inflammatory condition. Due to an episode of acute exacerbation of her clinical condition, she was later referred from a missionary hospital based in the city of Jos North Central part of Nigeria to a tertiary health centre located in southwestern part of Nigeria and thereby hospitalized for three and a half months with a diagnosis of RA. The generalised inflammatory joint condition was successfully managed but without any surgical intervention to release the ankylosed jaws at the time of her discharge from the aforementioned hospital. Drug history revealed that the patient had been on steroid (prednisolone) and non-steroidal anti-inflammatory analgesic (diclofenac sodium, celecoxib).

Physical examination revealed an alert, middle-aged woman in no respiratory distress with a slightly forward bending posture and walking with an aid of a "walking stick". Vital signs were within normal range. There was a fixed flexion deformity of the proximal interphalangeal joints (boutonniere deformity), Z deformity of the thumbs and painless bony swellings of both hands. A classic ulnar deviation of the fingers was also present (fig.1). The knee assumed a valgus position. Also the big and small toes exhibiting angulation from the mid-line of the body, overriding each other and stiffness of the adjoining metatarsal joints was noticed. Hallux rigidus was present on both feet (fig.2). There was no facial asymmetry and very slight transmitted movement felt at both TMJs and no scar was noticed on the chin region. The patient's oral hygiene was poor, with occlusal attrition of the incisors. There was Angle's class 1 type occlusion and full complement of teeth except third molars. The patient could only produce an inter incisal distance (IID) opening of about 2mm on maximum effort (fig.3). Radiographic evaluation comprised of reverse Townes and transcranial view of TMJs which revealed complete obliteration of the joint space on both sides. Further radiographs of wrist, knee and ankle joints were taken.

Based on these findings, a diagnosis of bilateral TMJ ankylosis secondary to RA was made. Routine biochemical investigations of the blood did not show any abnormality. Rheumatoid factor was found negative even though assay for citrulline antibody was not carried out because lack of facilities for such comprehensive investigation in the centre. Erythrocyte sedimentation rate and uric acid values were within normal range. No abnormality of the heart was detected on electrocardiogram. Patient was then prepared for surgery.

General anaesthesia was administered through a blind naso-tracheal intubation as fibre-optic intubation was not available in the centre. The blind intubation was successful at second attempt even though preparations were made for tracheostomy in the event the blind intubation had failed.

A pre-auricular incision was made on both sides thereby exposing the joint areas. Both heads of the condyle were found to be fused to the glenoid fossa. Bilateral condylectomy was carried out, patient mouth was opened with a side-action mouth prop in order to break any further adhesion and a mouth opening of IID 34mm was achieved immediately.

Post-operative recovery was uneventful and rigorous mouth opening exercise using an acrylic cork screw commenced on the second post-operative day. The jaw exercise was carried out at least three times daily during her hospitalization and to be continued by the patient for a period of 12 months after discharge from the hospital.

Twenty eight days after surgery the patient was discharged with an IID mouth opening of 26mm. She was under regular follow-up for a period of twelve months. Interincisal opening had been maintained and further increased to 27mm (fig.4). She has now regained considerable body weight though the deformities of the fingers and toes remain to be corrected.



Fig. 1

Fig. 2

Fig. 3

Fig. 4

3. DISCUSSION

The causes and treatment of TMJ ankylosis have been well documented, with trauma and infection or inflammatory condition as the two leading causes [3,4,10]. Furthermore, TMJ is susceptible to conditions that affect other peripheral joints in the body such as ankylosis, dislocation, developmental anomalies and arthritis [4]. RA is a systemic inflammatory condition that commonly affects the TMJ and they are the last to be affected among the joints involved during the natural course of the disease [5,11,12]. No known explanation has been given for this pattern of joint involvement; this may, however, be due to more range of activities at the joint. In this report it was noticed that the TMJs were the last to be affected during the generalised inflammatory joint episode which is still in agreement with the reported findings in the literatures.

In RA, pathological changes have been noticed in the affected synovial TMJ which include the condylar head erosion, degeneration, temporal subchondral cysts and decrease joint space [5,13-16]. The synovial lining cells which are normally one to three layers thick undergo hyperplasia and hypertrophy during inflammation. This inflamed synovium (pannus) often creeps over and erodes the articular cartilage with the adjacent bone. Eventually, the joint is destroyed and undergoes fibrous union in the early stage which will further lead to bony union on long-standing cases [14]. The history of long-standing inflammatory process in our report probably must have resulted in metaplasia of fibrous tissue of joint space to fibrocartilage which eventually led to bony ankylosis. Blackwood [4], in examining the post-mortem specimens of fibrous ankylosis caused by RA, highlighted destruction of bony architecture of the joint and entire joint space filled with fibrous tissue.

The presence or absence of rheumatoid factor is not specifically diagnostic for RA [5,14]. Patients suffering from RA may be seronegative for rheumatoid factor when the course of the disease is punctuated by remissions [5]. In this report the patient was not experiencing any acute inflammatory episode, hence rheumatoid factor was negative. The surgical release of TMJ ankylosis under general anaesthesia requires the experience of a competent anaesthesiologist because of the difficulty of achieving a clear airway for administration of anaesthesia. Therefore, a careful planning with the anaesthesia team is of paramount importance. Fibre-optic intubation is the recent technique for patients with ankylosed mandible [15]. However, because of non-availability of such facility in the centre, a blind naso-tracheal intubation was carried out and it was successful at second attempt. Though Adekeye [6] reported cases of death and pharyngo-mediastinal fistula complicating blind intubation, in our report it was well tolerated by the patient.

It is generally agreed that the only definitive treatment of TMJ ankylosis is surgical [17], precise site (intra-capsular or extra-capsular), type of tissue involved (bony, fibrous or fibrous-osseous), extent of ankylosis (complete or incomplete) invariably determines the surgical method to be employed. Condylectomy, gap arthroplasty or interpositional arthroplasty have been used to release ankylosis confined to the intra-capsular region [6]. Bilateral condylectomy via a pre-auricular approach was performed on the patient since the site of the ankylosis was between the head of the condyle and glenoid fossa. Anterior open bite was the attended complication from the procedure. It is believed that gap arthroplasty allows the mandible to rotate posteriorly and upwardly which eventually results in a patient developing anterior open bite [4]. Filling in the surgically created space with an interposing material such as autogenous grafts or alloplastics will prevent this complication. In advanced treatment centres, several prosthetic options for TMJ reconstruction exist including custom fossa implants, mandibular reconstruction plates with condylar heads and silastic sheeting materials [15]. But Adekeye [6] in his own opinion emphasized the importance of reduction of operative pain and facilitation of early active exercises than interpositional arthroplasty.

One of the major problems of prolonged ankylosis is the disuse atrophy and fibrosis of the muscles of mastication. It has been reported that patients with ankylosis for less than five years do not have disuse atrophy, whereas those with ankylosis for more than five years have atrophy [4]. In a long standing case in this report, disuse atrophy must have involved the masticatory muscles which invariably limited the maximum mouth opening even immediately after surgical release. Therefore, in this patient active jaw exercises began early and continued regularly for at least for one year thereby stretching scar tissue and preventing neo-osteogenesis [6]. Opening in the range 25mm to 30mm at the end of a year is considered satisfactorily. The mouth opening of the patient was 27mm at the end of a year after surgery (fig.3). Diligence and perseverance of the patient in exercising accounted for retention of mouth opening.

4. CONCLUSION

A rare case of TMJ ankylosis resulting from long standing RA hereby presented, this case was successfully managed bilateral condylectomy. Adequate resection of the ankylosed bone, early post-operative exercises, appropriate physiotherapy and close follow-up of the patient plays an important role in the prevention of post-operative adhesions.

5. LEGEND TO ILLUSTRATIONS

Fig. 1: .Showing fixed flexion deformity of the proximal interphalangeal joints (boutonniere deformity), Z deformity of the thumbs and classic ulnar deviation of the fingers.

Fig. 2: Showing Halus rigidis of both feet and overriding of little toes

Fig. 3: The patient could only produce an inter incisal distance (IID) opening of about 2mm on maximum effort

Fig. 4 : . Mouth Opening of about 27mm maintained a year after surgery

6. REFERENCES

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