CASE REPORT

Synovial Osteochondromatosis of the Temporomandibular Joint Manifesting as a Large Infratemporal Space Occupying Lesion

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Received: 5 July 2016 / Accepted: 27 September 2016 / Published online: 3 October 2016
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Keywords Synovial osteochondromatosis (SC) · Tumoral calcinosis · Infratemporal fossa

Introduction

Synovial chondromatosis (SC) is a rare, benign monoarthritic arthropathy of unknown etiology characterized by chondroid metaplasia of the synovium usually affecting larger diarthroidal joints of the axial skeleton, typically the knee (35 %), elbow (22 %), wrist (11 %) and hip (4 %) [1, 2]. Highly cellular metaplastic, cartilaginous nodules develop in the synovial membrane of the affected joint, which slowly enlarge and then detach to form loose bodies within the joint, the ossification of which lead on to osteochondromatosis. TMJ involvement is rare, with approximately 120 cases having been reported so far [3]. Females are affected more frequently than males [2].

Various synonyms include chondrometaplasia, synovial chondrosis, osteochondromatosis, and periarticular tenosynovial chondrometaplasia [4]. SC may arise either as a primary form or may be secondary to pre-existing joint disease, such as arthritis [5].

Case Report

A 30 year old male patient reported with a longstanding dull pain in front of the left ear, which had persisted for over a year and a half, and had recently, over the past 2 months, increased in intensity and was accompanied by a slowly developing swelling and difficulty in opening and closing of the mouth. On examination, the patient had a localized, oval, firm, smooth, non-tender swelling in the left TMJ region approximately 1 cm in diameter. Condylar movements were bilaterally palpable, smooth and synchronous. There was no evidence of mandibular midline shift or of any occlusal deviation of the mandible upon mouth opening and closing.

Radiographs revealed an increase in the left TMJ space and a radio-opacity medial to the left condyle and sigmoid notch. CT scans (Fig. 1) revealed a well-defined calcific density lesion, measuring 32 × 40 × 15 mm (AP × Tr × CC) near the left TMJ extending into the Infratemporal fossa. Multifocal calcific loose bodies were found scattered within a well-defined low-attenuated mildly enhancing soft tissue mass seen around the condyle (Fig. 1). The cortices of the mandibular condyle and glenoid fossa showed minimal erosions. There was sclerosis seen in the petrous part of the left temporal bone forming the roof of the TMJ and of the proximal zygomatic arch (the zygomatic process of the temporal bone).

Contrast enhanced MRI with T1 and T2 weighted images (Fig. 1j) showed a large non-enhancing low signal intensity heterogeneous left TMJ lesion, 30 × 40 mm in size, consisting of a circumferential cluster of partially calcified aggregates surrounding the mandibular condyle, expanding the joint space and invading the infratemporal fossa. The adjacent muscles appeared displaced without any infiltration. There was remodeling of bone around the

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Fig. 1 a–c A well defined calcific density lesion, in the *left* TMJ region, extending medially into the Infratemporal fossa, anteriorly into the sigmoid notch and laterally, bulging into the preauricular region. **d–i** CT scans revealing multifocal loose calcific bodies scattered around the *left* condyle (**j**) Contrast enhanced MRI revealing a large heterogeneous *left* TMJ lesion, consisting of a cluster of partially calcified aggregates surrounding the joint space and invading the infratemporal fossa

lesion, although no bony destruction or intracranial extension was evident.

CT scan and MRI findings favored a diagnosis of Synovial Chondromatosis in the Left TMJ. Differential diagnosis included Tumoral calcinosis, Pigmented Villonodular Synovitis, Osteochondritis Dissecans, or a primary bone neoplasm such as Chondroma or Chondrosarcoma.

A Fine Needle Aspiration Biopsy was attempted under Ultrasonographic guidance. However, it yielded only blood as the lesion could not be accessed being located too far medially within the infratemporal fossa due to which, a representative tissue sample was impossible to achieve.

The patient was planned for a surgical exploration and excisional biopsy of the mass using a lateral temporal and preauricular transzygomatic approach, with condylectomy kept as an option in case of inadequate access.

The Popowich and Krane's modification of the Al Quayat and Bramley's modified temporal incision was employed (Fig. 2a–c), extending it well down almost to the level of the lobule of the ear. The arch was exposed up to the body of the zygoma. The TMJ capsule, which appeared to bulge in the region of the condylar head, (Fig. 2d) was carefully slit open and the tumor mass popped out (Fig. 2e, f). It appeared white in colour and cartilaginous in consistency. A braided wire was threaded through a hole drilled through the left angle of the mandible, to exert a downward

traction on the mandible, enabling access into the infratemporal space. The superficial portion of the tumor mass was carefully removed and immediately sent for a frozen section biopsy to rule out malignancy. A section of the zygomatic arch was resected after stripping the masseter insertion, to gain better access into the infratemporal extension of the mass (Fig. 2g, h). The remainder of the tumor was then carefully dissected off the adjacent tissues such as the lateral pterygoid muscle and the deep aspect of the temporalis muscle (Fig. 2i, j). The osteotomised segment of the zygomatic arch was fixed back into place using a low profile titanium minibone plate (Fig. 2k), followed by layer wise closure of the site (Fig. 2l).

Histopathological examination (Fig. 3) revealed ossified hyaline cartilage and hypertrophied synovium, confirming the provisional diagnosis of Synovial chondromatosis of the TMJ (Fig. 3). Post operative CT scans confirmed complete elimination of the tumoral mass and there followed excellent healing of the operated site (Fig. 4).

Discussion

SC of the TMJ often presents a diagnostic dilemma [6], as the main clinical findings are pain, clicking, occlusal changes, volume augmentation of joint space and a limited

Fig. 2 a–d Hemicoronal incision to expose zygomatic arch and TMJ region, revealing a bulging fibrous capsule with the tumor mass protruding from it. e, f Fibrous capsule incised revealing the tumorous mass. g Segment of the zygomatic arch transected to gain access to

the medial aspect of the TMJ and the infratemporal fossa. h, i Friable cartilaginous mass excised. j–l Zygomatic segment replaced and fixed and the hemicoronal flap closed in layers

Fig. 3 **a** H&E (X200) Synovial lining with ossifying cartilage below, confirmatory of Synovial chondromatosis. **b** The synovium (indicated by the white arrows) seen in the excised bits appeared thickened and

hypertrophied and showed a mild mononuclear inflammatory infiltrate. **c** (X300) Mature hyaline cartilage with evidence of calcification (indicated by the black arrows)

Fig. 4 **a–d** Postoperative CT scans confirming complete elimination of the tumoral mass. **e–h** Excellent healing of the operated site with a good camouflage of the hemicoronal incision line by the hair

mouth opening [7], which are general findings that accompany most of TMJ pathologies including degenerative joint disorders. CT and MRI together with Histological examination play a vital role in establishing the correct diagnosis.

The differential diagnosis of Synovial chondromatosis are Osteoarthritis (joint mice), which is characterized by small calcifications [8]; Chondrocalcinosis (pseudogout), characterized by fine calcifications with an even distribution [9, 10]; Osteochondroma, which arises from the condylar head and has a cartilage cap [11]; Pigmented villonodular synovitis, which is not localized to a portion of the joint [12]; and Osteochondritis dissecans, which is seen in younger patients and is associated with osteochondral fracture.

Conclusion

Although Synovial chondromatosis of the TMJ, is a benign metaplastic process rather than a true neoplastic one, it is locally aggressive and progressively invasive, with the potential for displacing and compressing anatomically important structures in the region, such as cranial nerves exiting the skull base, leading to neurological deficits; and is also capable of eroding the adjacent bony surfaces, including the cranial base with a possibility of intracranial extension. Due to the nature of progress of this lesion, it is important to have a good understanding of the spectrum of clinical, radiographic as well as histopathological features presented by this rare pathology, so as to proceed with a prompt and efficient management, which entails its

complete surgical removal followed by total or subtotal synovectomy, which was diligently carried out in the patient.

Compliance with Ethical Standards

Conflict of interest None of the authors of this article have received any research grant, remuneration, or speaker honorarium from any company or committee whatsoever, and neither owns any stock in any company. Both the authors declare that neither of them have any conflict of interest.

Ethical Approval This article does not contain any studies with human participants or animals performed by any of the authors.

Informed Consent Informed consent was obtained from the individual participant in this study.

Research Involving Human Participants and/or Animals All procedures performed on the patient (human participant) involved were in accordance with the ethical standards of the institution and/or national research committee, as well as with the 1964 Helsinki declaration and its later amendments and comparable ethical standards.

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