Case Report A ring-like soft tissue osteoma in the temporomandibular joint capsule: case report

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Abstract: Soft tissue osteoma is an extremely rare tumor. All previously reported cases occurred in the tongue and extremities. This report is of a case of soft tissue osteoma in the temporomandibular joint capsule in a 58-year-old woman. Computed tomography (CT) examination with three-dimensional reconstruction revealed a ring-like, well defined densely ossified mass in the right temporomandibular joint, but with no relation to the adjacent bony condyle or fossa. The mass was resected, and reconstruction was done by total alloplastic joint replacement. Intraoperatively, the lesion was embedded in the joint capsule, and completely not attached to nearby bone. The histological examination showed mature dense bone, osteoblast cells, adipose tissue, and hematopoietic tissue. Pathology confirmed the diagnosis of soft-tissue osteoma. The clinical presentation, anatomic features, histologic characteristics, and differential diagnosis of this lesion are discussed.

Keywords: Soft tissue osteoma, temporomandibular joint, ring-like

Introduction

Osteoma is a rare benign bone tumor that usually arises from the long bones. Soft-tissue osteoma is exceedingly rare benign tumor containing both cartilaginous and osseous components [1]. Soft-tissue osteoma has been reported in different parts of the body, but most often in the tongue and skin [2-4]. There are some reports of these tumors in the extremities, commonly around the knee, foot, ankle, hip, and thigh [1, 5-8]. Soft tissue osteomas consist of mature lamellar bone with regular osteocytes and no atypia. These lesions are slowly growing and separated completely from adjacent bone. Although the exact etiology is unknown, it has been proposed that soft-tissue chondroma, myositis ossificans, soft-tissue osteochondroma, and soft-tissue osteoma lie in a spectrum related to soft tissue injury. The radiologic and histologic appearances of these lesions differ according to the relative amount and appearance of osseous and cartilaginous components [1, 6]. Involvement of the smaller joints is seldom reported. Reviewing English language literature, temporomandibular joint (TMJ) involvement by soft tissue osteoma was

not reported before. Therefore, there is a lack of understanding of the exact nature of the disease, and difficulty in making an accurate diagnosis. Therefore, this is a case study of soft-tissue osteoma in the TMJ capsule of a 58-year-old woman.

Case report

A 58-year-old woman with 1-year history of chronic pain, swelling in the right temporomandibular joint, and limitation of mouth opening without preceding trauma to the orofacial region. A long history of unilateral chewing habits in the right TMJ was recorded. The patient had a medical history of hypertension and diabetes mellitus. This patient presented to our institution for evaluation, diagnosis, and treatment.

Clinical examination showed an obvious preauricular swelling with tenderness on the right side. Maximal inter-incisal mouth opening was decreased to 15 mm. On mouth opening, slight deviation of the mandible to the right side was found, still with a stable occlusal relationship. There were no joint sounds. All laboratory val-



Figure 1. Panoramic radiograph showing round mass involving the right TMJ region (red arrow).

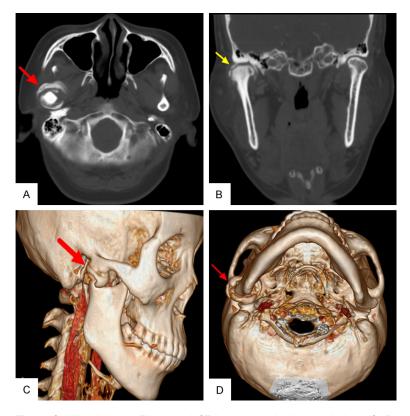


Figure 2. (A) Axial and (B) coronal CT images at the bone window, (C, D) three-dimensional reconstruction depicted a ring-like densely ossified mass in the TMJ with clear margin.

ues, including a complete blood cell count, calcium, phosphorous, alkaline phosphatase, blood urea nitrogen, creatinine, sedimentation rate, and rheumatoid factor, were within normal limits.

A panoramic radiograph (Figure 1) revealed a round mass related to the right TMJ. Axial and coronal CT scanss (Figure 2A, 2B) were obta-

ined, along with three-dimensional reconstruction (Figure 2C, 2D), demonstrating a ringlike mass surrounding the right condylar head. The lesion lacks the typical zoning pattern of myositis ossificans, with no direct communication with adjacent bone, and was extra-articular in location as opposed to synovial chondromatosis. Magnetic resonance imaging (MRI) showed cortical discontinuity between the condyle and the surrounding lesion, thus confirming complete separation (Figure 3). The disc was not found and severe degenerative bony changes were seen in condyle.

The mass was resected completely, and reconstruction was performed using total alloplastic TMJ replacement utilizing Biomet prosthesis (Jacksonville, FL, USA) (Figure 4A). Intraoperatively, a ringlike grey white osseous mass was identified in the joint capsule and surrounding the condyle with no attachment to the TMJ (Figure 4B). The disc was not explored. The cortical bone of the condyle and fossa contacted together. Severe degenerative changes were seen involving the condylar head.

Macroscopic examination revealed a gray white, ring-like, homogeneous mass with clear margins and firm consistency. Microscopic examination showed dense lamellar bone with well-defined haversian sys-

tems and regular osteocytes (**Figure 5**). Abundant connective tissue was found to in the marrow. An adult benign hyaline cartilage was seen at the periphery.

Follow-up plain radiographic and CT examination 3 years after surgery (Figures 6, 7) demonstrated no evidence of recurrence with stable TMJ prosthesis. The patient had a mouth open-

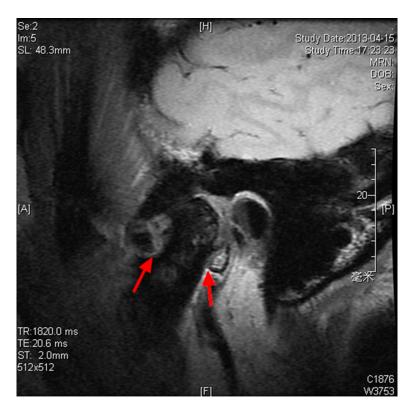


Figure 3. MRI showing cortical discontinuity between the lesion and the condyle.

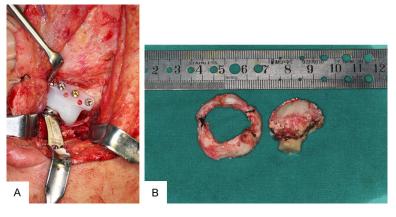


Figure 4. (A) The mass was resected and reconstruction with Biomet prosthesis. (B) A ring-like grey white osseous mass was removed with no attachment to the bony condyle.

ing of 28 mm, with no pain and no malocclusion.

Discussion

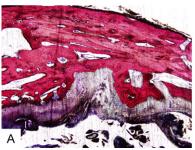
Soft tissue osteoma is a very rare tumor characterized by dense lamellar bone with no direct attachment to adjacent bony structures. Lesions typically occur in the tongue [2, 3] or large joints [1]. Involvement of the smaller

joints is unusual. To the best of our knowledge, there were no previous articles reporting such lesions in TMJ, therefore, the case presented in this article may be the first to report a ring like soft tissue osteoma in the TMJ capsule.

The differential diagnosis included synovial chondromatosis (SC), metabolic calcium disorders such as calcium pyrophosphate dehydrate crystal deposition disease (CP-PD), and tumoral calcinosis (TC). Three benign lesions were identified with aggressive clinical features that rarely occur in the TMJ. SC is characterized by loose bodies lying within the articular compartment [9], whereas the mass described in this article was located extra-articularly in the TMJ capsule. In addition, the histologic findings are distinctly different, with a large amount of hyaline cartilage seen in the SC specimen. CPPD and TC can be diagnosed by characteristic histological findings, which have specific calcified deposits rather than dense lamellar bone with well-defined haversian systems characterizing the discussed lesion [10, 11].

The pathogenesis remains unclear. It is thought that soft tissue osteoma, together with soft-tissue chondromas, soft-tissue osteochondromas, and myositis ossificans, represent a wide spectrum of response to soft tissue trauma with pro-

gressive maturation of histologic elements and endochondral ossification, representing the final most mature stage of a post-traumatic metaplasia [6]. Soft tissue osteoma, chondroma, and osteochondroma share many histologic characteristics that include adult hyaline-type cartilage at the periphery. Differentiation can be made based on the relative proportion and location of the cartilaginous tissue, to-





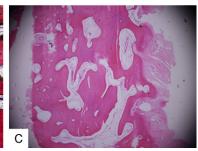


Figure 5. (A, B) Hard tissue biopsy revealed dense lamellar bone with well-defined haversian systems; (C) Hematoxylin-eosin stained photomicrograph of the mass showing mature bone areas.

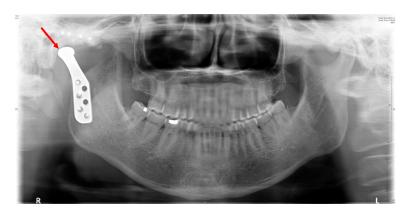


Figure 6. Follow-up panoramic radiograph demonstrated no evidence of recurrence with stable TMJ prosthesis.



Figure 7. Follow-up coronal CT images showed a good position and shape of prosthesis.

gether with the maturity of the osseous tissues. Myositis ossificans has a characteristic zoning pattern of ossification with areas of radiolucency within the central portion and a denser rim at the periphery in plain radiography.

The exact etiology of soft tissue osteoma is still unknown. To be classified as a neoplasm, the

lesion should arise spontaneously and not secondary to trauma or inflammation. Moreover, lesions should not be of developmental origin and should grow unattached to the periosteum or periarticular structures [5]. The case reported here fulfills the above-mentioned criteria.

In this case report, the mass was resected completely with soft tissues completely separating the lesion from the condylar bone. Owing to the old age of the patient with bad

bone quality of the costochondral graft, TMJ Biomet prosthesis was chosen for reconstruction. A ring like soft tissue osteoma in the TMJ capsule presented in this article is unusual, with a confusing term of diagnosis. However, the lesion had characteristic radiographic and histologic features as determined by imaging to show the extraarticular location of the lesion. These include: no direct attachment with adjacent bone confirmed directly intraoperatively and pathologic correlation was required to make an accurate and final diagnosis.

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