Case Report

Orthokeratinized odontogenic cyst: Report of eight cases and review of literature regarding its malignant transformation

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Abstract

Orthokeratinized odontogenic cyst (OOC) is an uncommon odontogenic cyst. It has been categorized as a subtype of odontogenic keratocyst (OKC). In 2005, it was classified as a distinct entity. OOC should be histopathologically differentiated from OKC, which has a higher recurrence rate and lower malignant potential. In addition, OOC should be examined for malignant transformation. The epithelium of odontogenic cysts may rarely show malignant transformation. However, malignant transformation has been reported in inflammatory cysts such as the residual cyst and periapical cyst. The number of carcinomas arising from an OOC is low. This paper describes eight cases of OOC; out of which, two showed the development of squamous cell carcinoma from their epithelial lining.

Keywords: Malignancy, malignant transformation, orthokeratinized odontogenic cyst, squamous cell carcinoma

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INTRODUCTION

Orthokeratinized odontogenic cyst (OOC) is an uncommon developmental odontogenic cyst. The jaws are the most common site of involvement. OOC was first reported as a subtype of OKC by Wright in 1981. OOCs account for about 7%–17% of all keratinized cysts in the jaws and 10% of odontogenic keratocysts (OKCs).[1,2] OOC is an odontogenic cyst that has an orthokeratinized epithelium in microscopic evaluation. It has been classified as a separate entity by the World Health Organization since OOC shows lower recurrence rate and higher malignant transformation than OKC.[1] The epithelium of odontogenic cysts can show keratinization or dysplasia.[3] Benign odontogenic tumors are the most common neoplasms arising from the epithelium of odontogenic cysts.[4] Nonetheless, mucoepidermoid carcinoma and squamous cell carcinoma (SCC) are two important malignancies that may arise from the epithelium of an OOC.[5] Malignant changes of the epithelium of odontogenic cysts are uncommon, and have a prevalence of 1–2/1000 cases.[6] Inflammatory cysts such as periapical or residual cysts (and not developmental cysts such as OOC) have the highest incidence of malignant

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transformation.[6] This paper describes eight cases of OOC; malignant transformation and development of SCC from the cyst lining occurred in two of them.

REPORT OF CASES

Case 1

A 31-year-old pregnant woman presented to dental clinic of Tehran University with a chief complaint of swelling of the anterior maxilla. Radiographic examination revealed a radiolucent, unilocular lesion in the anterior maxilla with corticated borders at the site of right canine tooth [Figure 1]. Radiographic and excisional biopsy findings were suggestive of a cystic lesion. Microscopic examination confirmed a cystic lesion as well. The cyst had orthokeratinized stratified squamous epithelium. Elongated bulbous rete ridges, nuclear pleomorphism, prominent nucleoli, and dyskeratotic cells with focal areas of invasion to the underlying connective tissue were noted in some areas of the epithelium. According to the histopathological findings, the diagnosis of SCC was made [Figure 2]. Another surgical procedure was suggested to the patient to obtain adequate safe margins but the patient refused to undergo another surgical procedure. After 10 months, the patient presented to another medical center complaining of an expansile lesion at the same area. The lesion was completely excised and sent for histopathological analysis, and the diagnosis of acanthotic ameloblastoma was made. The surgeon sent the paraffin blocks of the lesion to our center and asked for consultation. Microscopic examination revealed a tumor consisting of epithelial nests and islands of squamous cells with prominent acantholysis in a myxomatous stroma. Keratin pearls and necrosis were also seen in some foci. The tumor cells showed hyperchromatic nuclei, increased nuclear-cytoplasmic ratio, nuclear and cellular polymorphisms and increased mitotic activity [Figure 3]. Immunohistochemical staining for calretinin was performed to rule out acanthomatous ameloblastoma, which revealed no expression in the tumor cells [Figure 4]. Ki-67 immunostaining showed high proliferative activity of tumor cells [Figure 5]. The diagnosis of SCC was confirmed. The patient underwent *en bloc* resection, chemotherapy and radiotherapy, and showed no recurrence or metastasis after 1 year of follow-up.

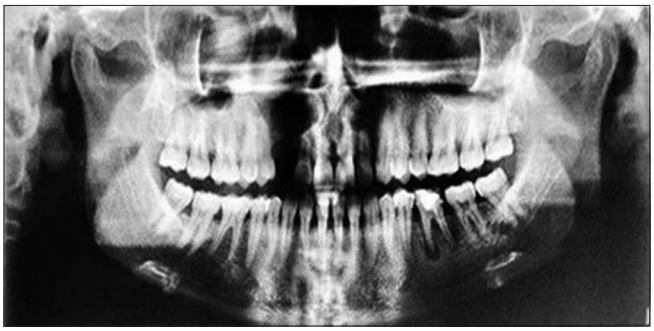

Figure 1: A unilocular radiolucent lesion with corticated borders in the anterior maxilla

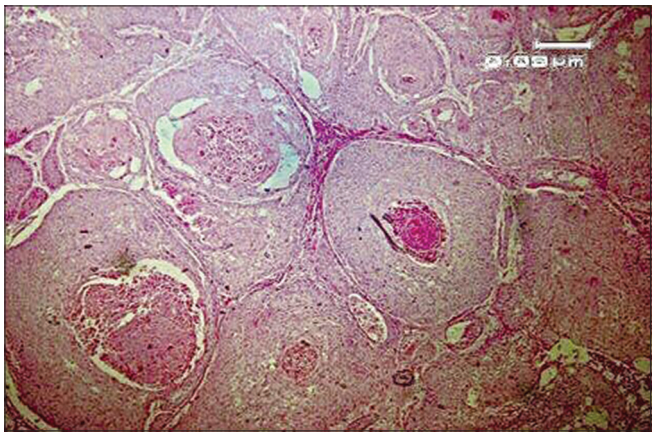

Figure 3: Squamous cell carcinoma with extensive necrosis

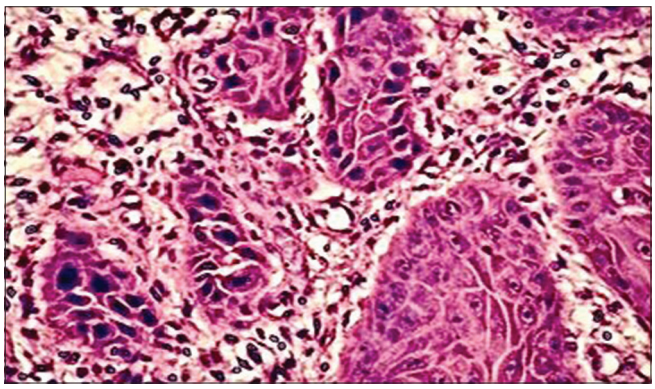

Figure 2: Nuclear pleomorphism, prominent nucleoli, and dyskeratotic cells with focal areas of invasion to the underlying connective tissue

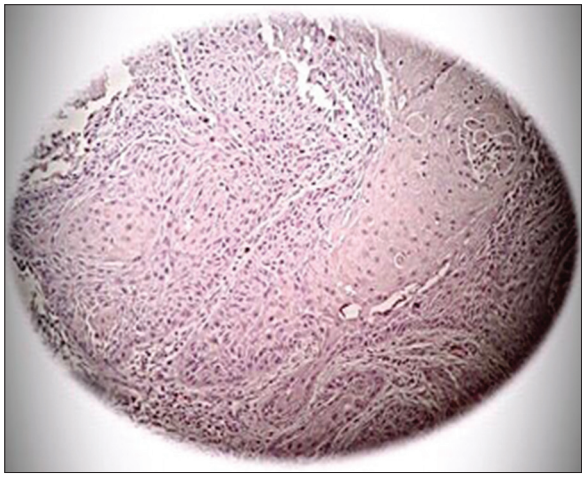

Figure 4: Immunohistochemical staining for calretinin was performed to rule out acanthomatous ameloblastoma, which revealed no expression in the tumor cells

Case 2

A 50-year-old male patient presented to dental clinic of Tehran University with a chief complaint of pain in the right maxilla for some time. Radiographic examination revealed a radiolucent unilocular lesion with corticated borders in the pericoronal area of maxillary right impacted third molar. Afterexcisional biopsy, gross evaluation showed several pieces of sheet-like tissue. Microscopic examination revealed a cystic lesion. The cyst had parakeratinized and orthokeratinized stratified squamous epithelium with elongated rete ridges that showed severe dysplasia. Verrucous hyperplasia was also present in some areas. Islands of dysplastic cells had invaded the connective tissue [Figure 6]. Atypical mitotic figures and keratin pearls were also seen. The cyst wall revealed mild, diffuse infiltration of chronic inflammatory cells and hemorrhage. The diagnosis of SCC arising from OOC was made. The patient died 2 years later.

Case 3

A 26-year-old female patient presented to dental clinic of Tehran University with a chief complaint of expansion and pain in the left mandible for 1 year. The covering mucosa was normal without any tenderness. Radiographic examination revealed a radiolucent lesion that was unilocular with corticated borders extending from the mandibular left second molar to the left impacted third molar [Figure 7]. After excisional biopsy, gross examination of biopsy specimen showed 2 pieces of cystic tissues, with a keratinized material inside their lumen. Microscopic evaluation showed a cystic lesion. The cyst had orthokeratinized stratified squamous epithelium with a prominent granular cell layer subjacent to the orthokeratinized layer. The cyst wall revealed mild diffuse infiltration of chronic inflammatory cells and hemorrhage. Orthokeratinized strands were seen within the cyst lumen [Figure 8]. The diagnosis of OOC was made.

Case 4

A 58-year-old female patient was referred to our department with a lesion that was accidentally detected in radiographic examination. The covering mucosa was normal without any tenderness. Radiographic examination revealed a radiolucent lesion that was unilocular with corticated borders extending from the pericoronal area of the right mandibular third molar to the mid-portion of

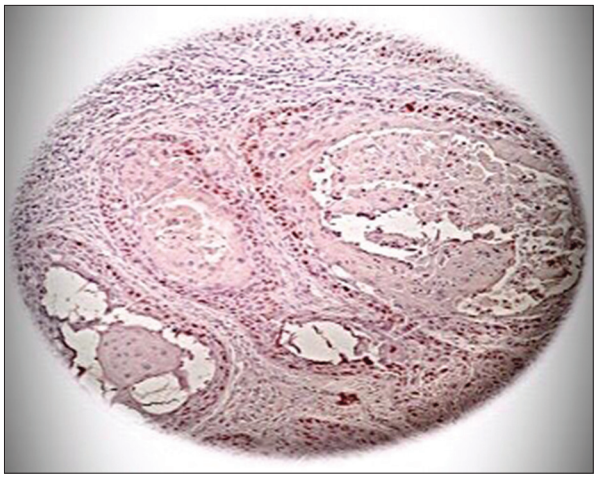

Figure 5: Ki-67 immunostaining showed high proliferative activity of tumor cells

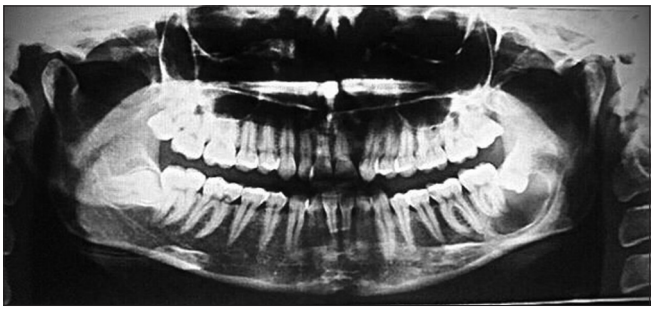

Figure 7: A unilocular radiolucent lesion with corticated borders extending from the mandibular left second molar to the mandibular left impacted third molar

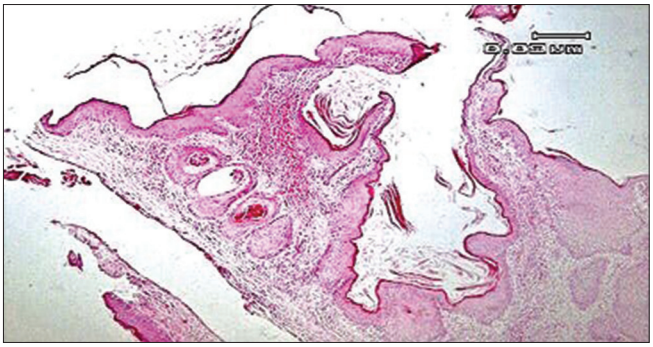

Figure 6: Verrucous hyperplasia seen in some areas. Islands of dysplastic cells invading the connective tissue are also seen

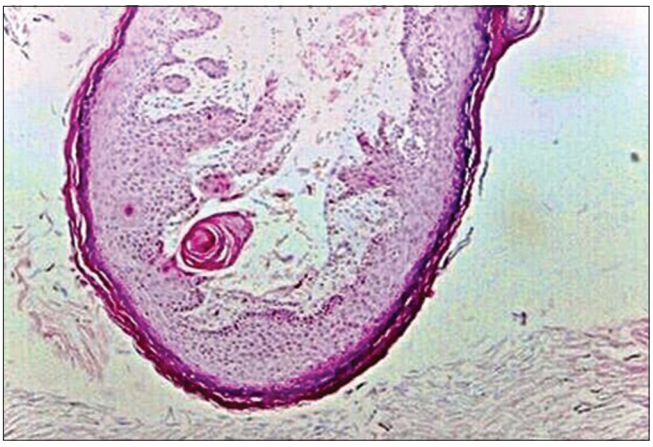

Figure 8: Orthokeratinized stratified squamous epithelial lining with prominent granular cell layer (H and E) and elongated rete ridges

the mandibular ramus [Figure 9]. Microscopic examination showed a cystic lesion. The cyst had orthokeratinized stratified squamous epithelium with varying thickness and prominent granular cell layer. The underlying connective tissue revealed diffuse mild infiltration of chronic inflammatory cells and sections of blood vessels. Orthokeratinized strands were seen within the cyst lumen. The diagnosis of OOC was made

Case 5

A 34-year-old male patient was referred to Tehran University with a chief complaint of swelling in the left posterior mandible. The covering mucosa was normal without any tenderness. Radiographic examination revealed an expansile unilocular radiolucent lesion with corticated borders extending from the mandibular left first molar to the left third molar. There was no impacted tooth [Figure 10]. Lingual expansion and perforation of the cortical plate were detected on cone-beam computed tomography scans [Figure 11]. Afterexcisional biopsy, gross examination of biopsy specimen revealed a cystic lesion that was elastic and creamy brown in color, measuring 2 cm × 1.5 cm × 0.2 cm. The microscopic examination showed a cystic lesion. The cyst had orthokeratinized stratified squamous epithelium with a prominent granular cell layer. Keratin strings were seen within the cyst lumen. The diagnosis of OOC was made.

Case 6

A 28-year-old female patient presented to Tehran University with a chief complaint of rapid expansion of the maxilla in the past month. The covering mucosa was normal without any tenderness. After excisional biopsy, gross examination of the biopsy specimen showed multiple pieces of irregular creamy tissues with fragile consistency totally measuring 2.8 cm × 2.2 cm × 1 cm, and two pieces of sheet-liked tissue totally measuring 2.5 cm × 0.5 cm × 0.4 cm. Microscopic examination showed a cyst. The cyst had orthokeratinized stratified squamous epithelium with a prominent granular cell layer. The underlying connective tissue revealed severe mixed inflammatory cell infiltration, collagen fibers, scattered fibroblasts, dystrophic calcification, extravasated red blood cells and bacterial colonies. The diagnosis of OOC was made.

Case 7

A 38-year-old male patient presented to the dental clinic of Tehran University. His chief complaint was expansion of the right side of the maxilla for 4 years. The covering mucosa was normal without any tenderness. After excisional biopsy, gross examination of the biopsy specimen showed multiple irregular pieces of soft tan brown tissue totally measuring 3 cm × 2.5 cm × 0.7 cm. Microscopic evaluation showed a cyst. The cyst had orthokeratinized stratified squamous epithelium with a prominent granular cell layer. The underlying connective tissue revealed diffuse severe chronic inflammatory cell infiltration. The diagnosis of OOC was made.

Case 8

A 53-year-old male patient presented to dental clinic of Tehran University. His chief complaint was swelling of the anterior maxilla. The covering mucosa was normal without any tenderness. Radiographic examination revealed a unilocular radiolucent lesion with corticated borders in the anterior right side of the maxilla with no associated impacted tooth. After excisional biopsy, gross examination showed one piece of gray, brown, cystic lesion measuring 3.2 cm × 2 cm × 1.2 cm. Microscopic examination showed

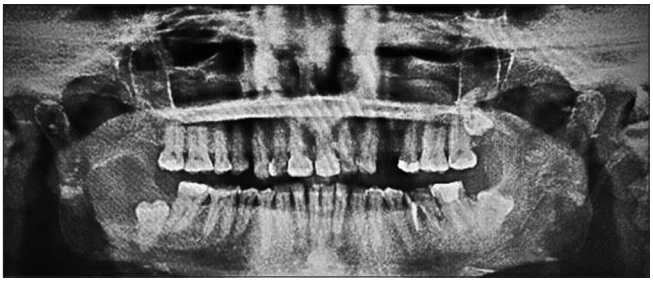

Figure 9: Panoramic radiograph showing a unilocular radiolucent lesion with corticated borders extending from the pericoronal area of the mandibular right impacted third molar to the mid-portion of the mandibular ramus

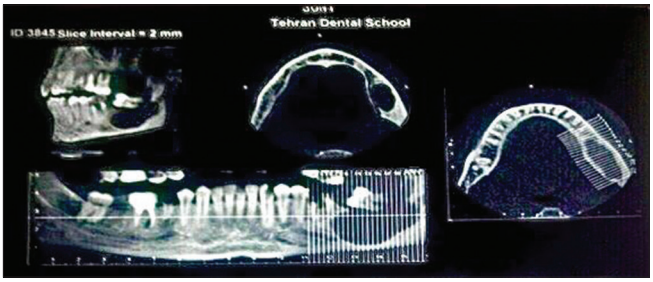

Figure 10: A unilocular radiolucent lesion with corticated borders extending from the left mandibular first molar to the left mandibular third molar

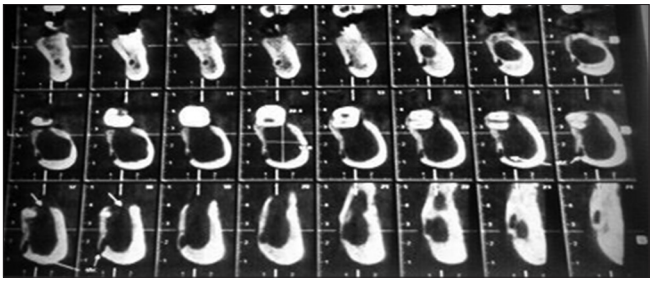

Figure 11: Cone-beam computed tomography scans showing lingual expansion and perforation of the cortical plate

a cystic lesion. The cyst had orthokeratinized stratified squamous epithelium with nodular thickening toward the lumen in some areas. A prominent granular cell layer was evident in the epithelial lining [Figure 12]. The diagnosis of OOC was made.

DISCUSSION

OOC is an uncommon developmental odontogenic cyst, and 5.2%–16.8% of the cases of OKC are re-categorized as OOC.[1] OOC accounts for approximately 11% of all the jaw cysts.[7] MacDonald and Li,[7] and Li *et al*.[8] reported that OOC was more common in males. However, in our case series, 5 out of 8 cases were female and only 3 were male.

Table 1 presents the OOC cases. Evidence shows that OOC more commonly occurs in the third and fourth decades of life,[1,7,8] which was the case for 6 out of 8 cases reported in this study; however, two of them were in their sixth decade of life. Moreover, MacDonald and Li[7] reported a preponderance for females in the second decade of life. The most common areas of involvement are the posterior mandible and the mandibular ramus region[8,9,12] but in our patients, maxilla and mandible were equally involved. OOCs are usually asymptomatic and the most frequent symptom is a slow-growing swelling, which is occasionally associated with pain.[12] In our study, one patient complained of pain and seven patients had swelling.OOC is a cystic lesion. The cyst has orthokeratinized stratified squamous epithelium with a very prominent granular cell layer. OKC must be considered as a differential entity. The epithelial surface of OKC is corrugated and the basal cell layer is palisaded.[9,13,14] The difference between OKC and OOC in keratin expression explains the difference in the characteristics of these two lesions.[15]

Malignancy has been rarely reported in OOC or OKC.[16] Evidence shows that the cysts with orthokeratinized surface have a higher risk of malignancy, but evidence is

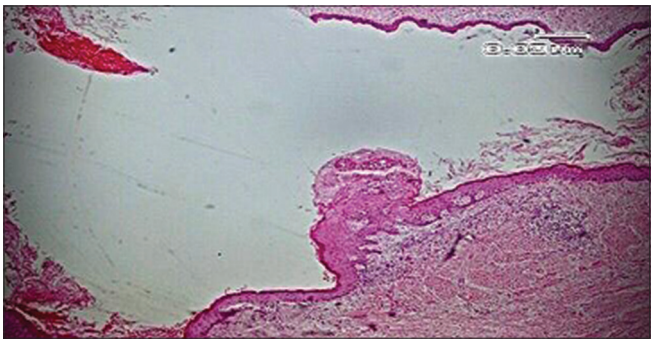

Figure 12: A cystic lesion lined by orthokeratinized stratified squamous epithelium with nodular thickening toward the lumen in some areas

Table 1: Orthokeratinized odontogenic cysts reported in the literature

Author/year	Number of cases	Gender Male:female	Mean age	Location Maxilla: Mandible	Pain Yes:no	Incidental findings Yes:no	Shape Unilocular: multilocular	Root resorption Yes:no	Tooth Yes:no	Swelling Yes:no	Mean size	Unerupted tooth Yes:no	Recurrence Yes:no
Dong *et al*., 2010[1]	61	ING	ING	6:55	13:48	ING	47:7	ING	ING	46:61	4:8	27	0:42
Macdonald and Li 2010[7]	5	2:3	34:20	2:4	2:3	0:5	3:2	0:5	4:1	ING	ING	3:2	ING
González Galván Mdel *et al*., 2013[9]	3	2:3	33.6	0:3	1:2	0:3	2:1	0:3	1:2	2:1	ING	2:1	ING
Manickam *et al*., 2014[10]	11	8:3	45.5	ING	ING	ING	ING	ING	ING	ING	ING	ING	ING
Vera-Sirera 2016[11]	12	7:5	35,50± 12,10	0.0019	ING	ING	ING	ING	ING	ING	ING	ING	ING
Our case series	8	3:8	37.6	4:4	1:7	ING	5	ING	5:3	7:1	ING	3:5	ING

ING: Information not given

Table 2: Cases of squamous cell carcinoma arising from Orthokeratinized odontogenic cyst and odontogenic keratocyst reported in the English literature from 2006 to 2016

Authors/ yearsdiagnosis	Age/sex	Duration	Clinical features	Site	Diagnosis
Falaki *et al*.,2009[19] SCC arising OKC	20/male	25 days	Pain, swelling, mass	RT MN posterior	SCC arising OKC
Chaisuparat *et al*.,2006[20] IC arising OC	18/female	-	Swelling	Anterior maxilla	IC arising OC
Chaisuparat *et al*.,2006[20] IC arising OC	46/male	-	Pain, paresthesia	Posterior mandible	IC arising OC
Sato *et al*.,2006[21] SCC arising KOT	76/female	3 days	Pain, swelling	sinus (D) LT MN posterior	SCC arising KOT
Mitchell *et al*.,2010[22] SCC arising OKC	63/male	4 months	Painful swelling, epistaxis	LT MX posterior	SCC arising OKC
Maria *et al*.,2011[23] SCC arising OKC	54/male	6 months	Swelling	RT MX posterior Opacification-MX sinus	SCC arising OKC
Lee *et al*.,2011[24] SCC arising KOT	27/male	-	Pain, swelling	Sinus (D) Rt MN posterior Unerupted 3rd molar	SCC arising KOT
Bereket *et al*.,2013[6] IC arising OC	26/male	3 years	Pain, swelling, sinus (D), FNAC– brownish liquid	RT MX anterior	IC arising OC
Acharyaa *et al*.,2013[25] SCC arising OOC	30/male	8 months	Pain, swelling, sinus, (D), FNAC+	RT MN posterior	SCC arising OOC
Tamgadge *et al*.,2013[26] SCC arising OKC	20/female	Several months	Pain, swelling	LT MN posterior	SCC arising OKC
Jain *et al*.,2013[27] SCC arising OC	70/male	8 months	Swelling, numbness	RT MN anterior	SCC arising OC
Akheel *et al*.,2014[28] CT – mass	66/female	1 month	Swelling, FNAC+	LT MN posterior	CT – mass
Sexena *et al*,2015[29] SCC arising OKC	60/male	7 month	sweling, dull pain, paresthesia	RT MN	SCC arising OKC
Martínez-Martínez *et al*., 2016[30] SCC arising OKC	37/female	9 months	Slow growth, swelling	RT MN	SCC arising OKC

SCC: Squamous cell carcinoma, IC: Intraosseous carcinoma, OC: Odontogenic cyst, KOT: Keratinizing odontogenic tumor, OKC: Odontogenic keratocyst, MX: Maxilla, MN: mandible, RT: Right, LT: Left, CT: Computed tomography

inconclusive on this topic.[2] However, OKCs have a more aggressive behavior than other types of odontogenic cysts.[17] A review study reported 15 cases of SCC developing from OCC or other types of odontogenic cysts.[3] A review on primary intraosseous SCCs arising from odontogenic cysts showed that 60% of cases were related to inflammatory odontogenic cysts such as residual and radicular cysts, and 14% (16 cases) were related to OKC or OOC.[18]

Table 2 shows malignancies such as SCC arising from OOC and OKC. In our study, two of eight cases showed malignant changes in the cyst lining. Carcinomas developing from an odontogenic cyst are scarce. However, SCC is the most common type of malignancy arising from odontogenic cysts.[6] In our study, two patients had SCC. The most important etiologies for carcinomas include genetics, smoking and alcohol consumption.[17] Malignant changes of odontogenic cyst epithelium often require 30–40 years.[18] Malignant transformation occurs faster in inflammatory cysts.[18] In contrast, in our report, both cases of OOC with malignant transformation occurred in young women (26 and 31-year-old). This may suggest that the nature of the lesion in cases in whom, the malignancy develops at a younger age within a shorter period of time is different from SCC that develops in odontogenic cysts after a long period of time. The lesion may be a primary intraosseous carcinoma with a cystic growth pattern in cases in whom, the malignant lesion develops within a short period of time; whereas, in patients who have a history of a preexisting cystic lesion for many years, malignant transformation occurs in the epithelium of the cyst after a long period of time.

CONCLUSION

OOCs are scarce developmental cysts that require careful consideration. They should be differentiated from the more common OKCs, which have a higher recurrence rate and lower malignant potential. OOCs should be carefully examined to rule out any malignant transformation.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient (s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal.

The patients understand that their names and initial s will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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