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Ectopic upper third molar embedded in a dentigerous cyst of the maxillary sinus: a case report and literature review

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Abstract

Background Dentigerous cysts are the second most common form of a developmental cyst. These cysts might be the cause of ectopic teeth. Dentigerous cysts associated with ectopic teeth in the maxillary sinus are seldom seen in clinical practice.

Case presentation We reported a 23-year-old woman who presented with nasal obstruction and facial pain on the left side 4 months ago. Three courses of antibiotics and analgesia were prescribed for her without improving the presenting features. Furthermore, extraction of the left upper second premolar and second molar, but the condition persisted. An orthopantogram and computerized tomography scan revealed the ectopic position of the left third upper molar tooth, which was embedded in a cystic lesion occupying the whole maxillary sinus. Removal of the cystic lesion with the ectopic tooth was performed with a Caldwell-Luc procedure. The histopathological evaluation confirmed the diagnosis of a dentigerous cyst. The presenting symptoms were resolved following surgery with no intra- or postoperative complications.

Conclusion We are reporting another case of a dentigerous cyst occupying the whole left maxillary sinus with an ectopic left third upper molar tooth. Reporting such a case will enrich the literature regarding this rare clinical entity.

Keywords Dentigerous cyst, Upper third molar, Ectopic tooth, Maxillary sinus, Case report

Background

Paget, in 1853, described the term "dentigerous cyst." A dentigerous cyst is one of the developmental odontogenic cysts. It is the second most common form of these cysts. It could arise from the crowns of embedded, impacted, or unerupted teeth [1]. Dentigerous cysts comprise around 20% of the jaws' cysts lined by epithelial tissue. The mandibular third molar and maxillary canine are the most commonly involved teeth. Dentigerous cysts occur in the mandible in 70% of cases, while only 30% of them are in the maxilla [2].

Displacement of the impacted tooth can occur due to a dentigerous cyst in an ectopic location like the mandibular condyle, coronoid process, nasal cavity, and maxillary antrum [3]. There are possible etiological theories that explain the pathogenesis of ectopic teeth, including developmental abnormalities, trauma, infection, and pathological problems like dentigerous cysts. However, the exact mechanism remains unclear [2].

In the majority of cases, dentigerous cysts involve the lower jaw with third molar involvement. While the upper

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jaw is less affected and mostly involves canines. Dentigerous cysts are usually not associated with pain. However, if they are associated with an infection, there is a painful swelling [1]. The ectopic tooth embedded in a dentigerous cyst of the maxillary antrum is extremely rare in daily clinical practice. Only 20 such cases were reported in the midline from 1980 to 2010 [2]. The only reported case from Iraq when searching the midline was an ectopic upper third molar tooth in the maxillary sinus, but it was not associated with a dentigerous cyst [4]. We reported a further case of the left ectopic maxillary third molar tooth being into the maxillary antrum and embedded in a dentigerous cyst, as well as a review of similar cases in PubMed from January 1990 to April 2022.

Case presentation

A 23-year-old female was referred to the Department of Oral and Maxillofacial Surgery complaining of left-sided facial pain and nasal obstruction for the last 4 months. The patient received three courses of medicine, but the presenting features persisted. Moreover, the patient underwent extraction of the left upper second premolar and upper second molar to remove the pain just a few weeks ago, but without benefit. Clinically, the patient was healthy with an unremarkable medical history. Intra-oral examination revealed normal mucosa with slight bony expansion on the left side of the zygomatic buttress and buccal vestibule, as well as the absence of the upper left third molar tooth. Radiographic orthopantomogram (OPG) examination and computerized tomography (CT) scan revealed the presence of a tooth-like structure within the left maxillary sinus just below the orbit (Figs. 1 and 2). It was suggested that an ectopic erupted upper left third molar with a large dentigerous cyst involving the whole antrum was compressing the lateral wall of the nasal cavity and floor of the orbit, depending on the clinical and radiological judgment. However, we put the following as differential diagnoses: maxillary sinusitis, infected dentigerous cyst, and odontogenic keratocyst.

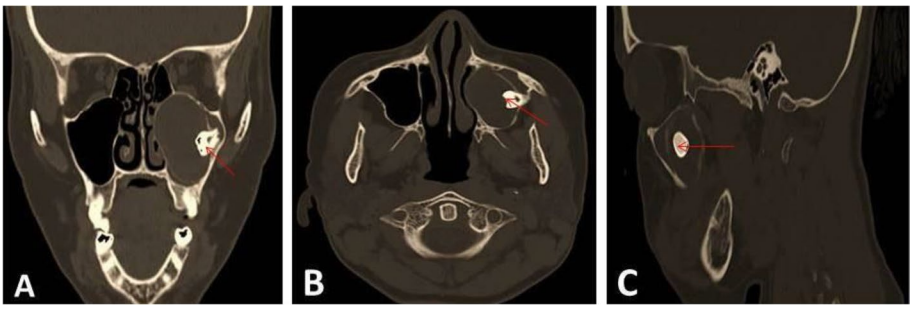

Fig. 1 Computerized tomography (CT) scans (**A** coronal, **B** axial, and **C** sagittal) revealed an ectopic left third molar tooth embedded in a cystic lesion occupying the whole maxillary antrum (red arrows). The lesion compresses the left lateral nasal wall and orbital floor

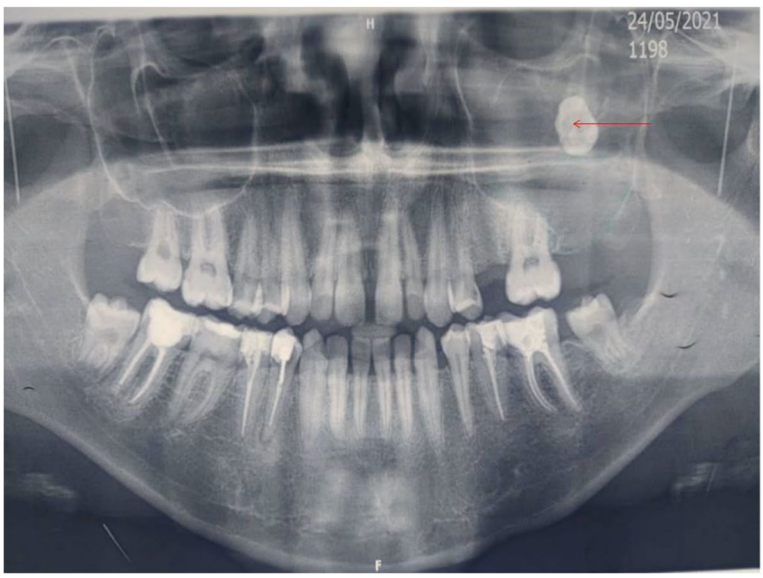

Fig. 2 Orthopantogram (OPG) shows total haziness of the left maxillary sinus and tooth-like structure (red arrow)

We prepared the patient for enucleation of the lesion as well as extraction of the ectopic tooth. Under general anesthesia with orotracheal intubation, enucleation of the cyst through a Caldwell-Luc operation was planned. The surgical procedure was done by an oral and maxillofacial surgeon with 12 years of experience. The operation starts by making a left sub-labial incision, creating a bone window in the anterolateral wall of the maxillary antrum. Complete removal of the whole cyst with the ectopic tooth (Figs. 3 and 4) was done, and the specimen was sent for histopathological examination. The pathological report confirms the diagnosis of a dentigerous cyst, as the lining of the cyst was

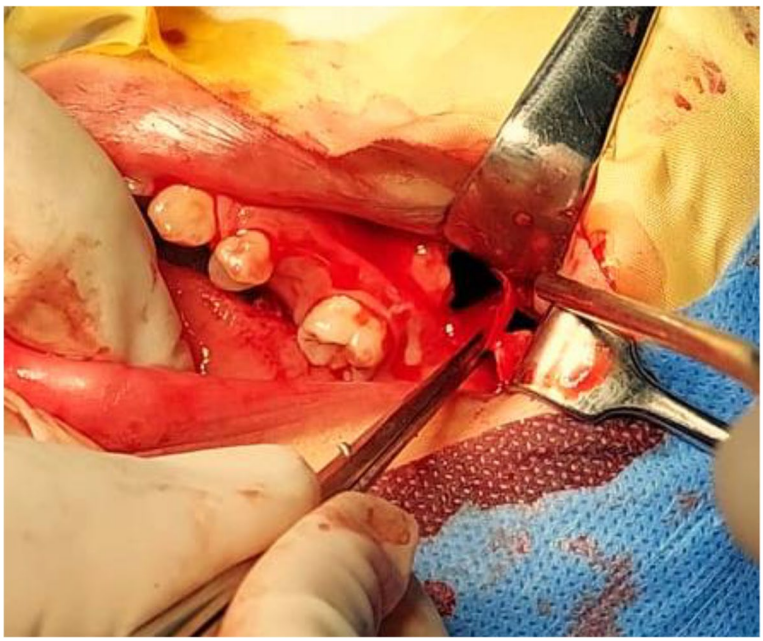

Fig. 3 Intraoperative image shows the opening in the anterolateral wall of the left maxillary sinus

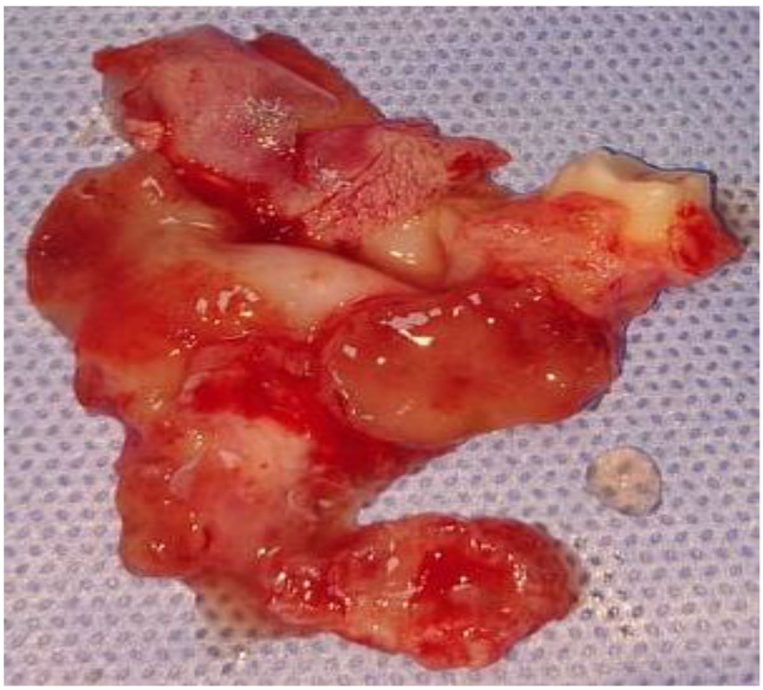

Fig. 4 The surgical specimen contains the dentigerous cyst and an ectopic third molar tooth

non-keratinized squamous epithelium (Figs. 5 and 6). Copious irrigation of the involved sinus was performed with normal saline to clear the operative field. Closure of the wound was performed with catgut sutures. The patient was put on an antibiotic (Augmentin 625 mg three times daily), analgesia (paracetamol 500 mg three times daily), and Medrol tablet (methylprednisolone) 4 mg once daily to lessen the postoperative cheek swelling. The patient was advised not to blow her nose for 14 days. The patient became completely free of the presenting complaints following surgery. After 6 weeks of follow-up, there was complete healing with good function and excellent patient satisfaction. Informed consent was obtained from the patient for publishing the case and its associated images. The study was approved by the Ethical Approval Committee of the University of Anbar (Reference Number 90 on June 25, 2023).

Discussion

There are two types of jaw developmental cysts: odontogenic which arises from odontogenic tissue like dental lamina or enamel organ remnants and non-odontogenic which arises from ectodermal tissue responsible for facial tissue development. Radicular cysts are the commonest type of odontogenic cysts followed by dentigerous cysts (70% in the mandible and 30% maxilla) [2].

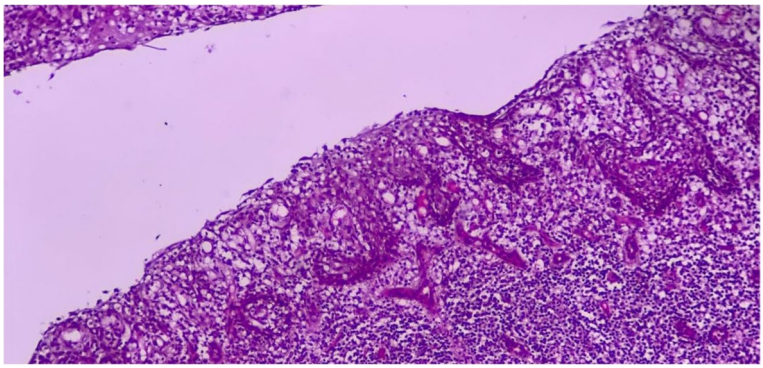

Fig. 5 Histopathological image shows the cystic lesion lined by non-keratinized squamous epithelium (hematoxylin–eosin, magnification × 10)

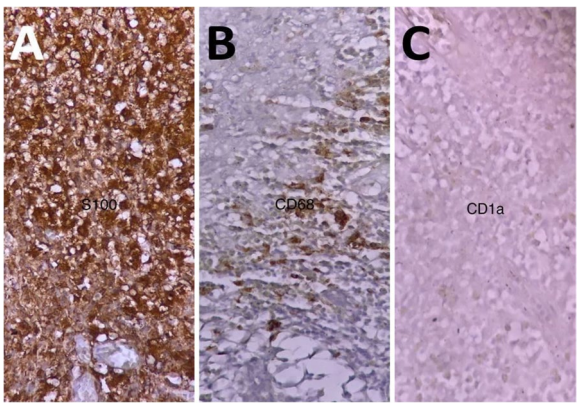

Fig. 6 An immunohistochemical study shows: **A** S100-positive Langerhans cell numbers in the lining epithelial and subepithelial connective tissues confirmed the association of high-grade inflammation and thick lining epithelium with the increased Langerhans cell number in a dentigerous cyst. **B** CD68 expression in Langerhans cells was present in defense mechanisms with the release of pro-inflammatory cytokines, which is responsible for inducing the continued proliferation of cystic epithelium. **C** CD1a-positive Langerhans cell numbers in the lining epithelial and subepithelial connective tissues confirmed the association of inflammation and thick lining epithelium with the increased Langerhans cell number in a dentigerous cyst (magnification × 10)

Eruption of teeth in their non-anatomical positions is a rare clinical entity. It is usually without symptoms and is discovered on routine radiological examination for other reasons [17]. However, if it is associated with a dentigerous cyst, various symptoms might develop, including, but not exclusively, nasal blockage, purulent discharge, cheek swelling, Epiphora, etc. (Table 1). Our patient presented with left-sided facial pain and nasal obstruction. The patient received three courses of antibiotics as well as the extraction of two left upper teeth but without benefit. Therefore, great care is necessary to catch the diagnosis early with a simple diagnostic tool (OPG) to avoid unnecessary treatment, particularly the extraction of the teeth, which is not the cause of the patient's complaints.

As reported in the literature, the majority of the cases were from Egypt (12/28), India (7/28), and Turkey (6/28). The age of patients with dentigerous cysts and ectopic upper molar teeth in the maxillary antrum ranged from 13 to 63 years. However, only 4 out of 28 cases occurred in children. There was a male predominance (16/28). The majority of the cases were unilateral (26/28) (Table 1). Our patient was a female, 23-year-old, and on the left side.

Table 1 The reported cases of the dentigerous cyst with a third molar ectopic tooth in the maxillary sinus from January 1990 to April 2022 in the midline. *CT*, computerized tomography; *OPG*, orthopantomogram

Authors	Year	Country	Age per years	Gender	Side	Presenting symptom	Radiological tool	Treatment approach
Hasbini et al. [5]	2001	Lebanon	21	Male	Left	Left-sided nasal obstruction	CT scan	Endoscopic sinus surgery
Di Pasquale and Shermetaro [1]	2006	USA	14	Female	Left	Asymptomatic	CT scan	Endoscopic sinus surgery
Srinivasa et al. [6]	2007	India	45	Male	Right	Pain, swelling, and purulent nasal discharge	CT scan	Caldwell-Luc
Micozkadioglu and Erkan [7]	2007	Turkey	24	Female	Right	Headache, facial pain, and facial swelling	OPG and CT scan	Endoscopic sinus surgery
Buyukkurt et al. [2]	2010	Turkey	19	Female	Left	Left cheek swelling	CT scan	Caldwell-Luc
			30	Male	Left	Enlarged soft swelling of the left maxilla	CT scan	Caldwell-Luc
Thakur et al. [8]	2011	India	25	Male	Left	Chronic cough, recurrent nasal purulent discharge, and facial pain	OPG	Caldwell-Luc
Kasat et al. [9]	2012	India	22	Male	Right	Oral and nasal discharge	OPG and CT scan	Caldwell-Luc
Guruprasad et al. [6]	2013	India	21	Female	Right	Cheek swelling and purulent nasal discharge	Paranasal sinuses X-ray, OPG, and CT scan	Caldwell-Luc
Mamatha et al. [10]	2014	India	17	Male	Left	Offensive nasal discharge with mouth salty discharge	OPG and CT scan	Caldwell-Luc
Demirtas et al. [11]	2014	Turkey	19	Male	Right	Pain, discomfort, fullness in the right cheek, and blurred vision	OPG and CT scan	Caldwell-Luc
Muhammed et al. [12]	2015	Turkey	16	Female	Right	Painful cheek swelling	OPG and CT scan	Caldwell-Luc
Liau et al. [13]	2018	Australia	63	Male	Right	chronic nasal obstruction and intraoral discharge	OPG and CT scan	Endoscopically assisted modified Caldwell-Luc
Sharma and Chauhan [3]	2019	India	27	Female	Bilateral	Purulent nasal discharge and recurrent facial swelling	OPG and CT scan	Caldwell-Luc
Elmorsy et al. [14]	2020	Egypt	13	Female	Right	Offensive oral discharge at the upper right second molar	OPG and CT scan	Caldwell-Luc
Balaji SM, Balaji P [15]	2020	India	42	Male	Right	Occasional pain in the right upper posterior tooth	OPG and CT scan	Caldwell-Luc

Table 1 (continued)

Authors	Year	Country	Age per years	Gender	Side	Presenting symptom	Radiological tool	Treatment approach
El-Fattah et al. [16]	2021	Egypt	43	Male	Left	Loosening of tooth	CT scan	Endoscopically assisted Caldwell-Luc
			30	Male	left	Asymptomatic	CT scan	Endoscopically assisted Caldwell-Luc
			35	Female	Right	Features of sinusitis	CT scan	Endoscopic sinus surgery
			39	Female	left	Proptosis	CT scan	Endoscopic sinus surgery
			25	Male	Right	Asymptomatic	CT scan	Endoscopic endonasal pre-lacrimal
			40	Female	Right	Features of sinusitis	CT scan	Endoscopically assisted Caldwell-Luc
			33	Female	left	Asymptomatic	CT scan	Endoscopic sinus surgery
			31	Male	left	Ocular pain	CT scan	Endoscopic sinus surgery
			48	Male	Right	Features of sinusitis	CT scan	Endoscopically assisted Caldwell-Luc
			31	Male	left	Loosening of teeth	CT scan	Endoscopically assisted Caldwell-Luc
			28	Male	Right	Asymptomatic	CT scan	Endoscopic endonasal pre-lacrimal
Arici et al. [17]	2022	Turkey	32	Female	Bilateral	Postnasal discharge, headache, nasal obstruction, and pain in the blinking right eye	CT scan	Caldwell-Luc

The majority of the reported cases of maxillary dentigerous cysts and ectopic teeth were single cysts; however, multiple cysts were also reported [18].

The diagnosis of an ectopic tooth in the maxillary sinus is usually straightforward through radiological investigations. Various tools are used, as described in the literature, including plain X-rays, OPG, CT scans, and cone beam computerized tomography (CBCT). The sophisticated tools (CT scan and CBCT) have a precise ability to localize the lesion and are superior to panoramic imaging. Moreover, CBCT gave a three-dimensional image of the ectopic tooth as well as its inclination and relationship to the maxillary antrum, which is helpful in the planning of surgery [3]. MRI was used by some researchers in the assessment of dentigerous cysts with an ectopic tooth in the maxillary sinus [19].

The standard operative technique for the dentigerous cyst is complete enucleation of the cyst and its associated ectopic tooth through a Caldwell-Luc surgical procedure. Marsupialization might be an initial step in the removal of large cysts before the complete enucleation process. However, the major drawback of marsupialization is the remnant of an early recurrence of the lesion. Owing to the high experience of functional endoscopic sinus surgery, this approach is advocated. The endoscopic approach has fewer intra- and postoperative complications than the Caldwell-Luc procedure [1]. In children, where a natural eruption is still possible, the Caldwell-Luc approach is not possible. Therefore, marsupialization of the cyst with the ectopic tooth is an option [20]. The majority of the reported cases in the midline were treated by cyst enucleation and extraction of the associated ectopic tooth through a Caldwell-Luc procedure. Our case was treated through the Caldwell-Luc approach and total enucleation of the cyst and the ectopic tooth because this procedure has many characteristics: direct exposure of the maxillary antrum, making easy usage of the instruments; excision of large lesions; and irrigation of the sinus at the end of the operation. Recently, the endoscopically assisted Caldwell-Luc approach was used [13, 16]. This approach differs from the classical Caldwell-Luc procedure in that the incision and bone drilling in the anterolateral maxillary wall are much smaller. Therefore, we can greatly avoid the classic Caldwell-Luc complications like

postoperative pain and edema, injury to the infraorbital nerve, and oroantral fistula [16].

Langerhans cells provide important insight into the immunopathogenesis of chronic periapical lesions such as dentigerous cysts, especially when an inflammatory process is noticed in patients who complain of pain. A clear correlation was observed between Langerhans cell-positive immunohistochemical expression in the lining epithelial tissues and epithelial thickness, as well as the inflammatory intensity in the reported dentigerous cyst. For this reason, it is advisable to conduct an immunohistochemical staining of Langerhans cells in chronic periapical lesions, including dentigerous cysts to properly assess the case, better understand the response of these lesions, and confirm any malignant transformation.

Conclusion

An ectopic upper third molar tooth associated with a dentigerous cyst in the maxillary sinus is seldom seen in clinical practice. This entity mostly affects one side. A CT scan and OPG are diagnostic tools to identify this lesion. Endoscopic or Caldwell-Luc approaches or both are the treatment options for this condition. Owing to the rarity of ectopic teeth embedded in the dentigerous cyst in the maxillary antrum, there is no consensus about the best option of treatment that deserves to be included in the literature.

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None.

Authors' contributions
Aldelaimi TN, Khalil AA, and Abdulkareem SM analyzed and interpreted the patient data regarding the clinical and pathological findings as well as writing the basic manuscript draft. Al-Ani RM formatted the references and wrote the final manuscript draft. All authors read and approved the final manuscript.

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Availability of data and materials
All patients' data was presented in the article.

Declarations

Ethics approval and consent to participate
This study was approved by the Ethical Committee of the University Of Anbar (Reference number 90 on 25–6-2023). Written informed consent was obtained from the patient for the publication of this case report and accompanying images.

Consent for publication
The patient gave written informed consent for the publication of the data and materials contained within this study.

Competing interests
The authors declare that they have no competing interests.

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