Intra-articular loose osteochondroma of the temporomandibular joint

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Abstract. Osteochondroma or osteocartilaginous exostosis is one of the most common benign bone tumours. It usually develops in the metaphyses of long flat bones, but rarely occurs in the oral and maxillofacial area, where it is generally associated with the coronoid process or the condyle. Loose osteochondromas inside the joint are generally a rare pathology usually described in large joints. The authors present a case of an intra-articular loose osteochondroma of the temporomandibular joint (TMJ), which represents the first case of such a pathological entity in the literature. The authors suggest that it should be considered in any differential diagnosis of loose bodies described in the TMJ. Case Report TMJ Disorders

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Osteochondroma or osteocartilaginous exostosis, one of the most common benign bone tumours, consists of an exophytic bone proliferation with a hyaline cartilage cap protruding from the surface of the affected bone. It usually develops in the metaphyses of long flat bones, but rarely occurs in the oral and maxillofacial area, where it is generally associated with the coronoid process or the condyle.³ This tumour can occur singly or as part of an autosomal dominant syndrome known as osteochondromatosis.⁶ Different aetiologies have been proposed. The most recent theory is based on the presence of nests of chondrocytes in the periosteum.⁸ Mechanical stress may lead to hyperplasia of these cells, because the lesion is usually located in areas such as the tendon insertions. Malignant change is rare in solitary osteochondromas, approximately 2% of cases, but does occur in approximately 5% of cases of multiple hereditary osteochondromatosis. No case of malignant transformation has been reported in the skull.¹ Most cases of condylar osteochondroma manifest with facial asymmetry or malocclusion (ipsilateral posterior open bite, contralateral cross bite) with limited temporomandibular joint (TMJ) movements. Loose osteochondromas inside the joint are a very rare pathology. Sarmiento and Elkins9 have reported a case that showed a large intra-articular osteocartilaginous loose body in the knee joint and termed it a giant intra-articular osteochondroma. Milgram and Dunn⁵ described a case that presented with an osteochondral tumour and two cases that presented with chondral tumours in the knee joint defined as an intra-articular osteochondroma and an intra-articular

chondroma, respectively. Lim reported a case of extraskeletal osteochondroma of the buttock.⁴ There are no cases in the literature of TMJ involvement. The authors present a case of intra-articular loose osteochondroma of the TMJ, which represents the first case of such a pathological entity in the literature.

Case report

A 22-year-old woman presented at the authors' department with facial asymmetry. She reported that it had appeared at the age of 15 years and had been gradually increasing. Her past medical history was unremarkable. She did not have any history of TMJ dysfunction and denied any recent trauma to the ear or mandible. Physical examination showed facial asymmetry with right lateral deviation





Fig. 1. Patient showing (a) facial asymmetry; (b, c) dental III class malocclusion with (d) right lateral mandibular deviation.

of the chin (Fig. 1a). The dental occlusion was characterized by class III malocclusion with a right lateral mandibular deviation (Fig. 1b–d). The patient had normal mouth opening, without deviation, and the mandibular range of motion was normal in all directions. There was no sound in the TMJ and no pain during the mandibular functionality examination. There was no facial nerve paralysis or paresis. The rest of the head and neck examination was unremarkable. A



Fig. 2. (a) Orthopantomogram showing an oval hyperdense mass adjacent to the left condyle of the TMJ. (b) Sagittal CT scan (bone window) showing a bony dense formation of about $2 \text{ cm} \times 1.5 \text{ cm}$ in the left glenoid fossa (coronal CT).

clinical differential diagnosis should include all the pathological conditions that can occur with mandibular deviation and malocclusion. The most common should include giant cell tumour, condylar hyperplasia, fibro-osseous lesion, vascular malformation, osteoma, chondroma, and osteochondroma. More rarely reported condylar tumours have included chondroblastoma, chondrosarcoma, osteoblastoma osteoid osteoma, enchondroma, osteosarcoma, and metastatic tumours.

Orthopantomography revealed an oval hyperdense mass adjacent to the left condyle of the TMJ (Fig. 2a). A computed tomography (CT) scan was obtained, which showed a bony dense formation of about 2 cm \times 1.5 cm in the left glenoid fossa. The formation had an oval appearance with a well-defined peripheral sclerosis rim with some patchy areas of low density. The mandibular condyle was dislocated anteriorly with a new joint reported with the tubercle of the temporal bone (Fig. 2b). The radiographic features were compatible with an osteochondroma and it appeared completely detached from the articular bone surfaces. The patient was taken to the operating room and access was gained to the left TMJ with a preauricular approach extending to the temporal region. The incision was made through the skin and subcutaneous tissues (including the temporoparietal fascia) as far as the temporal fascia (superficial layer). An incision was made in the superficial layer of the temporal fascia and dissections were taken at the level of the zygomatic arch as far as the lateral surface of the capsule of the TMJ. Afterwards, an incision of the joint capsule was performed to gain access to the lower compartment of the TMJ cavity (Fig. 3a). A 2 cm \times 1.5 cm mass, adjacent to and behind, the left condule of the TMJ was found. At surgical exploration the mass had no connection with the adjacent bony structures; the condyle appeared completely intact. The mass was carefully isolated and removed from the TMJ cavity (Fig. 3b). The wound was sutured in layers and a pressure bandage applied, which remained in place for 3 days. Histological examination showed a nodular mass consisting of lamellar bone, partially bordered by a thin rim of cartilage tissue (Fig. 3c). These aspects were consistent with the results of a complete detachment of osteochondroma. A resolution of the facial asymmetry and dental malocclusion was obtained (Fig. 4a). The follow-up examination at 18 months revealed no clinical or radiological sign of recurrence (Fig. 4b).



Fig. 3. (a) Joint capsule incision to gain access to the lower compartment of the TMJ. (b) A 2 cm \times 1.5 cm tumour mass removed from the TMJ. (c) Histological section (haematoxylin–eosin 10×) showing a nodular mass consisting of lamellar bone (*) partially bordered by a thin rim of cartilage tissue (arrows).

Discussion

Osteochondroma or osteocartilaginous exostosis, a cartilage-capped exophytic lesion that arises from the bone cortex, is one of the most common benign bone tumours. It usually occurs in the axial skeleton, especially in long bones, such as the distal metaphysis of the femur or the proximal metaphysis of the tibia.³ The oral and

maxillofacial region are not commonly involved. The coronoid is the most affected site in the skull followed by the condyle.³ The aetiology and pathogenesis of this lesion are not fully understood. The most recent theory is based on the presence of nests of chondrocytes in the periosteum, mechanical stress possibly leading to hyperplasia of these cells. The lesion in the TMJ is usually discovered incidentally



Fig. 4. Resolution of (a) facial asymmetry and (b) dental malocclusion. (c) Sagittal CT (bone window) showing no radiological sign of recurrence at 18 month follow up examination.

on radiographic examination or on palpation of a protruding mass in the affected area.

Malocclusion in the form of a lateral open bite on the contralateral side and progressive facial asymmetry are common findings in most cases of condylar osteochondroma. Pain may precede or accompany facial asymmetry in some cases. The lesion grows slowly and is almost painless. In the mandibular condyle it causes limited movement of the jaw. These conditions may also be found in unilateral condylar hyperplasia and other differential diagnoses including osteoma, chondroma, giant cell tumour, myxoma, fibroosteoma, fibrous dysplasia, fibrosarcoma and chondrosarcoma.¹⁰

In spite of the common clinical features, a definitive diagnosis should always be based on clinical, radiological and histological criteria. Conventional panoramic radiography and CT may help to delineate the anatomy of the lesion and surrounding structures. The lesions usually point away from the joint space and can have a pedunculated stalk or sessile base. Osteochondromas should be removed because of this dysfunction and the possibility of malignant transformation. Several surgical approaches have been suggested for the treatment of condylar osteochondroma, including complete resection of the tumour using condylectomy with or without reconstruction or selected tumour removal without condylectomy.² If the tumour involves only a limited area of the condylar surface, preservation of the remaining part of the condyle and reshaping should be undertaken.

In the present case, the authors removed only the intra-articular loose osteochondroma, which appeared completely detached from the articular bone surfaces. This osteochondroma probably originated from a hyperplasia of the chondrocytes in the periosteum⁷ due to persistent mechanical stress. The authors think that the same stress may have led to the detachment of the osteochondral fragment, although the possibility of extraskeletal osteochondroma has been described,⁴ the pathogenesis of which is unknown.

This is the first case of intra-articular loose osteochondroma of the TMJ reported in the literature. The authors suggest that it should be considered in any differential diagnosis of loose bodies described in the TMJ.⁷

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Competing interests

None declared.

Ethical approval

Not required.

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