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Heterotopic Ossification

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Heterotopic ossification (HO) is the formation of mature bone in soft tissue where bone normally does not exist. Although HO is among the most common complications after orthopedic surgery, it is not familiar to oral and maxillofacial surgeons. Here we report rare cases of HO. When a patient presents atypical osseous lesions, HO as well as similar lesions such as osteoma, osteochondroma should be considered in the provisional diagnosis. Three-dimensional reconstruction of preoperative computerized tomography imaging improves surgical success.

Key Words Heterotopic ossification · Formation of mature bone · Complications after orthopedic surgery · Atypical osseous lesions · Three-dimensional reconstruction.

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Introduction

Heterotopic ossification (HO) is defined as the formation of lamellar bone inside soft-tissue structures where bone normally does not exist. Heterotopic ossification (HO) is classified into two forms. The acquired form is usually caused by trauma or skeletal surgery, and the hereditary form is of very low prevalence. The acquired form is usually caused by trauma or skeletal surgery, and the hereditary form is of very low prevalence.

Very few cases of HO have been reported in the head and neck region. Here we report cases of HO in the masseteric space & zygoma or maxilla region

Case Reports

Case I

A twenty-three-year-old female with no specific medical disease visited our clinic with a complaint of palpable mass on right

ramus area. She said that there was no history of trauma as long as she could remember.

Clinical examination revealed a small palpable bony mass on right master area which was also observed on panoramic view (Fig. 1). On computed tomography (CT), a round hard tissue mass approximately 1 cm in diameter on right masseter area was confirmed (Fig. 2). Then three-dimensional reconstruction of preoperative computerized tomography imaging was performed (Fig. 3).

Based on clinical and radiographic examination, the clinical diagnosis was heterotopic ossification, masseter, Rt., which we planned to excise under general anesthesia. The surgical procedure for mass excision was performed intraorally.

Case II

A fifty-seven-year-old female with no specific medical disease visited our clinic with a complaint of trismus. She said that there was no history of trauma as long as she could remember. She had

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Fig. 1. Panoramic image at 1st visit (Case I).

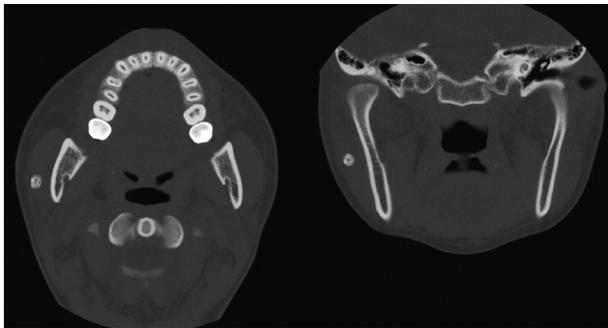


Fig. 2. CT image showing a round hard tissue mass approximately 1 cm in diameter in masseteric space (Case I).

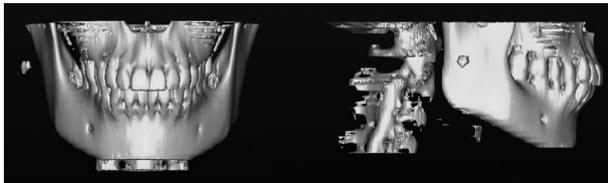


Fig. 3. 3D reconstruction image using SimPlant® software (Case I).

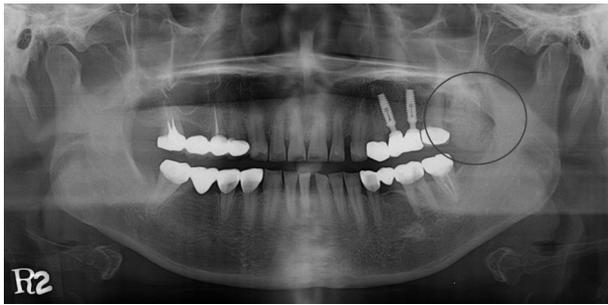


Fig. 4. Panoramic image at 1st visit (Case II).

an implant surgery on the left molar area about 15 months before.

Clinical examination revealed an elevated bony mass on the left buccal mucosa which was also observed on panoramic view (Fig. 4). On computed tomography (CT), a round pedunculated hard tissue mass approximately 3 cm in diameter arising from the zygomaticomaxillary suture and extending to the masseter anterior area was confirmed (Fig. 5). Then three-dimensional reconstruction of preoperative computerized tomography imaging was performed (Fig. 6).

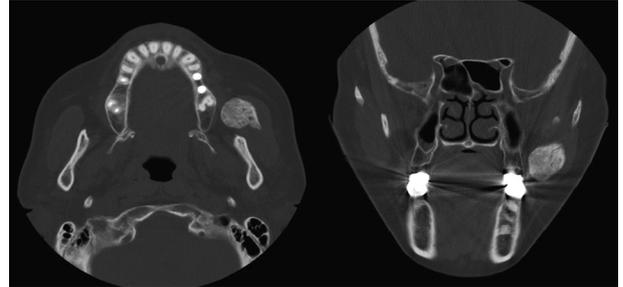


Fig. 5. CT image showing a round pedunculated hard tissue mass approximately 3 cm in diameter in masseteric space (Case II).

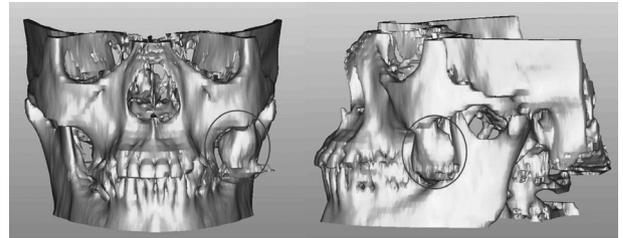


Fig. 6. 3D reconstruction image using SimPlant® software (Case II).

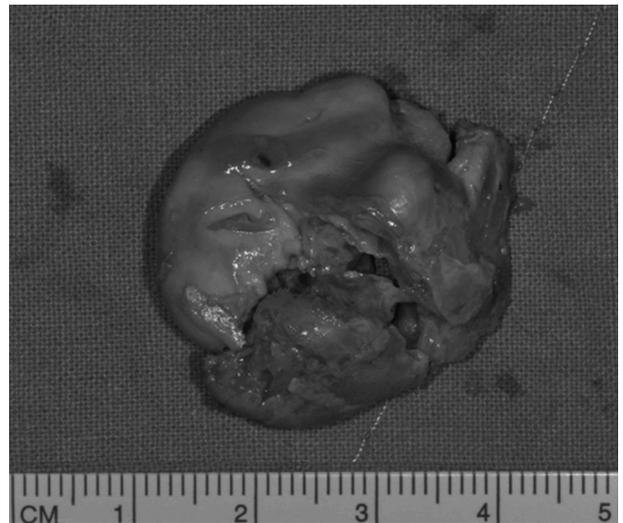


Fig. 7. Excised osseous mass circumscribed with cartilaginous tissue (Case II).

Based on clinical and radiographic examination, the first clinical diagnosis was osteochondroma, which we planned to excise under general anesthesia. The surgical procedure for mass excision was performed intraorally, with mucosal incision on palpable mass. The mass was exposed following blunt dissection and completely removed. It was well circumscribed, with cartilaginous tissue on the surface and softer than bone (Fig. 7).

Discussion

Osteochondroma, one of the most common benign bone tu-

mours, is cartilage-capped bony outgrowth characterized as a well-defined mass with no adjacent tissue invasion and with immature bone pattern and density. In second case, our first impression was osteochondroma due to its similar shape and radiographic features.

HO, the formation of mature bone in soft tissue, usually occurs in soft tissue such as muscle as a result of trauma or surgery. In our case, the bony mass was mainly located in the masseteric space, extending to the zygomaticomaxillary suture in second case. Histologically, it showed normal mature bone formation surrounded by osteoid. Consultation with a pathologist specializing in bone tumour yielded a final diagnosis of heterotopic ossification. We regard this as a very rare case of HO in the head and neck region.

The pathophysiology of HO is not yet precisely understood. Transformation of primitive cells of mesenchymal origin into osteogenic cells is assumed to be a brief process, but its specific pathway is not known.

Heterotopic ossification is one of the most frequent complications after orthopedic surgery, and is reported to follow implantation of internal spinal devices. In our case, a dental implant might be relevant or coincident. Oral and maxillofacial surgeons, although not familiar with HO, should know that it can occur in oral and maxillofacial regions. When a patient presents atypical osseous lesions, HO as well as similar lesions

such as osteoma, osteochondroma, and low grade osteosarcoma should be considered in the provisional diagnosis.

In addition, three-dimensional reconstruction of preoperative computerized tomography imaging improves surgical success.

References

1. Shehab D, Elgazzar AH, Collier BD. Heterotopic ossification. *J Nucl Med* 2002;43:346-353
2. Patil SG, Siddiqua A, Joshi UK, et al. Heterotopic Ossification: An Unusual Presentation. *Case Reports in Dentistry* 2012;2012:4
3. Shankly PE, Hill FJ, Sloan P, et al. Bizarre parosteal osteochondromatous proliferation in the anterior maxilla: report of a case. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 1999;87:351-356
4. Malhas AM, Sumathi VP, James SL, et al. Low-Grade Central Osteosarcoma: A Difficult Condition to Diagnose. *Sarcoma* 2012;2012:7
5. Urist MR, Nakagawa M, Nakata N, et al. Experimental myositis ossificans: cartilage and bone formation in muscle in response to a diffusible bone matrix-derived morphogen. *Arch Pathol Lab Med* 1978; 102:312-316
6. Mori M, Kasai T, Shrestha P, et al. Heterotopic ossifying tumors in head and neck and heterotopic ossification and calcification. *Journal of Oral and Maxillofacial Surgery, Medicine, and Pathology* 2012; 24:222-236
7. Chalmers J, Gray DH, Rush J. Observations on the induction of bone in soft tissues. *J Bone Joint Surg Br* 1975;57:36-45
8. Hsu JE, Keenan MA. Current review of heterotopic ossification. *Journal of Orthopaedics* 2010;20:126-130
9. Tian NF, Wu AM, Wu LJ, et al. Incidence of heterotopic ossification after implantation of interspinous process devices. *Neurosurg Focus* 2013;35:E3