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Odontogenic keratocyst of mandibular condylar region: A case report

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ABSTRACT

The Odontogenic Keratocyst (OKC) today is called Keratocystic Odontogenic Tumor (KCOT) by the WHO, consider as a benign intraosseous developmental odontogenic tumor and may be presented in unicystic or multicystic variants. The tumor is surrounded by stratified squamous parakeratinized or orthokeratinized epithelium and has invasive or infiltrative behavior. It happens in the second and third decades of life, and often is more prevalent among males. In this case report we describes an unusual case of OKC in the condylar region and its infiltration into parotid gland which is very rare and minimal clinical presentation indicative of a tumor.

Keywords: Odontogenic keratocyst, Keratocystic odontogenic tumor, Infiltration, Parotid, Mandibular condyle.

Introduction

Odontogenic Keratocyst (OKC) was first discovered by Philipsen in 1956 [1]. The OKC today is called Keratocystic Odontogenic Tumor (KCOT) by the WHO, which as a benign intraosseous developmental odontogenic tumor can have unicystic or multicystic-variants. The tumor is surrounded by stratified squamous parakeratinized or orthokeratinized epithelium and has invasive or infiltrative behavior [2]. It happens in the second and third decades of life, and often is more prevalent among males. The lesion occurs in the mandibular molars and ramus, and its prevalence in the mandible is twice more than in the maxilla [3-6]. The OKC is able to grow in anteroposterior direction in medullary cavities and unable to produce an obvious expansion in the bone, and can also be enlarged asymptomatically and detected by routine radiographs [3,7]. The OKC can be sporadic or associated with nevoid basal cell carcinoma syndrome (NBCCS) [8]. Radiographically, there are four OKC groups, including

1. Envelopmental.

2. Replacement.

3. Extraneous.

4. Collateral [1].

The presence of KCOT in the condylar region and its infiltration into parotid is very rare, which has not been reported in the literature.

Case Report

The patient was a 49-year-old man who referred to the Maxillofacial Surgery Clinics of Shariati Hospital complaining of left parotid swelling and pain. The patient reported a history of pain and swelling in the temporomandibular joint about 15 years ago, who had subsequently experienced three surgical procedures in the left mandibular condyle. The patient did not mention a history of condylar trauma or problems. He had been referred to a physician two years ago due to left parotid and anterior tragus swelling, who had undergone biopsy under general anesthesia the histopathology report findout the biopsy salivary duct cyst. However, the patient did not refer to continuing treatment due to problems and negligence, until he referred to the Maxillofacial Surgery Clinics of Shariati Hospital in Tehran since three months ago following the parotid severe pain and swelling, perforation of the area and the pus drainage. Initial examinations showed rigid parotid swelling whose skin was slough and erythematous, and lesion aspiration was also negative (Figure 1). A panoramic radiograph was requested for the patients to ensure the presence of intraosseous odontogenic and tumoral lesions. Attained findings revealed radiolucency in the left condylar region and also a blurred view in the left condylar neck (Figure 2). Therefore, a facial CT scan was requested for the patient. The findings showed a multilocular lesion with a well defineborders, which had spread from the condylar neck to the left parotid and its contents were homogeneous (Figure 3). After the routine work ups for surgery, the patient was admitted to remove the lesion under general anesthesia. Because the lesion was surrounded the condylar head and for better access for removal of the tumor we decided to remove condyle and because the lesion was recurrent the tumor removed more aggressively. Access was achieved by rhytidectomy incision and subcutaneous dissection to lateral extension of the lesion, and then rhytidectomy incision was extended to submmandibular incision. The lesion was raised from the parotid capsule. The lesion contained a thick capsule containing very rigid keratinous materials. Finally, the capsular lesion was removed from the parotid; the facial nerve was preserved and protectedthrough the lesion (Figure 4 a, b and c). Subsequently, mandibular ramus osteotomized and the lesion was removed along with condyle and ramus as a single piece (Figure 5). After receiving homeostasis in the area, a reconstruction plate with condyle was formed in the area and fixed to the mandible (Figure 6). The hemovac drain was embedded for the patient, which was removed two days after the surgery. Cefazolin antibiotic was given intravenously for five days for the patient, and there was no jaw deflection during opening and closing due to the presence of recon plate. Immediately after surgery, there was a degree of weakness in facial skeletal muscles, mostly due to stretching in the nervous system. Histopathological examination following H & E staining indicated that outer surface is rough and brown. On cutting the cyst is unilocular and filled with keratinous material. Maximal cyst wall diameter was 0.4 cm. Inner surface was smooth and grayish-brown. Epithelium was as stratified, squamous orthokeratinized, flat to cuboidal basal cells, and prominent granular cells were seen in most of the areas (Figure 7).

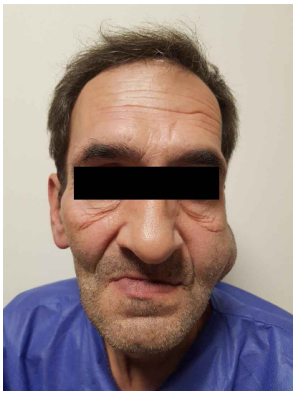

Fig 1. Photograph of the patient.

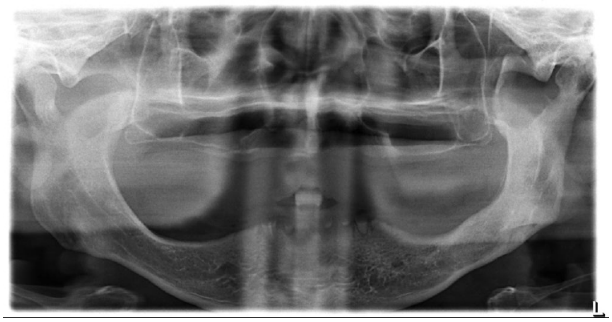

Fig 2. Panoramic of the patient, shows left side condylar head pathology.

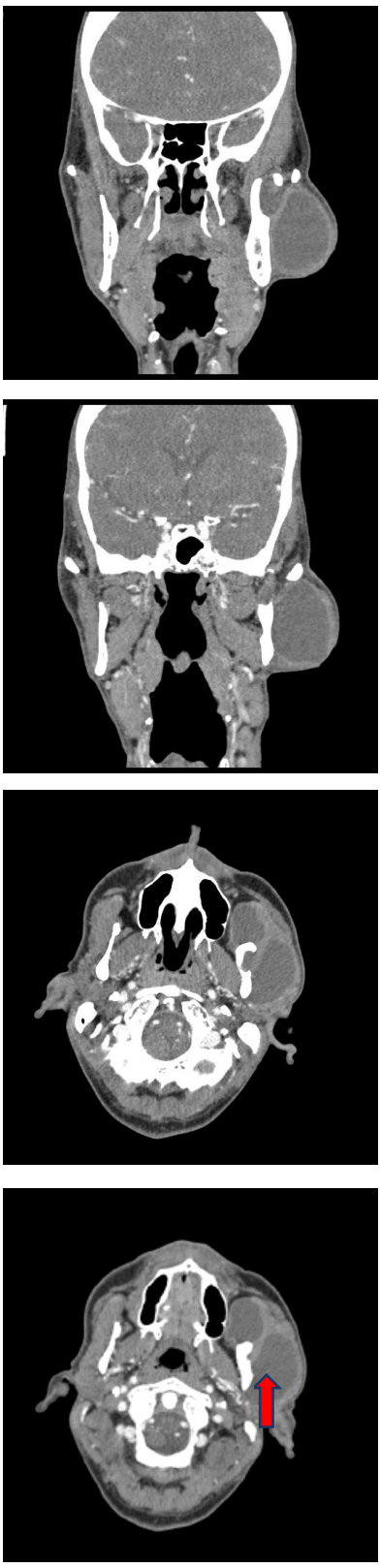

Fig 3. CT scan in axial and coronal cuts, tumor wrap around the mandibular condyle (red arrow).

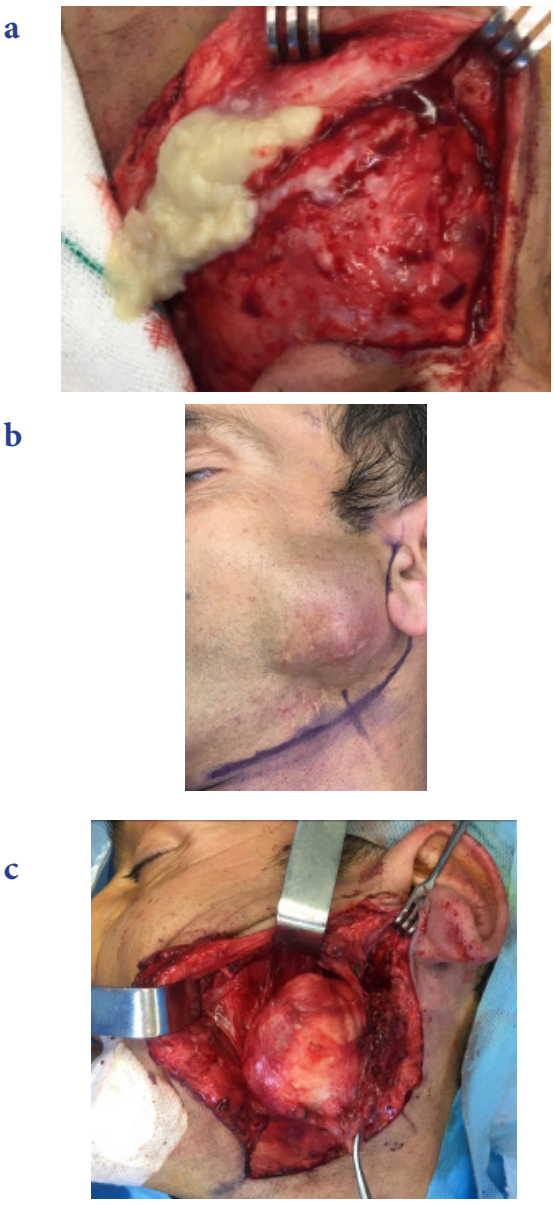

Fig 4. Intraoperative pictures of the patient.

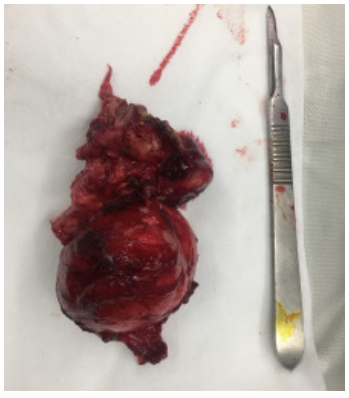

Figure 5. Excised specimen.

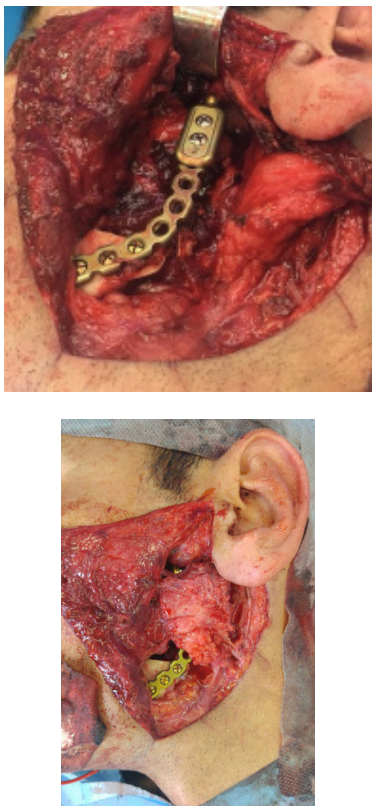

Fig 6. Mandible reconstructed with reconstruction plate and covered the plate with parotid gland.

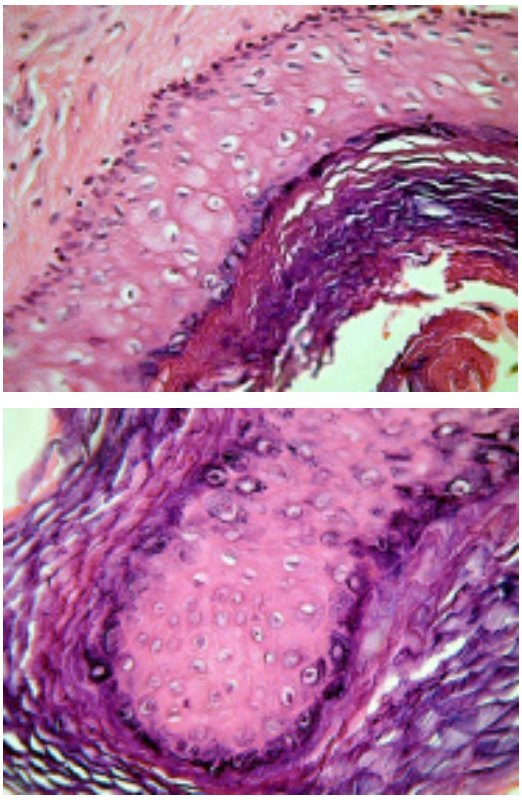

Fig 7. Histologic appearance.

Discussion

The keratocystic odontogenic tumor is a developmental cyst as described by Philipsen in 1956 [1], whose orthokeratinized variant is now called KCOT [2]. This lesion usually occurs in the molar and ramus region and its prevalence in the mandible is twice more than in the maxilla [3-5]. The occurrence of this lesion in the condylar region and its infiltration into parotid is rare. Filip Brzozowski et al. [6] reported that the OKC is generally present in mandibular ramus and body region and mostly associated with an impacted tooth, while a case showed the OKC in condyle. In a study by Stoelinga, the KCOT consider as a small unilocular cyst in tooth region or large multilocular cyst in posterior maxilla or angle- mandibular region [9].

The OKC is able to grow in anteroposterior direction in medullary cavities and unable to produce an obvious expansion in the bone, and can also be enlarged suddenly and detected by routine radiographs [3,7]. The OKC mostly occur among young adults, predominantly in the males and approximately 75 % of cases are associated with impacted teeth [6]. Managutti introduced a case report on the occurrence of OKC in the condyle [10]. The OKC clinically and radiographically is a painless enlarging massand has a radiolucencywith a clear border. It is an odontogenic cyst arising from remnants of dental lamina which is absent in condyle. However, extraneous type of cysts may even extend to theascending mandibular ramus.

The occurrence of cyst in the mandibular condyle is due to epithelial off-shoots (hermartias) of the epithelium from basal layer [11]. Histologically, OKC has a orthokeratinized epithelial lining with 4–8 cell-layer thick, whereas the KCOT has thick parakeratinized epithelial lining whose basal cells have palisading nuclei [10,12]. All these microscopic observations were in our case, as well as its contents were full of keratin.

Treatment includes enucleation and curettage for small lesions and marsupialisation for large lesions. Surgery resection is when the cyst is wide and the cortical plate of bone is perforated. Acceptable results are obtained from marsupialisation. Because of the high recurrence rate of the OKC following the enucleation, it is recommended to use a Carnoy solution to fix the surrounding tissues. High recurrence rate of the OKC is due to proliferative activity of the epithelium cellsof the cyst [3]. Dong et al. exhibited that enucleation with or without curettage, combination of enucleation followed by marsupialisation and peripheral osteotomy

for large multilocular lesion had no reoccurrence [4]. In a study of Crowley, Kaugars and Gunsolle, only 4 % of OKCs displayed the recurrence risk [13,14]. However, in our case, due to the history of recurrence twice, and previous relatively ineffective biopsy as well as secondary infection on the lesion, we decided to separate the lesion from parotid into a piece together with the ramus and condyle resection. Surgical complications associated with removal of the lesion and mandibular resection include infection, bleeding commonly caused by branches of facial artery, masseter muscle and inferior alveolar neurovascular bundle and maxillary artery, and temporary facial paralysis, as well as mandibular deviation due to disconnection between temporalis, masseter and medial pterygoid muscles. We followed up our patient for 12 months and found no symptoms of recurrence and postoperative problems for the patient.

Conclusion

The keratocystic odontogenic tumor is relatively common in mandibular body and ramus, and some cases of its occurrence have been reported in condyle, but its simultaneous occurrence in the condyle and its expansion to parotide and passing through parotid to reach subcutaneous region has not been reported until now. Therefore, this lesion should be examined in the differential diagnosis of condyle lesions and parotid enlargement, and its treatment should be more extensive.

Conflict of interest

There is no conflict of interest.

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we would like to aknowledge all the staff and personells in surgery ward.

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