Case Report

JOMP
Journal of Oral Medicine and Pain

pISSN 2288-9272 eISSN 2383-8493
J Oral Med Pain 2019;44(1):35-39
https://doi.org/10.14476/jomp.2019.44.1.35

Calcifying Aponeurotic Fibroma of the Temporomandibular Joint in an Adult Patient: A Case Report

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Received February 22, 2019
Revised March 18, 2019
Accepted March 18, 2019

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This study was supported by a Clinical Research Grant from Pusan National University Hospital (2019).

Calcifying aponeurotic fibroma (CAF) is an uncommon benign soft-tissue fibroblastic tumor with characteristic histological features. It mainly occurs in the distal part of the extremities, such as the hands and feet, in children and adolescents. Males are twice as commonly affected as females. CAF exhibits local invasiveness, and hence, its recurrence rate is also high. Therefore, complete surgical excision is both diagnostic and therapeutic. The occurrence of CAF in the maxillofacial region, especially the temporomandibular joint (TMJ), is very rare, and this necessitates its differentiation from other TMJ neoplasms. The differential diagnosis of CAF requires microscopic examination. Herein, we report a rare case of CAF located at the left mandibular condyle, which was confirmed by histopathological analysis.

Key Words: Adult; Calcifying aponeurotic fibroma; Temporomandibular joint

INTRODUCTION

Calcifying aponeurotic fibroma (CAF) is a rare benign soft-tissue tumor that predominantly occurs in children and adolescents, particularly males [1]. It primarily arises in the distal extremities, most commonly occurring in the fingers, palms, and soles. It is more often detected in males than in females, with a male:female ratio of approximately 2:1. Its typical clinical manifestation is a progressive, slow-growing, painless, and firm subcutaneous mass measuring less than 3 cm in diameter. The tumor is histologically characterized by the appearance of calcification within the proliferating fibroblasts. Although CAF is a benign tumor, it has a tendency to infiltrate the surrounding tissue, and recurrence has been well documented. The treatment of choice is surgical excision including a margin of the surrounding normal tissue, and histological evaluation is required for a definitive diagnosis [2,3].

To our knowledge, no cases of CAF arising in the temporomandibular joint (TMJ) have been reported to date. Moreover, the extremely low incidence of CAF in the TMJ, especially in older patients, might lead to its misdiagnosis as other TMJ neoplasms, which are common at the TMJ. In the present article, we describe a rare case of CAF affecting the left TMJ in a 49-year-old man and provide useful information on this rare entity.

CASE REPORT

A 49-year-old man visited the Department of Oral Medicine, Pusan National University Hospital, with the chief complaint of swelling and pain in the left TMJ area.

He had noticed a progressive, painless swelling in the left preauricular region during the past 3 years and experienced a limitation of mouth opening with pain during the past 3 months. He had no history of a systemic disease and previous trauma or surgery on the left TMJ or ear.

A clinical examination revealed a firm, non-fever, palpable mass in the left TMJ. The overlying skin showed no remarkable characteristics (Fig. 1). He had a maximum mouth opening of 38 mm without a TMJ sound and a deflected mouth opening path. The left TMJ area elicited tenderness on opening and closing the mouth. Mandibular asymmetry and occlusion change were absent. Neither sensitivity nor motor function was reduced on either side of the face. Panoramic radiography revealed no pathological findings, such as infections, fractures, or any other abnormalities, in the left mandibular condyle and its adjacent structures. No calcifications were seen peripheral to the left TMJ (Fig. 2). However, computed tomography (CT) clearly demonstrated the presence of the lesion. Although a calcified soft-tissue mass was identified, there was no involvement of the adjacent left mandibular condylar bone (Fig. 3).

We referred the patient to the Department of Oromaxillofacial Surgery, and he underwent complete surgical excision of the lesion after 4 weeks (Fig. 4). On the basis of a histopathological examination, the lesion was finally diagnosed as CAF (Fig. 5). No recurrence and symptoms have been observed during the 8-month follow-up period after the excision.

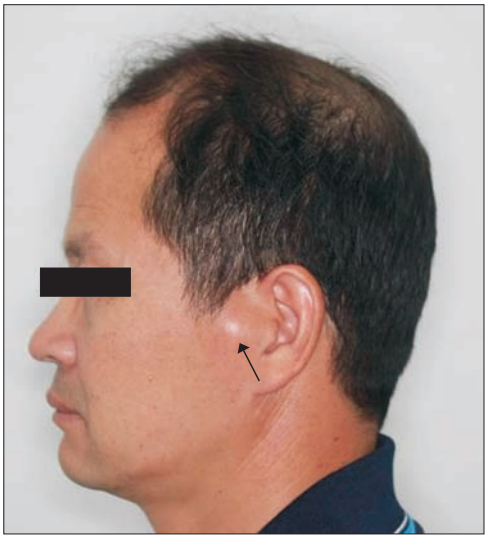

Fig. 1. Preoperative clinical photograph of patient with localized swelling on left temporomandibular joint (arrow). The overlaying skin is normal in color.

DISCUSSION

CAF was first described in 1953 by Keasbey as juvenile aponeurotic fibroma because this tumor usually develops

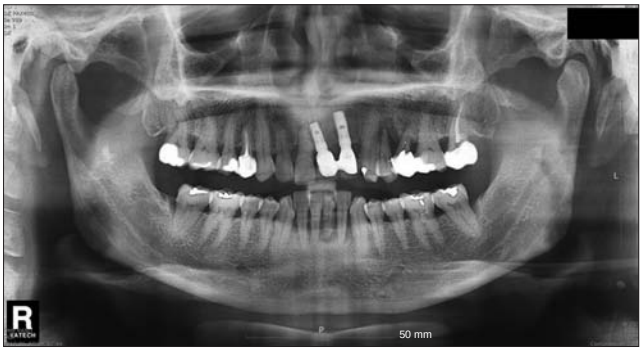

Fig. 2. Panoramic radiograph reveals that there are no pathological findings.

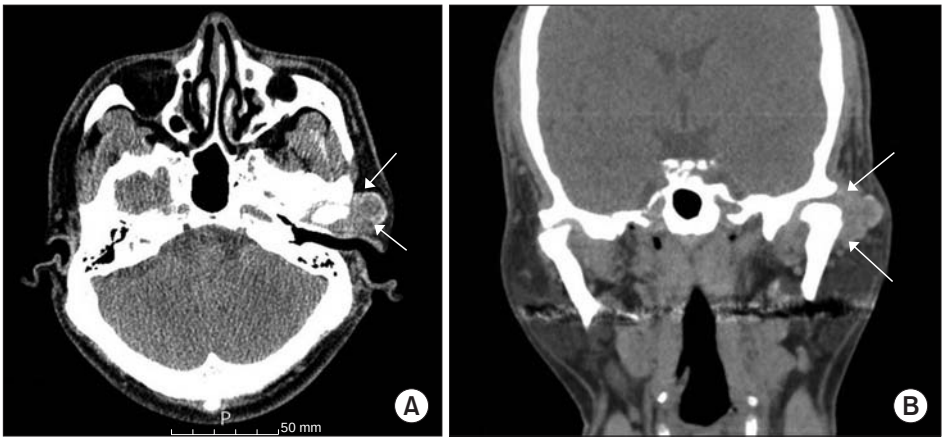

Fig. 3. Computed tomography (CT). (A) Axial CT image, (B) coronal CT image shows a non-specific soft tissue mass (white arrows) on the left temporomandibular joint and high density part. There is no adjacent bone involvement.

in the first to second decade of life. CAF is now known to occur over a wide age range and at various sites. The most common sites are the fingers, palms, and soles; however, case reports have documented CAFs arising rarely on the back, knee, thigh, forearm, and elbow. The maxillofacial region is one of the rarest locations of this pathology, and only few cases of CAF in this region, including the scalp and mandibular ramus, have been reported [4-7].

The clinical presentation of CAF is a firm and slow-growing subcutaneous mass that adheres to underlying soft tissues, such as tendons, fasciae, and aponeuroses [8]. This tumor is an extremely rare pathologic condition in the TMJ, and hence, the presence of a preauricular mass may be misinterpreted as other neoplasms of the TMJ. The TMJ contains bone, synovium, blood vessels, connective tissue, peripheral nerve fibers, and cartilage. Therefore, neoplasms of the TMJ can arise from any one or a combination of these tissues [9]. Because the mandibular condyle is cartilaginous in origin, osteochondromas, osteomas, and chondromas are expected to occur in this region [10]. Furthermore, some tumor-like masses, such as synovial chondromatosis—the most common lesion in the TMJ, should be considered when evaluating masses in the TMJ [11,12]. The signs and symptoms commonly associated with benign neoplasia of the mandibular condyle include a painless swelling in the preauricular region, reduction in maximum interincisal opening, mandibular deviation on mouth opening, and mandibular or facial asymmetry. Moreover, most of the same signs and symptoms are also present in cases of malignant lesions [9]. In the present case, the clinical signs and symptoms were similar to those associated with other TMJ neoplasms. Therefore, radiographic evaluation and histological analysis of the lesion were required for correctly diagnosing CAF.

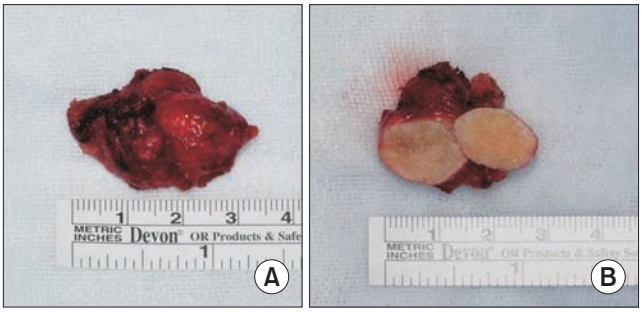

Fig. 4. Gross specimen. (A) Resection gross specimen shows a lobulated and circumscribed mass. (B) The sectioned specimen has a homogeneous yellowish white appearance.

The imaging appearances of CAF can vary depending on the patient's age or presence of calcifications. Some authors

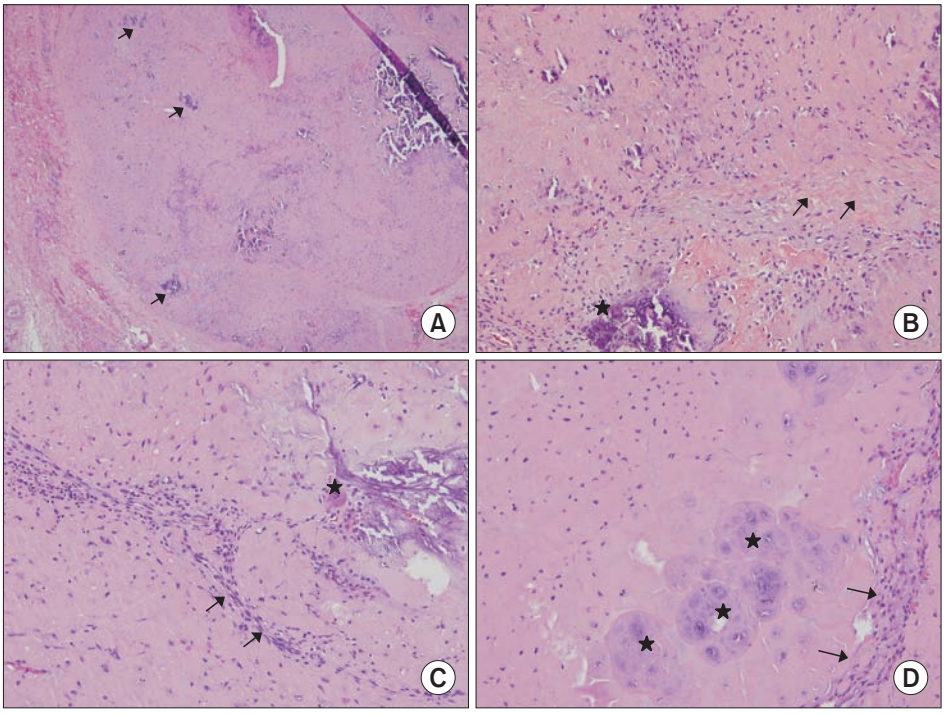

Fig. 5. Histologic examination. (A) The lesion demonstrates characteristic scattered foci of calcification (black arrows) (H&E stain, ×40). (B) Calcification area (black star) and collagenous stroma (black arrows) are observed (H&E stain, ×200). (C) Multinucleated giant cell (black star) is seen around the calcification area. Spindled fibroblasts are aligned linearly (black arrows) (H&E stain, ×200). (D) A local chondroid differentiation (black stars) reveals surrounded by peripheral less cellular and fibroblasic component (black arrow) (H&E stain, ×200).

have suggested the possibility of biphasic development of the tumor, consisting of the initial and late phases. In the initial phase, which is more common in the young, CAF is often not accompanied by calcification. Although calcification is a characteristic imaging finding of CAF, it is hard to detect the lesion in the initial phase. During the late phase, calcification and cartilage formation become clear, and the tumor is more compact and nodular [2]. In most cases, calcification is confirmed using radiography [13]. Panoramic radiographs may show an ill-defined margin of the soft-tissue mass, with a variable extent of finely stippled calcifications within the lesion. CT is useful in depicting the calcified area of the lesion and its association with the adjacent bone; other lesions demonstrate non-specific soft-tissue attenuation on CT. Nevertheless, adjacent bone involvement is very rarely observed [1,3].

Although the observation of calcification on radiography might suggest the presence of CAF, the reliability of imaging in the diagnosis of CAF is limited. Confirming the diagnosis of CAF requires a histological study, which demonstrates the proliferation of fibroblasts, collagenous stroma, small calcification, and areas with cartilage formation. Although the fibroblastic proliferation was impressive, the unique feature was the appearance of a local chondroid differentiation and calcification, as seen in the present case (Fig. 5). Microscopic analysis of the lesion reveals two components: 1) a less cellular, spindle-shaped, fibroblastic component, and 2) nodular depositions of calcification, accompanied by more rounded, chondrocyte-like cells and osteoclast-like giant cells [8]. The tumor cells are enmeshed in the dense collagenous stroma and have a tendency to extend into the surrounding fatty tissue and muscle cells. The tendency for a decrease in cellularity and an increase in collagenous stroma with age may cause tumor maturation. Mature bone formation has also been described in a long-standing lesion [6]. On the basis of the findings reported in the literature, the mass seen in our case was considered to clinically, radiologically, and histologically correspond to the late phase of CAF, because the mass had a nodular appearance with relatively dense calcifications in our 49-year-old patient with a long clinical history.

CAF exhibits growth in a diffuse, poorly circumscribed manner, and has a 50% probability of local recurrence after surgical resection [3]. However, because the aggressive growth potential of the tumor decreases with time and the patient's age, recurrence is higher in the younger age group. Complete local excision is important to prevent tumor recurrence, and regular follow-up is recommended after surgical excision [8].

In summary, we report the radiographic and histological features of the first case of CAF arising in the TMJ of an older patient. Patients with this condition show symptoms clinically similar to those of other TMJ neoplasms. Identifying the characteristic radiographic and histological features of CAF is the key to its correct diagnosis. At that point, the clinician should carefully review the patient's history, physical findings, and imaging findings. Ultimately, surgical excision will be required, both to treat the tumor and to establish the final histological diagnosis. Therefore, despite its rarity, CAF should be considered in the differential diagnosis of older patients with a soft-tissue mass in the TMJ. We believe this case report provides useful information for the diagnosis of CAF in the TMJ.

CONFLICT OF INTEREST

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

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