Limitation of mouth opening caused by osteochondroma of the coronoid process

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Background. Osteochondroma at the level of the coronoid process is unusual, causing a slowly progressive facial asymmetry and limitation of mouth opening. Histologically, it is a bone tumor covered by a thin capsule of cartilage. We present a literature review of cases published to date and present a new case in which osteochondroma originating in the coronoid process was associated with the formation of a cyst at the body of the zygoma, necessitating the reconstruction of the body of the zygoma.

Study design. A 55-year-old woman had a bone tumor in the right malar region, producing a limitation in mouth opening. After preoperative computerized tomography, we decided to excise the lesion and pseudocyst with the use of a combined subciliary and coronal approach, reconstructing the body of the zygoma with a cortical chip of calvarian bone.

Results. The patient regained normal mouth opening, without injury to the fronto-orbital branches of the facial nerve and no recurrence of the tumor to date.

Conclusions. Osteochondroma is a slow-growing tumor that causes progressive facial asymmetry and limitation of mouth opening. The treatment of choice for symptomatic osteochondromas is surgical resection. (Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2011;112:e64-e68)

Osteochondroma is the most common benign neoplasm of the skeleton. It is more frequently seen in long bones, but the most common locations in the facial skeleton and skull base are the mandible, maxillary sinus, and condyle. It is very uncommon in the coronoid process.1,2

Osteochondroma of the coronoid process is described as a solitary exophytic lesion of bone, which has varying amounts of cartilage tissue as a capsule. Sometimes both cartilage tissue and bone have an active growth, whereas in other cases both are diminished. Because of this variation, the injury is variously known as osteocartilaginous exostosis and osteochondroma. The tumor grows slowly and produces progressive limitation of mouth opening and facial deformity. Palpation of the zygomatic arch during jaw protrusion maximum aperture can be revealed by small vibrations, which are the shock of the tumor with the zygomatic arch. Using a stethoscope, noise or crackling at the arch can be heard if the coronoid process is rotated.1-4

Clinical examination, panoramic radiography, and computerized tomography (CT) with or without 3-dimensional reconstruction, are useful diagnostic tools, assisting in viewing the relationship between the zygomatic arch or malar bone and osteochondroma originating from the coronoid process.5

The facial asymmetry and limitation of mouth opening are indicative of the need for surgical removal of the lesion. The excision of the coronoid process with the tumor is the definitive treatment. The approaches may be intraoral, extraoral, or a combination. The reconstruction of bone defects in the zygomatic arch or malar bone can be performed with the use of calvarian bone grafts, as we have previously reported.5,6 The rate of recurrence after excision of the tumor is very low, ~2%.7 Mouth opening exercises promote postoperative rehabilitation.5

CASE REPORT

A 55-year-old woman attended the Oral and Maxillofacial Department, presenting a solid tumor in the right malar region. The lesion had been growing slowly and progressively over a 20-year period, producing a limitation of mouth opening and slight facial asymmetry (Fig. 1). Clinical examination of the patient revealed a prominence at the level of the zygoma body produced by the tumor. There was a mechanical trismus with a mouth opening of ~2 cm, slight facial asymmetry, and no facial pain on examination. Additional tests were requested: a panoramic radiograph, a Waters x-ray, and a 3-dimensional CT reconstruction, revealing an osteogenesis image located inside the zygomatic arch
and with continuity to the right coronoid process of the mandible (Fig. 2).

Having completed a preoperative study, we performed a surgical excision using a combined coronal and subciliary approach. After viewing the tumor, we performed an osteotomy at the zygomatic arch and a coronoidectomy including the lesion and the pseudocyst originating in the body of the zygoma. We repositioned the zygomatic arch and reconstructed the defect produced by the pseudocyst with a bone graft from the calvarian bone (Fig. 3).

The pathology of the lesion showed a polypoid exophytic lesion formed by thick bone trabeculae surrounded externally by a layer of hyaline cartilage and a superficially dense band of tissue, consistent with osteochondroma (Fig. 4).

In recovery, the patient had no involvement of the frontoorbital branches of the facial nerve, and normal mouth opening was restored over the following 2 years (>4 cm). There had been no recurrence of the tumor at the time of writing (Fig. 5).

**DISCUSSION**

Osteochondroma is the most common benign tumor in persons between 10 and 30 years of age. It accounts for ~20%-50% of all benign tumors and 10%-15% of all bone tumors. It is more commonly located at the level of the metaphysis of long bones. However, osteochondroma is rare at the level of the facial bones and skull base. It has been reported in the maxillary sinus and in different parts of the mandible, such as the condyle, ramus, body, and symphyseal region. It is a sessile lesion composed of bone covered with a cartilaginous capsule.3,7-10

Osteochondroma originating at the level of the coronoid process was first described in 1943 by Shackleford. Epidemiologically, the lesion usually occurs in young men <40 years old. It grows insidiously, and the most common symptom is the limitation of mouth opening with facial deformity (75% of patients). Other symptoms are remodeling, destruction, or expansion at the zygoma and/or zygomatic arch or pain with mouth opening. Over the long term, a new joint may develop between the coronoid process and zygomatic arch (Jacob disease), leading to a loss of mouth opening.2,5,7,11

In their review of published cases up to 1993, Kercher et al.3 included 30 cases, with age of onset ranging from 10 to 73 years and with 2 peaks of incidence: in the second and the fourth decades of life. Over 60% of the patients had facial deformity due to the involvement of peritumoral tissues. In >60% of the cases, the lesion had a sessile form.

Normally, the etiology of mandible hypomobility is related to an alteration in the mobility of the temporomandibular joint and/or diseases affecting the muscles...
of mastication. However, the alteration of the coronoid process should always be considered as a possible cause. Among the causes that may lead to a limitation of mouth opening and involving the coronoid process are muscle retractions after neurosurgery, coronoid hyperplasia, osteomas and osteochondromas, fibro-osseous ankylosis of the coronoid to the zygomatic arch, and pseudocampylodactyly syndrome.4,12

The pathogenesis of osteochondroma is not known, but there are several theories. Weinmann and Shichen have reported that the continued activity of the temporalis muscle tendon inserted at the level of the coronoid process can result in a hyperplastic growth of embryonic cartilage cell differentiation, presenting as endochondral ossification and causing osteochondroma. Lichtenstein suggests that the periosteum has pluripotency to produce cartilage and bone tissue. Kurita’s theory of abnormal development argues that osteomas of the coronoid process are sequels after a total ossification of cartilage. In some cases, both bone and cartilage show activity, causing resorption of the adjacent cortical bone. In other cases, the lesions are relatively quiescent. Some authors believe that the osteochondroma is a reactive exostosis, and others say that is a true neoplasm.1,2,5,10,11

Histologically, the tumor has a central region surrounded by endochondral ossification cartilage. The chondrocytes migrate from the periphery to the center of the lesion and produce bone tissue.7,9,13
Diagnosis is based on both clinical and imaging tests. Panoramic radiography or Waters projection can be used; CT, with or without 3-dimensional reconstruction, is the imaging modality of choice. CT visualizes the deformity of tissues and the size of the lesion and allows its shape and composition (the lesion appears as a radiolucent mass at the central level with a thin layer of cartilage that covers it), the location, and its relationship with the neighboring structures to be seen. A percutaneous biopsy of the tumor can also be used for histologic confirmation.

The differential diagnosis of osteochondroma of a pathologic enlargement of the coronoid process that causes a decrease in the opening includes rare entities such as unilateral coronoid hyperplasia due to hormonal changes, trauma or hereditary syndromes, and osteomas (which don’t have a capsule cartilage). Solitary osteochondroma may show sarcomatous changes in a small percentage of cases (1%). If the osteochondroma is part of an autosomal dominant syndrome, osteochondromatosis, the risk of sarcomatous transformation grows to 11%.7,11

The goal of treatment is to restore the normal range of acceptable oral opening. Surgical excision of the lesion, including the coronoid process, is the standard treatment. Recurrence is unusual. Facial asymmetry and/or limitation of mouth opening indicate the need for removal of the tumor. The surgical approach can be performed intraorally or extraorally.1

Almost all reported tumors until 1961 were resected by transzygomatic approach, with or without osteotomy of the zygomatic arch. Using this surgical approach, the upper branches of the facial nerve can be damaged and the possibility of producing an unsightly scar is increased. Therefore, the intraoral approach, if it allows a complete tumor resection, should be the initially preferred surgical approach. The intraoral approach avoids a skin scar and the risk of facial nerve injury, although sometimes herniation of the Bichat fat pad can make it hard to display the tumor.8,10,11

The alternative is the extraoral approach, through the submandibular or coronal incisions. The disadvantages of the submandibular approach are the risk of damage to the mandibular nerve and submandibular scarring, so it is rarely used. When the osteochondroma is very large and is stuck on the zygomatic arch, or mouth opening is minimal, the removal must be performed by coronal approach. This has the advantage of optimizing the visualization of the lesion. It produces an acceptable scar in the hairline and allows the transfer of the temporalis muscle flap and the removal of calvarian bone grafts in the same operation.

The coronal incision approach, described by Hernández Alfaro,8 is used in the following situations:

- When surgical excision using the intraoral approach is rendered difficult by the size and/or position of the lesion.
- When temporomandibular joint pathology is concomitant.
- In bilateral cases.

For Hernández Alfaro, the approach through a coronal and transzygomatic incision is currently the surgical approach of choice. It gives an excellent exposure of the surgical field. It facilitates the reconstruction of the zygomatic arch or malar bone if they are eroded or destroyed by cortical grafts of calvarian bone and...
avoids damage to the fronto-orbital branches of the facial nerve and skin scars.1,5,7,8,11

After surgical excision of the tumor, immediate postoperative rehabilitation is done through physiotherapy. Mouth opening exercises, for up to 6 weeks, are important to ensure therapeutic success.5,12

CONCLUSIONS
Osteochondroma is one of the most common benign tumors of the axial skeleton, but it is uncommon at the level of the facial bones. When originating in this anatomic region, the tumor mainly affects the coronoid process and mandibular condyle. Growth of the tumor is usually slow, gradually producing facial asymmetry and limitation of mouth opening. Radiographically, the lesions are radiolucent, and CT with or without 3-dimensional reconstruction imagery is the test of choice. The main treatment for symptomatic osteochondromas is surgical excision, which can be performed intraorally, extraorally, or using a combined approach. Recurrence and sarcomatous degeneration of the tumor are very unusual.

REFERENCES

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