CASE REPORT

Orthokeratinized Odontogenic Cyst (OOC) of Condylar Head: A Rare Entity

Anil Managutti¹ · Sunita Managutti¹ · Harsh Patel¹ · Shailesh Menat¹

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Abstract Orthokeratinized odontogenic cyst (OOC) is a rare developmental jaw cyst, considered as distinct entity from odontogenic keratocysts as it exhibits a less aggressive behaviour and a very low rate of recurrence. Most commonly occurs in mandibular molar and ramus region, rarely seen in maxilla and mandibular premolar area. But till now never reported in condyle, this article describes a case of OOC involving the mandibular condylar head in a 41-year-old male. The initial clinical diagnosis was given as TMD based on the clinical features, but radiographic evaluation revealed osteolytic lesion in right side condylar head. Condylotomy was performed and the specimen was sent for histopathological examination. A definite diagnosis of OOC was made by histopathological examination of the biopsy specimen. We report a rare entity in this article with its management.

Keywords Orthokeratinized odontogenic cyst · Condylar head · Condylotomy

Introduction

TMJ is a complex structure with many muscles and ligaments regulating its function by working in harmonious pattern. Pain in relation to TMJ region can be difficult to diagnose due to its complex structure. Possible causes for pain in this region can be due to infection or inflammation i.e. arthritis, myalgia, otitis, parotitis, temporal arteritis;

neuralgic pain i.e. trigeminal neuralgia; referred pain i.e. from 3rd molar; or pain due to pathology within the joint or associated structures. The common pathologies of the condyle are tumour/tumour-like lesions like osteoma, osteochondroma, chondroma, and synovial chondromatosis [1]. Osteolytic lesions of TMJ are rare which are difficult to diagnose without radiographic interventions. They may be caused by primary bone cysts and tumours, metastatic lesions of bone and degenerative changes of joint [2]. Of all radiolucent lesions, traumatic bone cysts, aneurysmal bone cysts (ABC), giant cell lesions of the bone, are common.

These lesions are usually asymptomatic until secondarily infected or alter the morphology of TMJ to cause pain. Diagnosis of these condylar lesions is difficult owing to rare occurrence of osteolytic lesions within the condyle and thus, histopathological analysis can only provide final diagnosis. Surgical enucleation or surgical excision of the lesion is the treatment of choice for these lesions; this article describes the diagnosis of the condylar radiolucent lesion as infected OOC and treatment modality of such lesion.

Case Report

A 41 year old male patient reported to the Department of Oral and Maxillofacial Surgery, Narsinhbhai Patel Dental College & Hospital, Visnagar, Gujarat, with a chief complaint of pain on right side of his face since 15 days. Pain was of sudden onset, severe in intensity, sharp and continuous in nature, aggravated on jaw movements along with episodes of electric shock type pain which remained for about 5 s every 30 min. Patient gave history of trauma and fracture of upper and lower jaw due to road traffic accident which was treated by inter maxillary fixation 20 years back. Clinical examination revealed tenderness present

✉ Anil Managutti
dranilman12@gmail.com

¹ Department of Oral and Maxillofacial Surgery, Narsinhbhai Patel Dental College and Hospital, Visnagar, Gujarat, India

over right TMJ region and pus discharge from right ear. His mouth opening was reduced to 3.2 cm and submandibular lymph nodes were tender on palpation. On radiological examination orthopantomogram (Fig. 1) showed a well defined oval radiolucency with corticated borders confined within the condylar head. CT scan (Fig. 1) revealed radiolucent area of 1.2 cm diameter in the medio-posterior region of right condylar head and perforating the condyle medially. Pus from right ear was sent for culture sensitivity test which revealed absence of any organism. The clinical and radiographic features were suggestive of non specific jaw cyst.

It was decided to operate the patient under general anaesthesia for condylotomy and excision of the lesion as

we could not rule out any specific diagnosis. Pre auricular approach was used followed by layer wise dissection. On dissecting the TMJ capsule there was discharge of pus (Fig. 2), which was causing intracapsular pressure and resulting in severe pain. Muscle attachments to the condyle were detached and condylar head was then resected taking appropriate care of medial vital structures. Resected specimen contained cheesy white material (Fig. 2). The wound was thoroughly irrigated with antiseptic solution and closed in layers with an active drain in place. Patient was put on a course of I.V antibiotics i.e. Amoxicillin sodium clavulanate 1.2 gm twice for 5 days. Mild weakness of temporal branch of facial nerve of transient nature was observed in immediate post-operative period. Drain was

Fig. 1 Pre operative OPG and CT scan

Fig. 2 Pus discharge and excised specimen

Fig. 3 H&E stain and immunohistochemistry

removed on 2nd post-operative day. Pus discharge from right ear was not seen post operatively and healing was uneventful. There was increased deviation of mandible towards right side on opening. Physiotherapy was advised for jaw opening and guiding into occlusion.

On histopathological examination, H&E stain (Fig. 3) revealed cystic epithelium of variable thickness and inflamed connective tissue capsule. Epithelium was stratified, squamous orthokeratinized, flat to cuboidal basal cells and prominent granular cells were seen in most of the areas. Few areas showed only keratin flakes. Stroma showed thick collagen bundles with spindle fibroblasts and few areas showed chronic inflammatory cell infiltrate comprising of lymphocytes, plasma cells, macrophages and few multinucleated giant cells. Immunohistochemistry showed presence of Ki67 and P53 (Fig. 3) markers which were confirmatory to Orthokeratinized Odontogenic Cyst.

Discussion

Orthokeratinized odontogenic cyst is a developmental odontogenic cyst described by Schultz in 1927 [3] as an orthokeratinized variant of the formerly called Odontogenic Keratocyst, which is today known as the Keratocystic

Odontogenic Tumour (KCOT). OOC was described as distinct individual entity from other odontogenic cysts by Wright in 1981 and the term Orthokeratinized Odontogenic Cyst was given by Li et al. in 1998 which is the most widely accepted term at present [4, 5]. This lesion is located mainly in the molar and ramus region but its occurrence in the condyle is rare.

According to Filip Brzozowski et al. [6] OKC is more commonly found in mandible body and ramus region and it is frequently associated with an impacted tooth, while in one case they reported OKC in condyle.

According to Stoelinga, KCOTs can present as relatively small unilocular cyst in tooth region or frequently large multilocular cyst in posterior maxilla or angle-ramus region [7]. OKCs tend to grow in an anterior-posterior direction within the medullary cavity of the bone without causing obvious bone expansion [8].

However, OOC is most common in mandibular posterior region and its extension or origin in the condyle has not been reported in literature. World Health Organization (WHO) redefined OKC as a neoplasm and redesignated it as keratocystic odontogenic tumour (KCOT) in 2005 so it became imperative that OOC had to be separated out from KCOT as a distinct entity. OOC's incidence varies among different series, ranging from 16.8 % of cases previously

Fig. 4 18 months post operative OPG

classified as odontogenic keratocyst [9] to only 0.4 % of all odontogenic cysts [10]. OOC has been reported to occur among young adults, with a male predominance and about 75 % of OOCs are associated with impacted teeth [11]. Clinically and radiographically OOC present as hard, progressively growing non-tender enlargement, with a well-defined radiolucency. It is a developmental odontogenic cyst arising from the cell rests of the dental lamina which is absent in condyle; however extraneous variant of cysts occur in the ascending ramus of mandible and away from tooth. The occurrence of cyst in the condyle could be due to epithelial off-shoots (hermarias) of the oral epithelium from basal layer [12]. Histologically, OOC is characterized by a 4–8 cell-layer thick, orthokeratinized epithelial lining, with prominent granulosum and low cuboidal basal cells; while KCOT epithelial lining is thick, parakeratinized with the basal cells exhibiting typical palisading of the nuclei. All these microscopic findings were present in our case, also level of expression of P53 and Ki67 is comparatively lower than that found in OKCs which confirms the OOC. In our case the cyst was infected secondarily, which is an uncommon event in developmental odontogenic cysts.

Treatment recommended for OOC usually involves conservative measures owing to its less reoccurrence rate as compared to OKC which sometimes require extensive procedures. Dong et al. [4] in their study found that enucleation with or without curettage, combination of enucleation followed by marsupialisation and peripheral osteotomy for large multilocular lesion of OOC showed no reoccurrence. In a study done by Crowley, Kaugars and Gunsolle only 4 % of OOCs showed recurrence rates [13, 14]. However for our case posteromedial position of the cyst within the condylar head did not allow direct access, so condylotomy was planned and performed. Post operative complications associated with condylotomy are infection, haemorrhage commonly from messeteric artery and maxillary artery, facial nerve paralysis and disocclusion of teeth. In our case except for transient paralysis of

temporal branch and deviation of the jaw while closing no other complications were encountered. We followed up this case for 18 months; there were no postoperative complaints and radiograph showed no signs of reoccurrence (Fig. 4).

Conclusion

To conclude, Orthokeratinized Odontogenic Cyst is relatively common in mandibular body and ramus, but its occurrence in condyle has not been reported so far. This is the first reported case of OOC originating in mandibular condyle. So OOC should be considered in the differential diagnosis of benign tumours of mandibular condyle and treated accordingly.

Compliance with Ethical Standards

Conflict of interest None declared.

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