Management of a case of osteoma of coronoid: A rare case report

ABSTRACT
Coronoid process osteoma is an extremely unusual and slow growing tumor which causes functional limitations of the temporomandibular joint. Till December 2014 only 7 cases have been reported worldwide. This case report is about a 40-year-old male patient with osteoma of left coronoid process. Treatment plan constitutes of surgical resection of the mass and post operative physiotherapy.

Keywords: Coronoidectomy, osteoma, pseudoankylosis

INTRODUCTION
Extrinsic joint pathologies constitute one of the chief causes of restricted mandibular movements. Trauma, infection, ankylosis, and arthritis are few etiological factors that lead to progressive functional limitation of the temporomandibular joint. Coronoid process osteoma is an extremely unusual and slow-growing tumor that can lead to restriction of mandibular movements. The first case of compact osteoma of the coronoid process of mandible was reported by Lewars in 1959. Osteomas comprise histologically normal membranous bone varying from insignificant thickening to large masses affecting the skeletal system largely. They are predominantly restricted to the craniofacial skeleton, and the most common sites include the paranasal sinuses and the mandible. True osteomas of the jaws may arise centrally or subperiosteally or in a peripheral location and might be either cancellous or compact in nature.[2,3] The cause of osteoma in the maxillofacial region is reported to be reactive bone hyperplasia or advanced bone ossification. Coronoid osteomas are largely asymptomatic and are nontender until their size and position incommodes with functioning. The present report describes one such case. Aspects dealing with the differential diagnosis, treatment, surgical access, possible complications, and outcomes have been elucidated. Until December 2014, only seven authors have described this rare entity in scientific literature [Table 1].

CASE REPORT
A 40-year-old male reported to us with a complaint of progressive restriction in mouth opening for 1 month. The restriction in mouth opening had gradually increased to the present state [Figure 1a]. There was no history of associated maxillofacial trauma, infections, or any surgical procedures. The patient presented with mild facial asymmetry toward the left with an accentuated prominence over the left malar region. Temporomandibular joint examination showed restriction of translatory and lateral excursive movements.

On intraoral examination, a tender, ill-defined bony mass could be palpated in the coronoid process of the ipsilateral side.

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Table 1: Courtesy: Table form of published cases on coronoid osteoma till date, da Costa Araújo et al.\textsuperscript{[7]}

| Authors          | Gender age (years) | Affected side | Clinical aspects                          | Mouth opening (before) (mm) | Mouth opening (after) (mm) | Image exams | Access type   | Lesion dimension (cm) |
|------------------|--------------------|---------------|------------------------------------------|-----------------------------|---------------------------|-------------|--------------|----------------------|
| Lewars, 1959     | Male, 15           | Right         | Lockjaw, edema in zygomatic region       | 3                           | 12                        | X ray       | Extraoral    | 3*1.25*1.5           |
| Ord et al., 1983 | Female, 40         | Left          | lockjaw, edema, paresthesia, fracture of zygomatic arch | Not mentioned              | Not mentioned             | X ray       | Extraoral    | 4*3.5*2              |
| Plezia, 1984     | Female, 26         | Right         | lockjaw, edema                          | Not mentioned              | Not mentioned             | X ray       | Intraoral    | Not mentioned        |
| Wesley et al., 1987 | Female, 12        | Bilateral     | Garden’s syndrome                       | 12                          | 32                        | X ray       | Not mentioned | 5*3*2               |
| Kurita et al., 1991 | Female, 40         | Right         | lockjaw, edema, fracture of zygomatic arch | 17                          | 30                        | Tomography  | Extraoral    | 3*2*1.5             |
| Chen et al., 1998 | Female, 28         | Right         | lockjaw                                 | 11                          | 28                        | X ray       | Tomography   | 2.5*3*3             |
| Vashishth et al., 2013 | Female, 26       | Not mentioned | Lockjaw, chewing difficulty, deviation on opening to the right side | 20                          | 35                        | X ray       | Tomography   | Not mentioned        |
| Araujo et al., 2013 | Female, 45        | Right         | Lockjaw, Fracture of zygomatic arch, edema | 8                           | 25                        | Tomography  | Intra and Extraoral | 3.5*4*2.5 |

Intraoperatively, the mouth opening improved and was recorded as 30 mm, following which contralateral coronoidectomy was carried out intraorally, thereby increasing the mouth opening to 45 mm. Zygomatic arch repositioning and stabilization was done using titanium miniplate and screws [Figure 1b].

The resected bony mass had a mixed nodular and smooth surface, measuring about 4 cm × 3 cm × 2 cm [Figure 3].

Microscopic examination of the specimen revealed a dense lamellar bone with marrow space and trabeculae lined by periosteum. Unlike osteochondroma, it lacked the typical cartilaginous cap [Figure 4].

The postoperative recovery was uneventful with no facial nerve deficits. No occlusal discrepancy was observed and the mouth opening was found to be 35 mm 1 month later. The patient was advised to continue aggressive physiotherapy and regular follow-up.

**DISCUSSION**

Osteoma is a benign tumor which arises from the proliferation of compact or cancellous bone, which leads to hypomobility of the mandible. It can reach a significant size, causing an increase in volume, facial asymmetry, limited mouth opening, and fracture of zygomatic complex in some cases. Central type, peripheral type, and extraskeletal type are the three variants based on the origin.\textsuperscript{[4]} A central osteoma arises from the endosteum, while the periosteum gives rise to the peripheral variant. An extraskeletal soft-tissue osteoma is of muscular origin.\textsuperscript{[5]} Coronoid osteomas lead to limitation...
Osteoma is most common in the fourth and fifth decades, but lesions are reported from 10 to 79 years. They are usually solitary. However, Gardner’s syndrome is associated with multiple such tumors.

Osteochondromas of the coronoid process have been reported in literature by Shackelford and Brown in 1943. The etiology is unknown, however it might be associated with aberrant activity of the surrounding periosteum. An endochondral ossification may occur around a foci of such metaplastic cartilage, leading to the formation of exostoses.

Another supporting theory is that it may arise post trauma, wherein a hematoma may undergo fibrosis and lead to the formation of chondrocytes. Histopathological evaluation of osteoma reveal absence of cartilaginous cap which can be attributed to shifting of the growth to the juxtaposition of new bone rather than that of endochondral ossification.

Coronoid hyperplasia can also be a striking differential to our diagnosis as it may occur as a reactive bone hyperplasia secondary to endocrine stimulus, trauma, increased temporalis activity, or genetic influences. Another differential to restricted mouth opening is Jacob’s disease, which differs from the osteoid osteoma owing to the histopathological evidence of regions of endochondral ossifications enclosed by hyaline cartilage.

Histopathological evaluation, therefore, is a key tool in establishing a diagnosis in such cases.

Kersher et al. reported a theory presuming that osteomas of the coronoid process are the sequelae of osteochondromas after total ossification of cartilage. Exhibition of cellular pleomorphism of chondrocytes and disturbances in
endochondral ossification are suggestive of neoplastic nature. Hence, they have been correctly categorized as tumors by several authors.

Facial asymmetry is the most common presentation of such patients owing to the lack of other symptoms apart from functional limitations, if any. Continuous growth might push the cortices of zygoma, resulting in an evident malar bulge and leading to the resorption of zygoma, which is followed by remodeling into a state of pseudo-ankylosis, resulting in trismus.

The aim of treatment is to restore the function, that is, optimal mandibular movements and chewing. Depending on the size, location, and type of restriction, surgery can be performed using a submandibular, coronal, retromandibular, preauricular, or intraoral access. It may accompany myotomy of masseter and temporalis muscles to relieve the limited range of motion as the facial musculature gets adapted to the existing scenario. Surgical excision of the osseous mass is the preferred treatment for patients with functional impairment. The coronoid process can be approached extraorally, intraorally, or using a combination of both. However, extraoral approach is preferred for larger coronoid process lesions. Osteomas of the coronoid process have a satisfactory prognosis, with no recurrence. The procedure involved is carried out with utmost care to avoid severing of the facial nerve. The intraoral approach involves an incision from the superior limit of the coronoid process to the retromolar trigone region with exploration of the entire anterior mandibular ramus. This procedure offers safety in terms of associated nerve damages and extraoral scars. However, the accessibility is compromised.

Postoperative mouth opening exercises, physiotherapy, and regular follow-up of treated cases are advocated. The present report is a pursuit to draw the attention of clinicians to such rare cases, where trismus is among the clinical findings. Such a case, if diagnosed and managed effectively, will result in impressive functional and esthetic outcomes.

Declaration of patient consent
The authors certify that they have obtained all appropriate patient consent forms. In the form, the legal guardian has given his consent for images and other clinical information to be reported in the journal. The guardian understands that names and initials will not be published and due efforts will be made to conceal identity, but anonymity cannot be guaranteed.

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Conflicts of interest
There are no conflicts of interest.

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