

A Rare Case of Solitary Osteochondroma at the Temporomandibular Joint: A Case Report

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Osteochondroma is a bone tumor with cartilaginous growth potential that generally appears near the growth plate of long bones in areas such as hip, knee, and shoulder joints, related to the nature of endochondral ossification and it is known a common benign bone tumor. However, it has been very rare in craniofacial region possibly because craniofacial bone is largely formed by intramembranous ossification. Moreover, reports on the solitary type of osteochondroma in mandibular condyle has been extremely rare. Osteochondroma in mandibular condylar may show various symptoms similar to general temporomandibular joint disorders (TMDs), such as pain in the condylar area during mouth opening, internal derangement, facial asymmetry or posterior open bite. Therefore, it can be disregarded for a long time period without any adequate treatment. Surgical excision has been the treatment option for the solitary osteochondroma with very low recurrence rate reportedly. In this case report, a rare case of solitary osteochondroma developed in unilateral mandibular condyle is presented with emphasis on differential diagnosis with general TMDs.

Key Words: Mandibular condyle; Mouth opening deviation; Osteochondroma; Solitary type; Temporomandibular joint disorder

Introduction

Osteochondroma is generally known as osteocartilaginous exostosis and one of the most common benign tumors of long bones, accounting for about 35%~50% of benign tumors and 8%~15% of primary bone tumors. However it is very rare in the facial

skeletal bone especially as a solitary type^{1,2)}. Osteochondroma is formed as a result of endochondral ossification and increase in size via endochondral ossification from an abnormal cartilaginous epiphyseal growth plate tissue. In the craniofacial region, the most common sites of occurrence are the mandibular coronoid process and the condyle of mandible¹⁾.

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Possibly due to the nature of the endochondral ossification during embryonic growth, the temporomandibular joint (TMJ) tends to be one of the most frequent site for osteochondroma in the craniofacial region³. Osteochondroma may develop from any endochondral bone structures during childhood and adolescence^{4,5}.

Radiographically, these lesions are easily visualized on plain radiography and computed tomography (CT) because of the distinct borders^{6,7}. Osteochondroma appears as a radiopaque lesion in the radiographic image that shows relatively clear boundary. Since osteosarcoma or chondrosarcoma occurs commonly among the malignant tumors, differential diagnosis of them is crucial⁸. A scintigraphy has also been suggested to detect the intense uptake in the lesion⁹.

Morphologically it may present as condylar hyperplasia or increase in the condylar length for which traditional therapies include condylectomy, surgical excision, and TMJ reconstruction, and relapses have been rarely reported¹⁰. The outline of tumor by CT imaging was clearly outlined and subsequent histopathology confirmed it as osteochondroma¹¹.

Osteochondromas occur as solitary tumor or as multiple exostoses. Multiple exostoses can occur sporadically but more commonly results from a hereditary disorder with autosomal dominant transmission known as hereditary multiple exostosis (also referred to as osteochondromatosis, Bessel-Hagel syndrome, or diaphyseal aclasis)¹². Solitary osteochondroma (osteocartilaginous exostosis or exostosis) has been rare and was first described by Reid¹³ in 1843, and it accounts for 10% of all primary osseous tumors¹⁴. Solitary osteochondroma in the condylar area has been even more rare and only a few cases has been reported in the literature.

Osteochondroma in the condyle may show various clinical signs and symptoms such as pain in the condylar area during mouth opening, internal derangement in the TMJ, facial asymmetry and/or posterior

open bite. These features can be often regarded as routine temporomandibular disorders (TMDs), however, common treatment modalities for regular TMDs would not be appropriate for the treatment of the disease. Management of osteochondroma of the mandibular condyle includes condylectomy or locally resection of the lesion and condyle replacement, usually with a costochondral graft or total joint prosthesis^{1,15}. In our case, because of the solitary type of the osteochondroma, simple excisional removal of the mass with dissection around soft tissue through preauricular approach was conducted. In this report, we present a very rare case of solitary osteochondroma developed in the space of the mandibular condyle.

Case Report

A 65-year-old male was referred due to discomfort of right side TMJ. He complained of chronic pain and some annoying joint noise when his mastication of daily diet, however he did not present mouth opening limitation. He also had chin deviation to the right side since two years ago and had malocclusion with noncontacting maxillary and mandibular molars. There was no trauma history in the region (Fig. 1). Before the referral to our hospital, he was diagnosed as having regular TMDs and received routine TMD treatments such as stabilization splint



Fig. 1. Pre-operative clinical photograph.

and physical therapy in the local clinic. Despite of those treatments for two months, his problems were not improved.

According to the radiological examination of this patient, a solitary radiopaque lesion was found and initially assumed to be a benign lesion in the posterior space of the right mandibular condyle (Fig. 2).

In the CT and magnetic resonance imaging (MRI) figures, the outline of the chondro-osseous lesion

was clearly demarcated and any adhesion to mandibular condyle or the disk was not observed. When compared to the image of normal condylar head of the left TMJ, it appeared to be more radiopaque in CT and less bright signal in T2 MR image.

The mass was measured as $2.3 \times 1.8 \times 0.6 \text{ cm}^3$ in volumn (Fig. 3, 4). Although this mass was preliminarily diagnosed as a benign tumor, *i.e.*, a solitary osteochondroma, surgical removal was chosen to confirm the diagnosis and to relieve the patient's discomfort.

Under general anesthesia, a routine surgical draping was performed and a local anesthesia (lidocaine

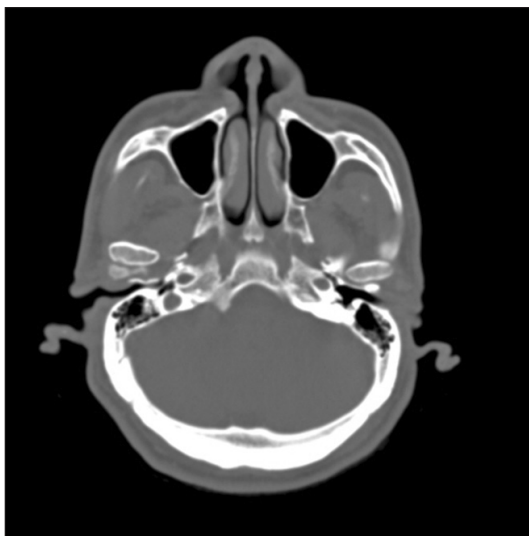


Fig. 2. Computed tomography image of horizontal plane. When compared to image of normal condylar head of the left temporomandibular joint, condylar displacement is observed on right side pushed by the solitary osteochondroma measured $2.3 \times 1.8 \times 0.6 \text{ cm}^3$ in volumn.

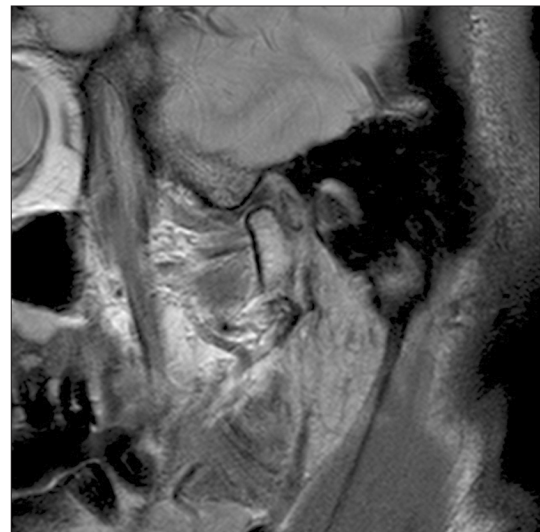


Fig. 3. Magnetic resonance image of the right condylar region.

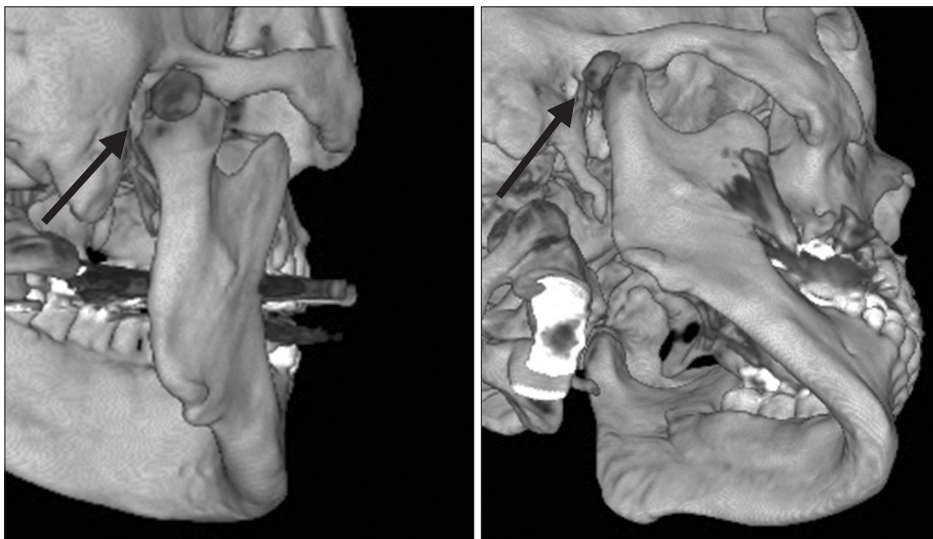


Fig. 4. Three-dimensional computed tomography image of facial bone (arrows: solitary osteochondroma).



Fig. 5. Isolated solitary osteochondroma during the operation.



Fig. 6. Excised tumor mass.

and 1:100,000 epinephrine) was given. Through a short preauricular incision along the patient's natural crease, the mass was exposed and removed with careful dissection.

Mouth opening exercise started at 3 days after surgery. At 1 month after surgery, chronic pain and mentioned discomforts disappeared and the mass did not recur during our observation period (Fig. 5).

The excised tumor was sent to the department of pathology and lining by cartilaginous tissue was observed. Its boundary partly adhered to adjacent soft tissues but was clearly separated from the soft tissue (Fig. 6). In the histopathological findings at low magnification of the tissue, we could find portion of the bone in the inner area and cap of cartilage in the outer area and we also observed endochondral ossification between the cartilage and the bone (Fig. 7). Accordingly, the mass was finally confirmed as the solitary type of osteochondroma that had occurred in the mandibular condylar space.

Discussion

In terms of etiology of the osteochondroma, Langenskiöld¹⁶⁾ insisted in 1967 that osteochondroma occurs via proliferation of undifferentiated cell layers near the epiphyseal area followed by inferior

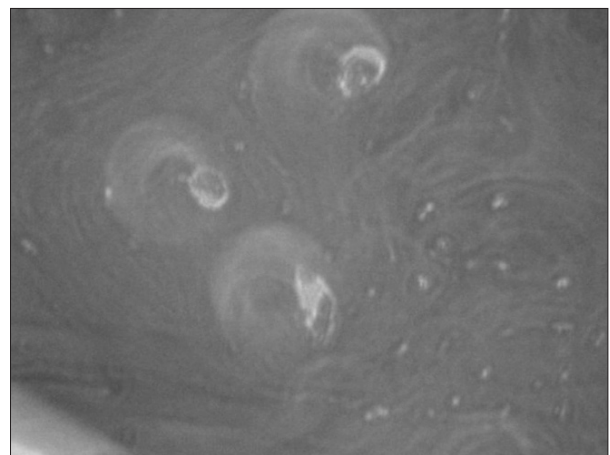


Fig. 7. Photomicrograph shows cartilaginous cap and endochondral ossification (H&E staining, x40).

displacement of proliferated cells onto near the metaphysis. This may explain rounded condylar head lesion but in the entire intramembranous bones such as maxilla, the pathophysiology may not be explained properly¹⁷⁾. In 1982, Kaneda et al.¹⁸⁾ reported that infection and trauma may serve as causative factors for the formation of osteochondroma in the mandibular condyle. Recently, Roychoudhury et al.¹⁹⁾ suggested that trauma may be an etiologic factor for osteochondroma of mandibular condyle, in accordance to others^{15,20)}. Therefore, it is believed that the stress in the regions of tendinous insertion

which may develop foci of cell accumulations with cartilaginous potential, may then lead to formation of osteochondroma²¹. In this case, the patient had no previous trauma history, however parafunctional habit of clenching and gum chewing were present. Further investigation is needed regarding etiology of the osteochondroma in association with parafunctional habits.

Solitary osteochondroma is a frequent lesion which account for 1%~2% of individuals undergoing extensive radiographic evaluation. The radiographic appearance of solitary osteochondroma in long bones is frequently pathognomonic. The lesion is composed of cortical and medullary bone protruding from and continuous with the underlying bone²².

Clinically the patient may present pain, limited mobility of TMJ, partial or complete hearing loss, asymmetry, bowing of lower border of mandible, prognathic appearance, malocclusion with ipsilateral open bite, lateral crossbite as well as anterior crossbite, or any combination of these clinical manifestation¹⁹. Therefore, these expression patterns need to be meticulously evaluated to rule out common TMD symptoms. For the diagnosis of osteochondroma-like lesions, two dimensional radiographies are not considered appropriate because of overlapping images of osseous structures¹². The CT images are useful to clarify the attachment zone of the osteochondroma, contributing to preoperative planning²³⁻²⁵. One may easily confirm the continuity of medulla and cortex of the osteochondrom from CT image for which CT is considered an optimal tool to demonstrate calcified cartilage^{3,26}. The CT image also allows diagnosis radiologically by showing medullary and cortical continuity between the bony tumor and normal anatomic structures^{12,27}.

In spite of the strengths of CT in the study of osteosarcomas²⁸, noncontrast CT scans and routine MRI have been suggested as the best to evaluate the cervical osteochondromas. As in our case, MRI is useful for several reasons. It can show cartilaginous caps

that are larger than 3 mm, for which, a CT scan may underestimate the size of the osteochondroma²⁸.

It is known that osteochondromas generally develop during growth of the skeleton and also it is widely agreed that osteochondromas hardly initiate after puberty^{4,5}. In most cases, the osteochondroma growth occurs in proportion to growth of skeletal bone²⁹. However, in the mandibular condyle, osteochondroma continues to grow³⁰, and this growth of osteochondroma in the mandibular condyle may be related to constant mandibular movement that stimulates the tendon³¹. Thus, parafunctional habits such as clenching or bruxism may also contribute to the development osteochondroma. In our case, the patient had severe clenching habit and other TMD symptoms which stimulate condylar soft tissues of the mandible.

Different from osteblastoma in long bones, osteochondromas in the jaw bones tend to occur mainly in females, and the onset of average age is older than 40 years⁷. As our case showed, osteochondroma contraction in the condyle of mandible often leads to cosmetic and functional abnormalities, for which surgical excision is needed for both function and esthetics^{10,32,33}.

In consideration of its extremely low recurrence rate and the nature of the benign tumor, the aim of treatment was surgical excision of the osteochondroma expecting the mandible to swing back to normal occlusion leading to the correction of facial midline³⁴. Radical excision including the surrounding periosteum has been strongly recommended to reduce the recurrence of osteochondroma^{9,35}. Large lesions with bulky cartilaginous caps, and lesions that continue to grow after skeletal maturity should be considered as evidence of possible malignancy¹². Afterwards, periodical observation of the present case is required for evaluation of the prognosis.

In conclusion, osteochondroma may occur in the TMJ, which needs careful differential diagnosis in patients with TMD symptoms or obvious jaw deformities.

Conflict of Interest

No potential conflict of interest relevant to this article was reported.

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