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CASE REPORT

Temporomandibular Joint Synovial Chondromatosis Posing as Diagnostic Dilemma: A Case Report

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Introduction

Synovial Chondromatosis is a chronic process and a benign tumor-like disorder which forms a loose cartilaginous nodule in the synovial membrane. These nodules can be in different shape and form, pedunculated, single or multiple in number, free or attached to joint space, can be detached from synovial membrane and also process a chance of calcification. It was first described by Auhausen in 1933 in the literature. Patients usually report with a history of difficulty in mouth opening gradually with time and pain with mild swelling on the preauricular area when involving temporomandibular joint. The lesion hinders the normal movements of temporomandibular joint. It is commonly reported in the long bones [1]. In temporomandibular joint, both unilateral and bilateral cases are reported with more gravity to right side in unilateral cases [2]. Male/female ratio is 1:1.5 in occurrence [2, 3]. It mainly occurs at the age of 30–50 [3]. It has an idiopathic etiology, but the literature reports joint disorders or trauma [4]. In cases of severe reduction of joint movement, deviation can also be a clinical feature toward unaffected side. Lesion is not associated with pathology as reported till now. Clinically, the examination has various differential diagnoses, but magnetic resonance imaging or temporomandibular joint tomography is helpful. Histopathology confirms the diagnosis. Removal of lesion is the treatment of choice.

Case Report

We report a case of 36-year-old male who was referred from department of ENT as the patient was having a history of ear pain. Ultrasonography elicited a heterogeneous nodule of 12 mm × 19 mm × 23 mm located posterior to the posterior capsule of parotid gland closely attributing the neck of mandibular condyle without cortical erosion. He also complains about having pain in the temporomandibular joint region of left side for 2 weeks and gives history of difficulty in mouth opening and gradual reduction for 9 years which is idiopathic in origin. On extra oral clinical examination, mild swelling was present on preauricular area on the left temporomandibular joint region. Tenderness was present on forceful wide mouth opening. Jaw deviation was absent. Intraoral dental occlusion was stable. Mouth opening was 33 mm. Clinical diagnosis was temporomandibular joint disorder. Further radiographic evaluation was performed by taking OPG which does not give significant diagnosis. So magnetic resonance imaging was advised which reported a small heterogeneous lesion measuring 21 mm anteroposteriorly, 18 mm craniocaudally and 17 mm transverse along the margins of temporomandibular joint surrounding the condyle and neck of hemimandible involving parotid space (Fig. 1). The lesion is of low-to-intermediate signal on T1W1, hypo intense, somewhat heterogeneous in T2W1 and heterogeneously

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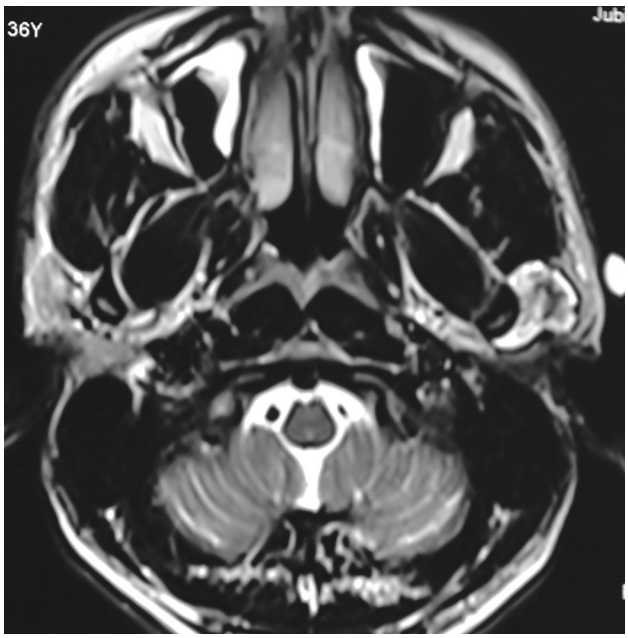

Fig. 1 Magnetic resonance imaging-axial section

enhancing. Mucosal thickening was present in bilateral ethmoid and maxillary sinuses. Possibility of calcium pyrophosphate dihydrate deposition disease and tophaceous/tumoral form was suspected, and differential diagnosis was given as temporomandibular joint synovial chondromatosis as no remodeling or mass effect on condyle with intracranial involvement was noted. Plain helical computed tomography scan of left temporomandibular joint showed a soft tissue density lesion (40–55 Hounsfield Unit) in close relation to condyle of mandible and posterior aspect of ramus and left masseter measuring 20 mm craniocaudally, 14 mm transverse and 20 mm anteroposteriorly (Fig. 2). Minimal scalloping was noted at the lateral aspect of condyle. There was no extension into temporomandibular joint or infratemporal fossa. Lateral and inferior part of the lesion is closely abutting superficial lobe of parotid gland. After a thorough evaluation of clinical and radiographic diagnosis, excision of lesion was planned under general anesthesia. A preauricular approach was taken, and joint space was exposed (Fig. 3). Lesion was separated from the surrounding vital borders and margins, and excised. Excised tissue was sent for histopathologic evaluation, showing macroscopically a gray-brown nodule of irregular surface with cut sections showing small yellowish white bits. Esthetic closure was performed. Histopathology (× 40) reported a small metaplastic cartilaginous nodule of varying size with calcification and also synovial tissue along with nodules of cartilage, normal chondrocytes are noted with occasional foci of calcification which confirmed the clinicoradiographic diagnosis. Postoperatively, patient had an adequate mouth opening with minimal scar on the preauricular incision area and no pain in normal temporomandibular joint movements (Fig. 4).

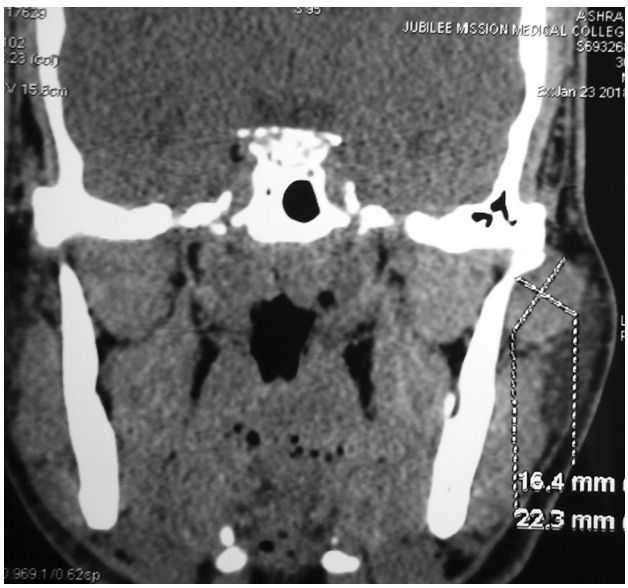

Fig. 2 Temporomandibular joint computed tomography

Discussion

Synovial chondrometaplasia, articular chondromatosis, osteochondromatosis, synovial osteochondromatosis, chondrocalcinosis articularis, tenosynovial chondromatosis, joint chondromatosis) metaplastic change of mesenchymal cells in the joint synovial tissue into cartilage [4]. Tgf-1 involvement in chondrogenesis abnormalities has been reported on chromosome 6 Milgram's phases i) active synovial disease, no loose bodies ii) active synovial proliferation, free loose bodies iii) multiple loose bodies, no synovial disease [5]. Temporomandibular joint synovial chondromatosis is classified histopathologically and radiologically. Histologically, synovial chondromatosis may be divided into three stages of development: (1) metaplasia found in the synovial membrane without the presence of detached particles, (2) metaplasia found in the synovial membrane with the presence of detached particles and (3) only detached particles, which may vary in size from less than 1 mm to greater than 10 mm [6]. Noyek and coworkers (1) pointed out radiologic features of synovial chondromatosis in the TMJ, namely (1) widening of the joint space, (2) limitation of motion, (3) irregularity of the joint surface, (4) presence of calcified loose bodies (cartilage) and (5) sclerosis or hyperostosis (overgrowth) of the glenoid fossa and mandibular condyle [7]. Differential diagnoses of this lesion are sarcomatous degeneration (rare), synovial chondrosarcoma, loose bodies,

Fig. 3 Exposure and excision of lesion

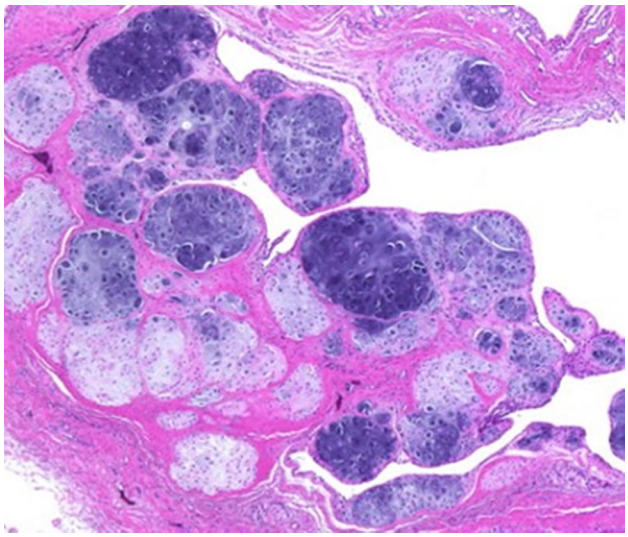

Fig. 4 Histopathologic slide of lesion

osteochondritis dissecans, neurotrophic arthritis, tuberculous arthritis, osteochondral fibrosis, osteonecrosis, periarticular pseudogout, amyloid arthropathy (when disease is active), pigmented villonodular synovitis (when no calcifications present). Familial synovial chondromatosis has been reported in three men of the same family (x-linked) in two generations. Multifocal synovial chondromatosis has been reported with bilateral TMJ occurrence, simultaneous synovial chondromatosis and pigmented villonodular synovitis (PVNS). Simultaneous involvement of both diseases in one joint has been reported TMJ: iatrogenic chondromatosis has been reported 8 months after the autologous chondrocyte implantation, and secondary synovial chondromatosis occurrence within a bursal sac overlying an osteochondroma has been reported which mimics a chondrosarcoma radiologically [8]. In the animal model, it is found in dog which had shoulder involvement has been reported as a cause of lameness [9]. The peculiar gross and histopathologic features are nodules of cartilage adjacent to synovium, often connected only by a stalk. Intra-articular cartilaginous loose bodies "snow storm knee" have been described with hundreds of loose bodies without synovitis. Central area of loose body loses nutrition source, undergoes necrosis and calcifies. Synovial metaplasia to cartilage substantiated by lack of immunohistochemical detection of the Ki-67 protein (proliferation marker) [10].

Conclusion

Temporomandibular joint synovial chondromatosis is rarely reported worldwide. To the best of our knowledge in the published literature, it is a rare case reported in India. As the lesion is benign and slow growing, it also creates functional disturbance and psychological imbalance to patient. Clinical examination alone is not self-satisfactory to come with a diagnosis regarding such cases, but also radiology and histopathology are equally important.

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