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Abstract

Background:

Osteoma is a benign tumor composed of both cortical and cancellous bones that increase in size with continuous formation of bone. The pathogenesis is unknown. Osteomas can cause symptoms depending on

their location and size. They can be asymptomatic or symptomatic, with trismus, limitation of mouth opening, and progressive malocclusion with facial asymmetry and can be painful.

Aim:

The aim of this paper is to report an unusual case of osteoma in the mandibular condylar neck and review the cases of mandibular condyle osteomas that have been reported in the last 15 years.

Conclusions:

Only a few cases involving the temporomandibular joint have been reported. We report an unusual case of osteoma in the mandibular condylar neck causing restricted mouth opening in addition to pain. Complete surgical excision in symptomatic cases is the therapy of choice with a low recurrence rate.

Keywords: Condyle, facial asymmetry, mandible, osteogenic tumor, osteoma, temporomandibular joint

Introduction

Osteoma is a benign tumor composed of both cortical and cancellous bones that increase in size with continuous formation of bone.[1] It represents an uncommon lesion that occurs mainly in craniofacial complex bones.[2] In jaws, it can appear on the bone surface as a polypoid or sessile mass, characterizing a peripheral osteoma, or can be a lesion in the medullar space, called central osteoma.[3] The majority of osteomas in the craniofacial skeleton are peripheral, while central osteomas are uncommon.[4]

The pathogenesis is unknown. Reaction mechanisms, traumas, or infections are suggested as possible causes.[5] They affect most young adults, with no preference by sex. The mandibular cases occur in the angle, mandibular condyle, and molar regions of the body, being frequent intraoral cases in the lingual regions close to molars and premolars.[6] When the mandibular condyle is involved, trismus, limitation of mouth opening, progressive malocclusion with midline shift toward the nonaffected side, contralateral mandibular deviation, and facial asymmetry are the common findings.[3,6]

In Gardner syndrome, multiple maxillary osteomas can be associated with multiple intestinal polyposes, osteomas, and epidermoid cysts. In addition, dental abnormalities include an increased frequency of multiple odontomas, as well as supernumerary and impacted teeth.[7]

We report an unusual case of osteoma in the mandibular condylar neck that presented in a 52-year-old female, causing restricted mouth opening in addition to pain.

Case Report

A 52-year-old female presented to the outpatient clinic of the Craniomaxillofacial Surgery Unit of the University of Ferrara (Italy) with a 7-year history of a slow-growing swelling over the preauricular area and pain of the temporomandibular joint (TMJ) area radiating to the right face.

The onset of the swelling was insidious and gradually increasing in size. It was not associated with trauma or infection and the patient had not undergone any recent surgery.

On physical examination, there was pain in the right TMJ during mouth opening and palpation. Maximum mouth opening was limited to 1.2 cm [Figure 1]. There was malocclusion with mandibular deviation toward the right side on mouth opening and limitation of lateral and protrusion movements. On palpation, there was a swelling in the right preauricular region. The swelling was bony hard in consistency and nontender and attached to the underlying bone. The overlying skin was normal and freely movable, and there was no local rise in temperature. No regional lymph nodes were palpable.

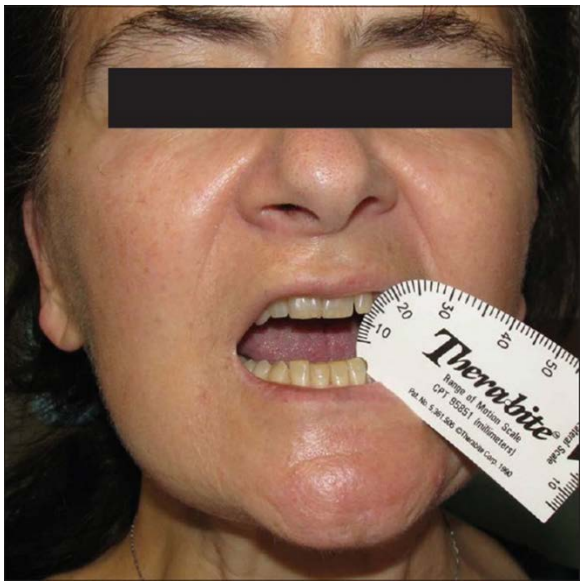

Figure 1 Preoperative mouth opening 12 mm

Computed tomography (CT) scan showed a well-defined radiopaque lesion involving the glenoid fossa and the right condyle [Figure [Figure2a2a and andb].b]. Routine hematological and biochemical investigations were within the normal limits.

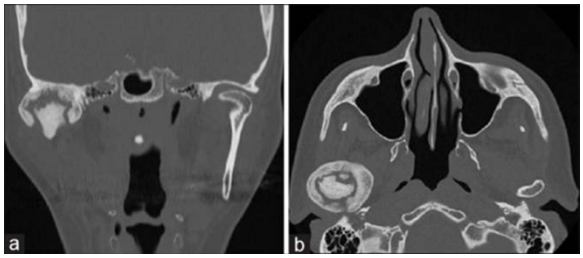

Figure 2 (a) Preoperative computed tomography scan coronal view. (b) Preoperative computed tomography scan axial view

Surgical correction was planned and performed under general anesthesia. The lesion was approached through a preauricular incision with temporal extension. After exposure of the entire lesion, an osteotomy was made with piezosurgery so that the condyle head with the lesion could be removed. A pedicled temporalis muscle flap was raised and positioned in the glenoid cavity, as interpositional material between the bony articular surfaces.

An intraoral incision was made and the right coronoid process was approached with stripping of the temporalis attachment on the anterior border of ramus. An osteotomy cut from the sigmoid notch to the anterior border of ramus was done, and the coronoidectomy was performed.

After the procedure, the maximum mouth opening was 5 cm. A subcutaneous suction drain was inserted. Sutures and dressing completed the operation. The specimen was sent for histopathological examination, which confirmed osteoma. The recovery was uneventful.

A 6-month postoperative CT scan showed a stable morphological result [Figure [Figure3a3a and andbb].

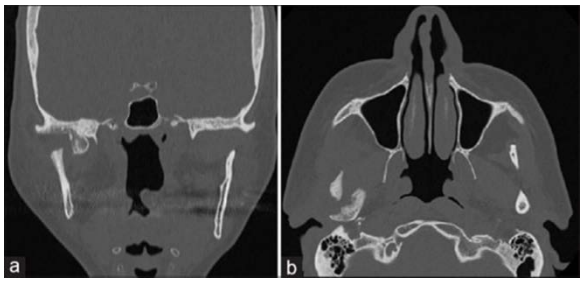

Figure 3 (a) Postoperative computed tomography scan coronal view. (b) Postoperative computed tomography scan axial view

At 12-month follow-up, the patient showed an improvement in facial symmetry with a good mouth opening (32 mm) [Figure [Figure4a4a and andb]b] and stable occlusion and with no limitations in mandibular movements or pain in mouth opening. No signs of recurrence were observed.

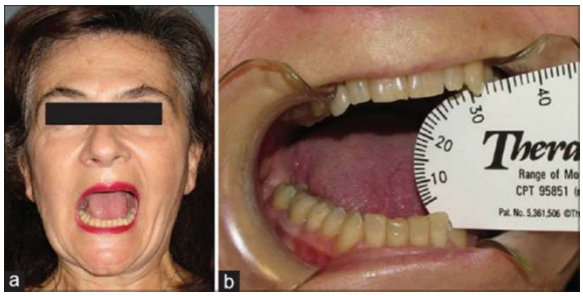

Figure 4 (a) Twelve-month postoperative follow-up. Mouth opening 32 mm. (b) Close-up of mouth opening

Discussion

Osteomas of the mandibular condyle are considered extremely infrequent lesions. The first case of a condylar osteoma was described in 1927 by Ivy.[8] Since then, only few cases of osteoma arising from the condylar process have been reported. The pathogenesis is unknown. Some authors consider osteoma true neoplasia, while others consider it a hamartoma. The precise location of an osteoma is usually in proximity to the regions of muscle attachment, suggesting that muscle traction may play a role in its development.[9] Reaction mechanisms, traumas, or infections are also suggested as possible etiologic factors.[5]

In jaws, osteomas may appear on the surface of the bone as a polypoidal or sessile mass, arising from the cortical bone (peripheral osteoma), or can be located within the medullary space, arising from cancellous bone (central osteoma). Thus, osteomas occurring in the mandibular condyle can appear as a sessile or pedunculated mass on the condylar head or neck (peripheral osteoma) or proliferate and replace the condyle (central osteoma).

Osteomas can cause symptoms depending on their location and size. They can be asymptomatic if the lesions tend to be small, solitary, slow-growing, and painless, and in these cases, they are only noticeable during routine examinations.[6] As these lesions have a progressive growth, they can cause trismus, limitation of mouth opening, progressive malocclusion with midline shift toward the nonaffected side, contralateral mandibular deviation, and facial asymmetry and be painful.

In the last 15 years, only nine cases of mandibular condyle osteoma have been documented in the literature [Table 1]. PubMed and Cochrane Library were used as a source of research.

Table 1 Cases of mandibular condyle osteoma documented in the last 15 years

Case number	Reference	Age (year)	Gender	Site	Clinical manifestation	Treatment
1	Heitz *et al*., 2018[10]	49	Female	Right condyle and ramus, temporal area	Swelling of the right hemiface in the temporomandibular region Painless at palpation Limited mouth opening with a maximum amplitude of 10 mm	Removal of the lesion, piezosurgery, myofascial strip pad of the temporal muscle
2	De Souza *et al*., 2017[6]	67	Female	Right condyle	Asymmetry in the left lower jaw Laterognathism to the left side Limited mouth opening Pain in the right TMJ during maximum mouth opening and palpation	Immediate reconstruction using a total TMJ prosthesis
3	Nojima *et al*., 2014[11]	46	Female	Left condyle	Facial asymmetry Pain on chewing	Tumor resection including left mandibular condyle, reconstructed with an artificial articular head made of silicone
4	Almeida and de Oliveira Filho 2011[3]	45	Male	Right mandibular condyle	Face deformity Malocclusion Limited mouth opening (25 mm)	Conservative surgical excision
5	Cogburn *et al*., 2008[12]	55	Male	Left condyle	Dental malocclusion Significant trismus Inability to chew Open bite on the left side with significant malocclusion	Endoscopic approach with resection of the osteoma and recontouring of the condyle
6	Yonezu *et al*., 2007[1]	5X	Male	Lateral aspect of the left condyle	Pain Interincisal mouth opening 15 mm	Tumor resection
7	Ortakoğlu *et al*., 2005[13]	22	Male	Condyle	Mandibular deviation Malocclusion	Condylectomy
8	Mancini *et al*., 2005[14]	19	Female	Condyle	Facial asymmetry Canting of the occlusal plane and a posterior unilateral crossbite	Condylectomy
9	Siar *et al*., 2004[9]	32	Female	Left condyle	Restriction of mouth opening	Condylectomy

TMJ=Temporomandibular joint

By analyzing the reported cases, we inferred that the majority of osteoma cases involving the mandibular condyle occurred during the fourth and fifth decades of life (mean age 42.8 years), with a slight female predilection (female-to-male ratio 1.25:1).

Facial asymmetry, malocclusion, and limited mouth opening were the most common clinical findings.

Surgical intervention with removal of the lesion was the preferred treatment.

Radiographically, osteomas appear as circumscribed masses similar in density to normal bone. At their centers, these masses may exhibit a mixed radiolucent-radiopaque appearance, depending on the number of marrow tissues present. Small endosteal osteomas are difficult to differentiate from foci of condensing osteitis, focal chronic sclerosing osteomyelitis, or idiopathic osteosclerosis.[2] Radiographic images, such as panoramic radiographs, CT, magnetic resonance imaging, and scintigraphy, are utilized for diagnosis. CT scan with three-

dimensional reconstruction is the most useful in locating the lesion, showing more details about the relationship between the osteoma and the adjacent structures. Activity in the lesion is diagnosed by scintigraphy.[15]

Histologically, osteoma is reported as either a normal-appearing dense mass of lamellar bone with minimal marrow tissues (compact osteoma) or trabeculae of mature lamellar bone, with intervening fatty or fibrous marrow (cancellous osteoma).[16]

There are no reports of osteomas undergoing malignant transformation.[9]

Patients with osteomas should be evaluated for Gardner syndrome,[9] which is defined as multiple osteomas of the jaws accompanied by colorectal polyposis with high malignant potentials,[16] anomalies involving soft and hard tissues, congenital retinal pigment hypertrophies, multiple impacted or supernumerary teeth, desmoids tumors, and epidermoid cysts. These patients may present with symptoms of rectal bleeding, diarrhea, and abdominal pain. The triad of colorectal polyposis, skeletal abnormalities, and multiple impacted or supernumerary teeth is consistent with this syndrome. Onset occurs at an average age of 16 years, with malignant transformation of the colorectal polyps approaching 100% by age 40.[9,17]

Differential diagnosis should include maxillary or mandibular exostoses, osteoid osteoma, and osteoblastoma. Exostoses are bony excrescences that occur on the buccal aspect of alveolar bone. These lesions are of reactive or developmental origin and are not thought to be true neoplasms. Osteoblastoma and osteoid osteomas are more frequently painful and may exhibit a more rapid rate of growth than osteomas.[9]

The treatment of this pathology is determined according to the clinical and radiological examination, the extension of the lesion, and the possible complications for the patient.

In general, symptomless osteomas do not necessarily require treatment. However, when symptoms such as functional disorders (limitation of mouth opening, malocclusion, and mandibular deviation) facial asymmetry and pain are also present, surgical treatment is indicated.[11,18,19,20,21]

The optimal treatment for osteomas consists of complete surgical removal at the base where it unites with the cortical bone,[9] without compromising the adjacent structures.

Reconstruction of TMJ is a complex surgical procedure, and it entails improved mandibular form and function and reduction of pain and disability.[6]

Total TMJ replacement using alloplastic prosthesis may provide satisfactory results in cases of functional alterations of the TMJ due to the presence of tumors with irreversible joint damage.[6]

For the treatment of this case, we preferred surgical excision, with a conservative removal of the lesion, TMJ arthroplasty, and TMF interposition.

In the present case, resection of the right mandibular condyle osteoma resulted in the restoration of facial symmetry and a greatly improved molar occlusal relationship. The results were stable at 12-month follow-up.

Conclusions

Osteomas of the mandibular condyle are considered extremely infrequent lesions.

CT scan is the examination of choice for surgical planning purposes. The treatment of this pathology will be determined according to the clinical and radiological examination, the extension of the lesion, and the possible complications for the patient.

Symptomless osteomas do not necessarily require treatment. When symptoms such as functional disorders, facial asymmetry, and pain are present, surgical treatment is indicated. Surgery with complete lesion excision is the most adequate treatment, with low recurrence rates.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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