Unusual causes of trismus: a report of two cases

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Abstract Chronic inability to achieve normal mouth opening can be a symptom of several diseases. Mostly the causes of inability to open mouth are articular and sometimes the cause may be extra-articular. In the two cases which are being reported in this article the causes of limited mouth opening are extra-articular and that too from rare pathologies, of which one case is of OKC of the coronoid process and the other case is of myositis ossificans of the buccinator and medial pterygoid muscle. Both the cases had adequate mouth opening both intra-operatively and at long term follow up.

Keywords OKC · Myositis ossificans · Trismus · Coronoid process

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Introduction

Restricted mouth opening is a commonly encountered problem in oral and maxillofacial surgical practice. The causes for this can be broadly classified as articular or extra-articular. Although Tveteras and Kristensen [1] have defined trismus as a prolonged tetanic spasm of the jaw muscles by which normal mouth opening is restricted (locked jaw), in common parlance, it is often used as a synonym for reduced mouth opening due to extra-articular causes. Luyk and Steinberg [2] have enumerated the possible causes of trismus as under (Table 1):

We present two cases of who presented with restricted mouth opening as their chief complaint with uncommon, though not unknown, etiologies.

Case 1

A 60-year-old female patient of low socioeconomic status reported with the complaint of inability to open her mouth since 7–8 years. There was no history of trauma, otitis media or externa, exanthematous diseases or space infection. She did not have any previous habit of pan, gutka, tobacco or betel nut chewing.

On examination, there was no facial asymmetry but her mouth opening was

Table 1

Some possible causes of trismus are:

1. Trauma
 - a. Acute
 - b. Chronic
2. Neoplastic
 - a. Benign
 - b. Malignant
3. Neuromuscular disorders
eg. Parkinson's disease
4. Reactive
 - a. Acute: masticator space infection
 - b. Chronic: TMJ ankylosis, oral submucous fibrosis, MPDS, post radiotherapy and degenerative joint disease
5. Congenital
6. Psychogenic – hysteria, hyperventilation syndrome
7. Drug induced – strychnine poisoning

restricted to 2 millimeters only (Fig. 1). Any attempt to manually open her mouth gave a 'hard-end' feel suggestive of an obstructive bony etiology. Radiological investigations were significant as the OPG revealed a single 3cm x 2cm radiolucency at the right coronoid process. The radiolucency was unilocular, had thick radiopaque borders and had no tooth or tooth germ associated with it (Fig. 2). Both the condylar heads

appeared normal in shape and size although the joint space was reduced bilaterally.

A provisional diagnosis of extraneous variety of primordial cyst or unicystic ameloblastoma involving the right coronoid was made. Mechanical obstruction due to expanded coronoid was constructed to be the cause for the trismus.

The patient was taken up for surgery under general anesthesia after routine investigations. Fiber-optic intubation was performed and the coronoid process was exposed via the Alkayat and Bramley incision with Popowich modification. An 'L' shaped excision of the coronoid process was performed thus removing the pathology in toto (Fig. 3). An improvement in mouth opening of 10 to 15 mm was noticed immediately. In order to further improve the mouth opening bilateral high condylectomy and gap arthroplasty was performed. Closure of the surgical site was done in layers and postoperative recovery was uneventful.

Histopathological examination of the specimen confirmed the diagnosis of an extraneous variety of odontogenic keratocyst (Fig. 4). 5 years postoperatively there is no sign of recurrence and patient has a satisfactory mouth opening of 33 mm (Figs. 5 and 6).

Fig. 1 Preoperative presentation

Fig. 2 OPG showing well-defined radiolucency at the coronoid process

Fig. 3 Planned surgical excision

Fig. 4 Histology showing OKC

Fig. 5 Postoperative mouth opening

Fig. 6 Postoperative OPG showing bilateral gap arthroplasty and 'L' shaped excision of pathology at right coronoid process

Fig. 7 Preoperative presentation

Fig. 8 CT showing calcification

Fig. 9 Preoperative OPG

Fig. 10 H & E staining at 10X showing muscle fibres undergoing hyalinization and signs of calcification

Fig. 11 Postoperative OPG

Fig. 12 Postoperative mouth opening

Case 2

A 20-year-old female reported with the chief complaint of inability to open mouth since past 1 year. She had sustained dento-alveolar trauma to her right mandible 2 years ago following which all the teeth of right mandibular quadrant were extracted by a dentist. However, the patient noted a

progressive decrease in mouth opening following the extractions resulting in almost complete trismus.

On examination, she had a noticeable hollowing over the right cheek with a maximum mouth opening of 1 mm (Fig. 7). Intraorally, a firm fibrous band was palpable extending from maxillary tuberosity region to mandibular ridge

involving the pterygomandibular raphe. The overlying mucosa was normal. Temporalis and masseter muscles bilaterally showed no abnormalities. The temporalis tendon could not be palpated intra-orally adequately because of the fibrosis present.

On radiographic examination, a calcified mass extending from the maxillary

tuberosity to the anterior border of the ramus on right side was observed (Fig. 8). Both her TMJs were deemed to be normal on plain radiographs (Fig. 9) as well as CT scans. Blood investigations were within normal limits, including serum calcium, phosphorous, alkaline phosphate and parathormone level. A provisional diagnosis of myositis ossificans traumatica was made.

Under general anesthesia, the fibrotic calcified mass extending into the buccinator and medial pterygoid was dissected out along with the overlying buccal mucosa. Considering the nature of the disease and to further improve the mouth opening, it was thought prudent to perform bilateral coronoidectomy. 35mm of mouth opening was achieved intraoperatively. The buccal mucosal defect was reconstructed by an ipsilateral palatal pedicle flap. Postoperative recovery was uneventful. Histopathologic examination of the specimen showed dense fibrous hyalinization (Fig.10) of the tissue and microcalcifications, which confirmed the diagnosis of myositis ossificans.

After 3 months, an associated lateral ankyloglossia was corrected by excision of the band and interposition of split thickness skin graft. The external hollowing was filled with free fat graft from thigh region in an attempt to correct the facial asymmetry. She maintained a mouth opening of 30 mm postoperatively on follow-up at 1year (Figs. 11 and 12).

Discussion

Ankylosis of the TMJ is the best known cause of long-standing restriction of mouth opening. Common causes of chronic reduced mouth opening due to extra-articular etiologies are few e.g. oral submucous fibrosis, radiation therapy and pathologies of the masticatory muscles. Therefore when a patient presents with an inability to achieve a normal mouth opening, a careful history must be obtained and a thorough clinical examination should be done.

Involvement of the coronoid process in pathological processes is a known cause for limited mouth opening. Elongation of the coronoid and tumours of the coronoid tip are unusual causes for trismus [3]. Aneurysmal bone cyst at the coronoid process has been reported [4,5] as a cause of restricted mouth opening. However, OKC of the coronoid process presenting as a cause for trismus, in our knowledge, has not been reported earlier.

The OKC is one of the most common cystic lesions of the jaws. The lesion is more frequent in the mandible than in maxilla [6]. More than half of the OKCs occur at the angle of mandible, and extend for varying distance into the body anteriorly and into ramus, condyle or coronoid regions posteriorly [7,8]. OKC can be associated with pain, swelling or discharge and occasionally paresthesia of the lower lip or teeth [9].

In our case however, the presentation of OKC was highly unusual clinically since trismus was the only presenting symptom for the patient. Radiographically, the finding was of an extraneous variety of a cystic lesion which was confirmed histologically as an OKC. The cyst occurred in this unusual location without the presence of any of the features of basal cell nevus syndrome (Gorlin Goltz Syndrome).

The second case of myositis ossificans traumatica as a cause of trismus too is rare and worthy of discussion. Myositis ossificans traumatica (eponyms: myositis ossificans circumscripta, ossifying hematoma, calcified hematoma, parosteal bone formation) was initially described by Thoma in 1958 as a condition generally caused by calcification and progressive ossification of an intramuscular hematoma after trauma [10].

Two distinct forms of myositis ossificans have been described, namely:

Myositis ossificans progressiva: is a rare hereditary connective tissue disorder of unknown origin. It has an autosomal dominant inheritance with variable penetrance and expressivity. It involves an over expression of the bone morphogenic protein 4 (bmp4) and its messenger Ribonucleic acid (mRNA). This protein has been mapped to the chromosome band 14q22–q23.

Myositis ossificans traumatica: (fibrodysplasia ossificans circumscripta, ossifying hematoma, periosteal bone formation, myositis ossificans circumscripta). This involves an aberrant reparative process which causes benign, heterotropic (i.e. extraskeletal), ossification of soft tissues after blunt trauma, fractures, surgical incision, or repeated minor trauma to muscle.

The differential diagnosis of myositis ossificans traumatica includes extraosseous osteosarcoma, Klippel-Feil syndrome, rheumatoid arthritis and dermatomyositis. Myositis ossificans traumatica has a rate of incidence in head & neck region of about 0.61×10^{-6} internationally. In their literature review

Arima et al. had discovered about 26 cases in the head and neck region [11].

Bisphosphonates (Glendronate, Risedronate, Etidronate) have been tried on study basis for myositis ossificans progressiva, which attempts to block aggregation of hydroxyapatite crystals and decrease the bone formation. Dosage of 400mg twice daily 2 weeks preoperatively and 4 weeks postoperatively can be administered. Bisphosphonates have been shown to alleviate some symptoms during active phases of myositis ossificans progressiva however they have minimal effectiveness in the treatment of myositis ossificans traumatica [12,13,14].

Myositis ossificans progressiva commonly involves the following muscles in decreasing order of involvement masseter, temporalis, genioglossus, buccinator, medial pterygoid [11,15].

Carey [16] summarized various theories regarding the pathogenesis of this condition as:

1. Displacement of bony fragments into soft tissues and hematoma with subsequent proliferation
2. Detachment of periosteal fragments into surrounding tissue with proliferation of osteoprogenitor cells
3. 'Leakage' of subperiosteal osteoprogenitor cells into surrounding soft tissue through periosteal perforations suffered via trauma
4. Differentiation of extraosseous cells exposed to bone morphogenic protein (BMP) [17].

Last theory is the most accepted explanation for myositis ossificans.

Conclusion

Trismus is a fairly common presenting complaint amongst patients seen by oral and maxillofacial surgeons, but long standing limitation of jaw opening due to causes other than TMJ ankylosis is unusual. An accurate diagnosis of these causative factors is necessary for appropriate management of the condition, and provides the patient their best possible chance to overcome their disability.

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