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CASE REPORT

Soft-tissue osteoma in the pterygomandibular space: report of a rare case

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Osteoma is a slow-growing, benign and uncommon neoplasm located primarily in the region of the maxillofacial skeleton. An extraskeletal soft-tissue osteoma is exceedingly rare. Here, we report a case of soft-tissue osteoma occurring in the pterygomandibular space in a 66-year-old woman. The patient complained of a hard mass superior to the left posterior teeth. Clinical examination did not reveal any extraoral swelling, facial asymmetry or difficulty in mouth opening, and the regional lymph nodes were non-palpable. CT images revealed well-circumscribed dense, radiopaque masses located between the left side of the maxilla and the lateral plate of the left pterygoid process and the left ramus of the mandible. Intraoperatively, the masses were completely surrounded by soft tissues with no attachment to the bone. Histological examination indicated the diagnosis of cancellous osteoma.
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Keywords: bone-forming neoplasm; extraskeletal osteoma; soft-tissue osteoma; pterygomandibular space

Introduction

Osteoma is a slow-growing, benign and uncommon neoplasm located primarily in the region of the maxillofacial skeleton. It is an osteogenic lesion characterized by proliferation of compact or cancellous bone. Osteoma can be central, peripheral or extraskeletal. Central osteomas arise from endosteum, peripheral osteomas arise from the periosteum and extraskeletal soft-tissue osteomas usually develop within a muscle.[1,2]

An extraskeletal soft-tissue osteoma is exceedingly rare; most previously reported cases have occurred in tongue and skin[3–6] with very few in extremities such as the hip,[7] thigh[8] and hand.[9] Here, we report a case of soft-tissue osteoma occurring in the pterygomandibular space.

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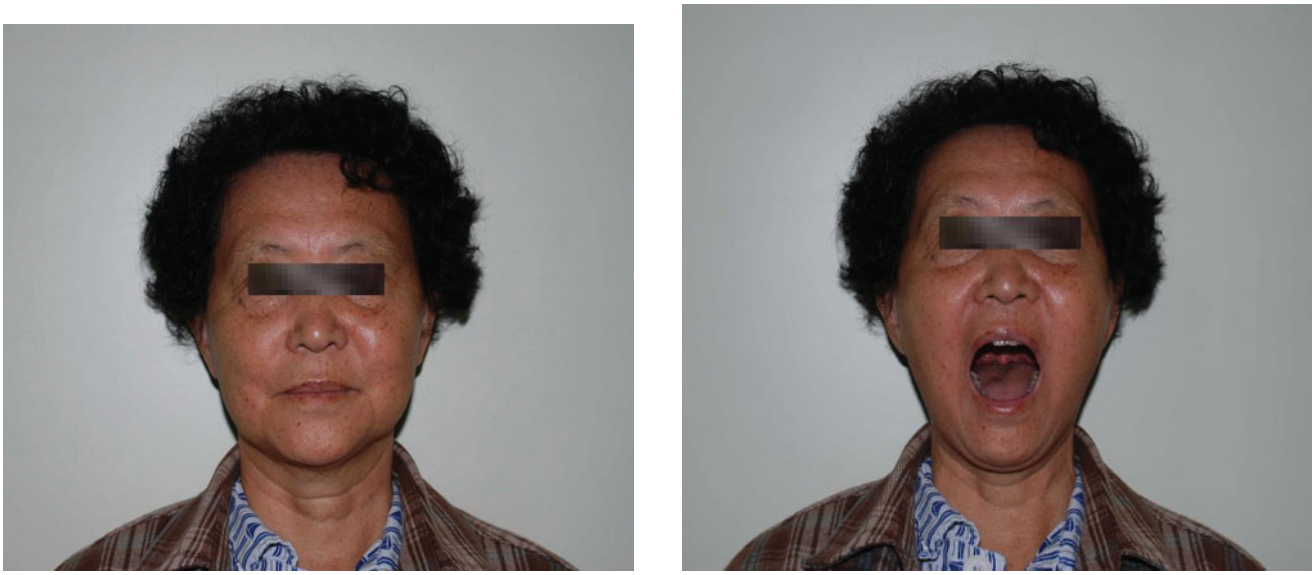
Case report

A 66-year-old woman was referred to the Department of Oral and Maxillofacial Surgery, Peking University School and Hospital of Stomatology, Beijing, China, with a complaint of a hard mass superior to the left posterior teeth that was without pain or swelling and did not cause difficulty in mouth-opening. The patient discovered the mass by chance when food particles became trapped in the space between the hard mass and the cheek. According to the patient, there was no history of trauma or inflammation to the area.

Clinical examination did not reveal any extraoral swelling, facial asymmetry or difficulty in mouth opening (Figure 1) and the regional lymph nodes were non-palpable. Intraoral examination revealed a well-circumscribed, movable, bony hard mass located at the buccal side of the maxillary left second premolar and extending to the anterior rim of the pterygomandibular fold. The mass was approximately 5×1 cm in size and covered with normal oral mucosa. Results of blood and serum chemistry analyses were within the normal ranges.

A panoramic radiograph showed two dense radiopaque masses with well-circumscribed margins in the



a b

Figure 1 Picture of the patient with (a) mouth closed and (b) mouth open. No facial asymmetry, swelling or difficulty in opening the mouth were observed

left side of the jaws. One was in front of the ramus and the other was superior to the sigmoid notch and the coronoid process of the mandible (Figure 2).

The CT scanned images revealed that the two masses were well-circumscribed and located between the left side of the maxilla and the lateral plate of the left pterygoid process and the left ramus of the mandible. The masses had no connections with any bone and the surface of the upper mass was lobulated (Figure 3a). Figure 3b showed that the density of the two masses was not homogeneous and that the posterior wall of the sinus was deformed. The masses looked like two collections of many small round and oval radiopaque masses.

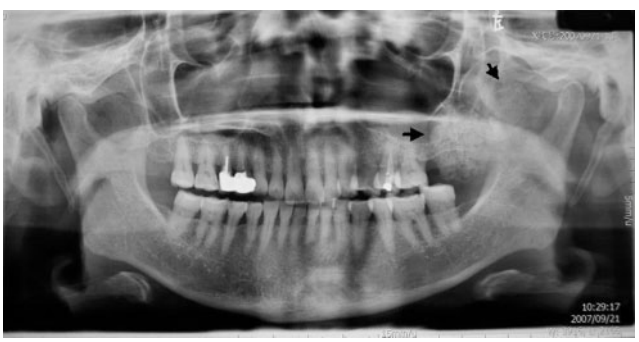


Figure 2 Two dense radiopaque masses with well-circumscribed margins were shown in the left side of the jaws. One was anterior to the ramus of mandible and the other was superior to the sigmoid notch and the coronoid process (arrows)

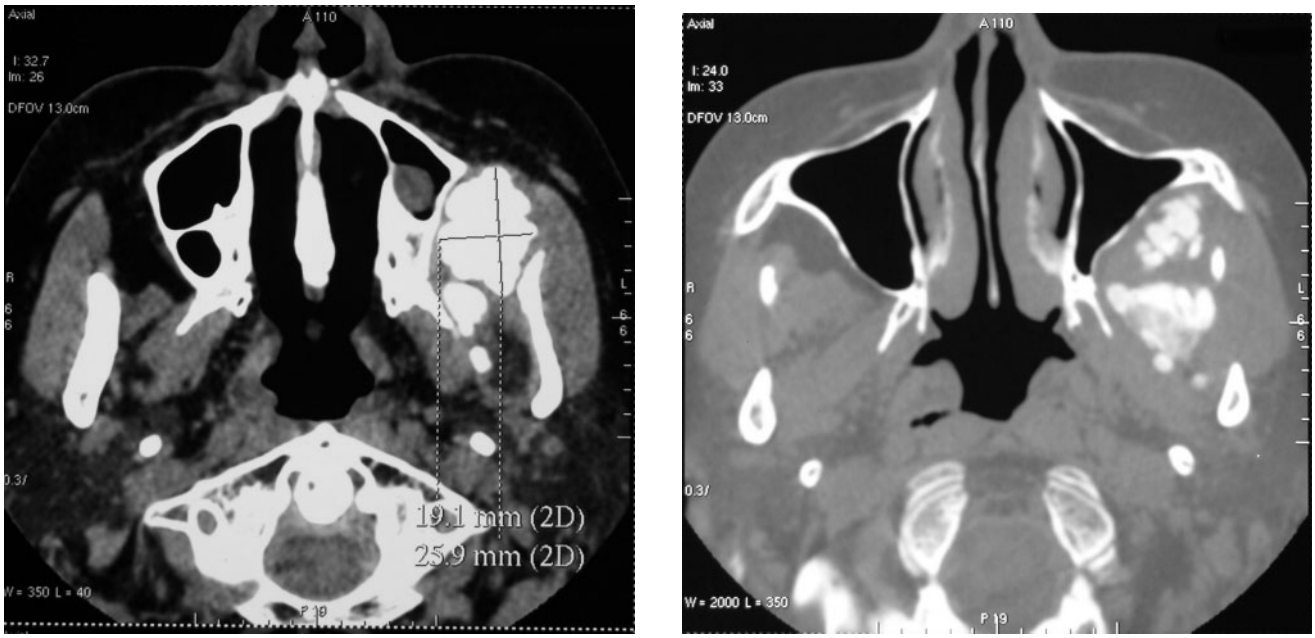
Intraoperatively, the masses were completely surrounded by soft tissues with no attachment to bone. The post-operative course was uneventful. The radiograph of the specimens showed the well-defined borders and many small radiopaque masses within the specimens (Figure 4). A three-dimensional image of the osteoma is shown in Figure 5.

Microscopic examination revealed round-shaped mature bones with prominent cancellous components and marrow function that were separated by fibrous connective tissues (Figure 6). The histological feature indicated the diagnosis of cancellous osteoma.

Discussion

Apart from osteomas occurring in the maxillofacial sinuses, approximately 98 solitary osteomas in the mandible[10,11] and 9 in the maxilla[12,13] have been reported in the English literature over the past 80 years. Most of the reported osteomas are of the peripheral type. For the so-called extraskeletal type, osteomas have been found most often in the tongue and skin.[3,6] In our search of the English literature, we did not find reports of osteoma occurring in soft tissues other than in the tongue and skin of the maxillofacial region. Thus, the present case is the first osteoma originating from the soft tissue in the pterygomandibular space.

The exact aetiology of benign bone-forming lesions of soft tissue remains unclear. Various hypotheses have been set forth. Some investigators consider the osteoma a true neoplasm, and others classify it as a develop-



a **b**

Figure 3 CT examination showed that (a) the two radiopaque masses had no attachment to the surrounding bones and (b) the density of the two masses was not homogeneous

mental anomaly, especially those occurring in the ''classical posterior tongue midline position''. The possibility of a reactive mechanism, triggered by trauma and infection, has also been suggested. The precise location of an osteoma is usually in close proximity to regions of muscle attachment, suggesting that muscle traction may play a role in its development.[7,10,12] It has also been proposed that myositis ossificans, soft-tissue osteochondroma and soft-tissue osteoma form a spectrum of changes related to soft-tissue injury. However, this dose not agree with Kasper et al, who described that central cystic degeneration in older myositis ossificans was absent in osteoma.[7]

To be classified as soft-tissue osteoma, a lesion should fulfil the following criteria: arise spontaneously and not be secondary to trauma or inflammation, not be of developmental origin and it should grow

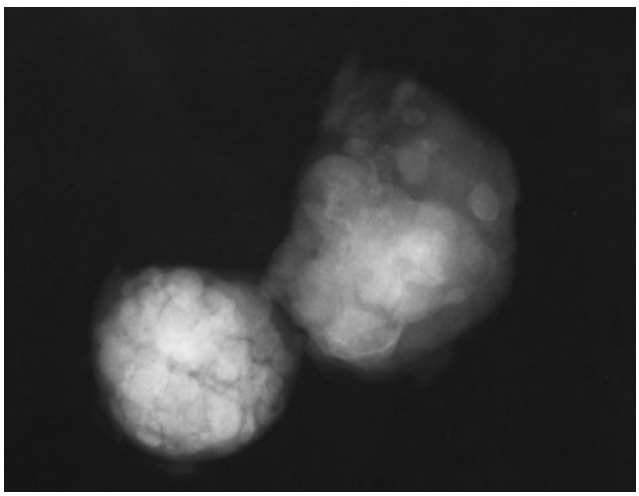


Figure 4 Radiograph of the specimens, showing the well-defined borders and many small radiopaque masses within the specimens

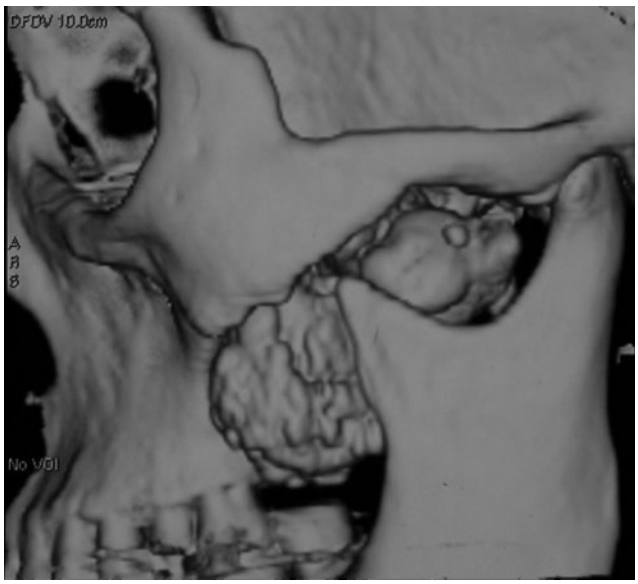


Figure 5 A three-dimensional image of the osteomas

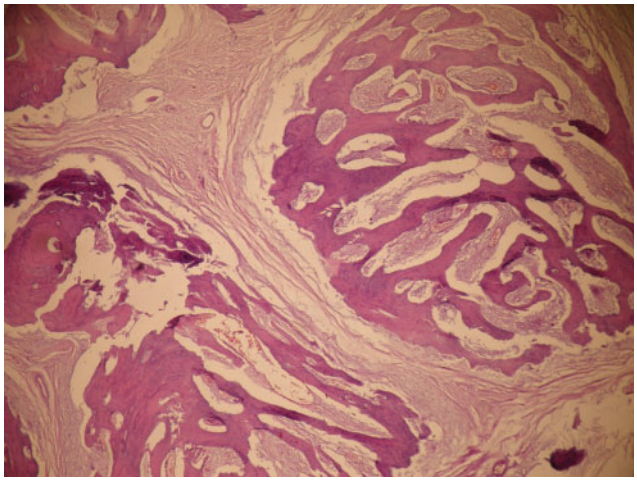


Figure 6 Microscopically, the round-shaped mature bones with prominent cancellous component and marrow function were separated by fibrous connective tissues (original magnification $\times 40$)

unattached to the periosteum or periarticular structures.[7] The present case appears to be such a lesion.

The main differential diagnosis of the present case should be osteochondroma – a lesion presenting a similar radiographic appearance to osteoma, but with tissues of cartilage observed microscopically. The lesion is often associated with the mandibular condyle.

Besides osteoma, the osseous lesions most often occurring in soft tissue are osteosarcoma, localized myositis ossificans, progressive myositis ossificans, fibro-osseous pseudotumour and osteoblastoma.[14] Osteosarcoma and osteoblastoma are lesions manifesting with severe pain and rapid growth. Localized myositis ossificans and progressive myositis ossificans in the maxillofacial region are often associated with difficulty in mouth-opening and are radiographically characterized by the calcification of a masticatory muscle. Fibro-osseous pseudotumour usually occurs in the soft tissue of the fingers, particularly in the region of the proximal phalange. No fibro-osseous pseudotumour was reported in the maxillofacial region.

The treatment of osteoma is surgical excision. Recurrence after surgical procedure is rare and there are no reports of malignant transformation.

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