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Calcifying epithelial odontogenic tumor with maxillary sinus extension: Case report and therapeutic review

Antônio Augusto de Melo da Silva, Tiago de Arruda Martins, Henrique Rocha Mazorchi Veronese, Michelle Inês e Silva

ABSTRACT

Calcifying epithelial odontogenic tumor (CEOT) is a rare benign neoplasm, with slow, localized, invasive, and asymptomatic growth. The involvement of the maxillary sinus by the neoplasm is rare, with its treatment controversial. The aim of this study was to describe the clinical, imaging, and therapeutic characteristics of a CEOT with maxillary sinus extension, as well as a literature review of therapeutic approaches and the prognoses obtained from cases of the same extension. In this case report, we report the case of a female patient, 49 years old, Caucasian, with mild asymmetry of the middle third of the face. Clinical and imaging examinations showed an intraosseous tumor in the posterior region of the left hemimaxillary, with imprecise limits and extension of 44×24×32 mm, compromising the alveolar process, maxillary posterior teeth, posterior hemipalatal region, left maxillary sinus, and orbital floor, associated with local expansion, tooth mobility, maxillary sinusopathy, and nasal obstruction. Calcifying epithelial odontogenic tumor diagnosis was obtained from incisional biopsy and histopathological examination. Surgical therapy of partial maxillectomy was performed from the Weber Ferguson Access with subsequent prosthetic rehabilitation. There were no postoperative complications. This case presented had satisfactory success with the therapy performed. The use of invasive therapies such as partial maxillectomies associated with transfacial approaches is an effective treatment for CEOT involving the maxillary sinus. Long-term follow-up is essential to avoid recurrences.

Keywords: Calcifying epithelial odontogenic tumor, Maxillary neoplasm, Odontogenic tumors, Pindborg tumor

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INTRODUCTION

Calcifying epithelial odontogenic tumor (CEOT), also known as Pindborg tumor, is a rare benign neoplasm, with slow, localized, invasive, and asymptomatic growth [1], responsible for approximately 1.6% of all odontogenic tumors and 0.03% of all oral and maxillofacial lesions biopsied [2]. Its origin, although not completely elucidated, has been associated with reduced enamel epithelium, the intermediate stratum of the enamel organ, remnants from the dental lamina, or oral epithelium [3].

Calcifying epithelial odontogenic tumor has no sexual predilection, being more frequent in the third and fourth

decades of life. Its intraosseous presentation is the most common (90%), with rare extraosseous cases. The mandible is the most prevalently affected region, with high involvement of the posterior region of premolars and molars, and dental association in half of the cases [4].

The characteristic histological findings of CEOT are cords and/or islands of polyhedral neoplastic epithelial cells, with eosinophilic cytoplasm, different degrees of nuclear pleomorphism, and presence of amyloid protein that tends to calcify [3, 5]. Its radiographic presentation is diverse, with interposed radiolucent and radiopaque areas of unilocular or multilocular appearance [3]. Other findings such as: presence of bone expansion and erosion, impacted or erupted teeth, tooth displacement, and root resorption can be found [2, 4]. Maxillary sinus involvement is rare and is commonly associated with the presence of facial edema, pain, nasal obstruction, respiratory distress, headache, epistaxis and proptosis [6].

In view of the infiltrative characteristics of the tumor, the treatment of choice is surgical, ranging from less invasive procedures, such as curettage and enucleation, to more invasive therapies for segmental or marginal resection [2, 4]. The patient's follow-up for five years after treatment is indicated due to the risks of recurrence, which vary between 15% and 30%, being higher in cases of conservative management [5].

Less than 400 cases of CEOT have been described in the literature [2, 4]. The incidence in the maxilla with involvement of the maxillary sinus is even rarer, with only 29 cases described in the literature. The present study reports a case of extensive CEOT in the maxilla with involvement of the maxillary sinus, showing its clinical, radiographic, histopathological, and therapeutic characteristics, and a review of the surgical management in similar cases previously described in the literature.

CASE REPORT

Female patient, 49 years old, Caucasian, referred by the Primary Care of the Municipality of Cataguases-MG to the Oral and Maxillofacial Surgery Department of the Dental Specialties Center-CEO Marília Guimarães Costa for evaluation of asymptomatic swelling in the posterior region of the left maxilla with six months of evolution. The patient was without medical comorbidities, with only previous anxiety cases. Extraoral examination revealed asymmetry of the middle third of the face, without the presence of lymphadenopathy (Figure 1A). The intraoral examination revealed the presence of a firm, nodular, sessile, and well-defined palpable tumor in the left hemimaxillary region, with involvement of the alveolar process, ipsilateral maxillary posterior teeth, and palatal region, with local expansion and tooth mobility associated. The adjacent mucosa had a normal appearance.

An orthopantomography revealed an extensive non-delimited unilocular mixed image (radiolucid and radiopaque) in the left maxilla, without dental association, with extension of the canine region to the maxillary tuber (Figure 1B). The maxillofacial tomographic examination revealed the presence of mixed intraosseous lesion with imprecise limits, affecting the left hemimaxillary region, palatine process, left lateral wall of the nasal cavity, orbital floor, and ipsilateral maxillary sinus, with the presence of maxillary sinusopathy, mild nasal obstruction and perforation of the cortical bone peripheral to the tumor (Figure 1C and D). From 3D reconstruction of computed tomography (CT) the dimension of the lesion was obtained, with an approximate measure of 44×24×32 mm. The diagnostic hypotheses were based on bone dysplasia, odontogenic myxoma and CEOT.

After routine preoperative examinations, an incisional biopsy of the lesion, under local anesthesia, using the Caldwell-Luc Accession, was performed. Histopathological analysis was compatible with CEOT.

The patient was referred for evaluation and therapy at the Department of the Maxillofacial Surgery and Traumatology of the Casa de Caridade de Muriaé Hospital São Paulo, MG. The surgical therapy based on left partial maxillectomy from Weber-Ferguson extraoral access by Dieffenbach's Modification (Figure 2A) was carried out by the team of specialists in maxillofacial surgery and traumatology of the hospital, under general anesthesia and nasotracheal intubation, due to extension of the lesion and the risks of tumor recurrence. After divulging and displacing the facial tissues, an osteotomy in the region of the maxillary midline, piriform contour, anterior maxillary wall, infraorbital margin, left maxillary zygomatic suture, and maxillary pterygoid process was performed to remove the lesion (Figure 2B). A flap of the facial fat pad was used to fill the remaining maxillary bone defect class 2 (Figure 2B) and a flap from the cheek mucosa was made to close the left palatal region. Extraoral and intraoral sutures with 4-0 vicryl and 5-0 black nylon were used for surgical synthesis (Figure 2C and D).

The patient was medicated and referred to the intensive care unit of the same hospital for monitoring of vital signs for 24 hours. The immediate postoperative period proceeded without any complications and the patient was discharged 72 hours after the surgery.

The surgical piece measuring 50×50×30 mm was sent for histopathological evaluation (Figure 3A). The microscopic findings revealed a neoplasm consisting predominantly of nests and strands of cells, with round and oval, hypercolored nuclei, eosinophilic cytoplasm, with some evident nucleoli and rare mitoses, dispersed in a collagenized stroma. Multiple focuses of concentric Liesegang ring-like calcifications and focal epithelial cells surrounding amorphous eosinophilic material were observed amid mature bone trabeculae (Figure 3B and C). Clinical, histopathological and imaging findings confirmed the final diagnosis of CEOT.

The patient was referred for routine post-surgical outpatient follow-up, with adequate extra-oral and intra-oral healing after a 5-month postoperative evaluation

(Figure 4A and B). A provisional dental prosthesis was made to rehabilitate the functions of the stomatognathic system (Figure 4C). The case is under follow-up, in order to minimize the chances of recurrence and provide early intervention in case of recurrence. A helical tomography performed ten months after surgery showed free margins and no tumor recurrence (Figure 5A and B).

This study was approved by the Research Ethics Committee of the FAMINAS University Center, Muriaé, MG, Brazil, under CAAE 39289720.3.0000.5105, and the Informed Consent Term (TCLE), containing the Confidentiality and Confidentiality Term was signed by the patient.

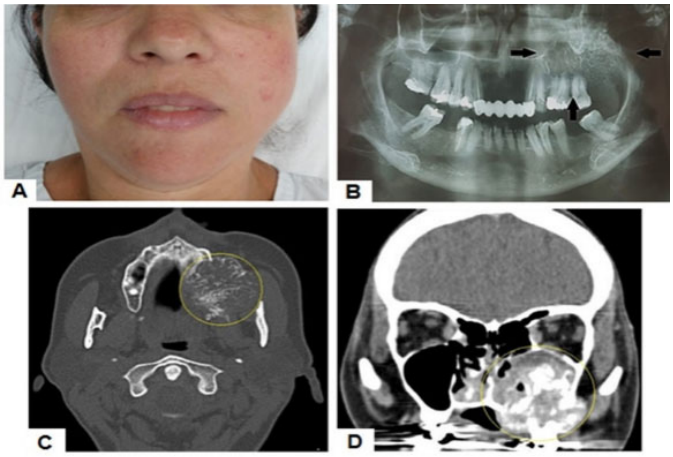

Figure 1: Clinical and radiological characteristics of the tumor. (A) Extraoral clinical appearance showing asymmetry of the middle third of the face. (B) Orthopantomography showing mixed unilocular lesion in the left maxilla (black arrows). Computed tomography of the lesion in the axial (C) and coronal (D) planes showing the extension of the neoplasm (yellow circles).

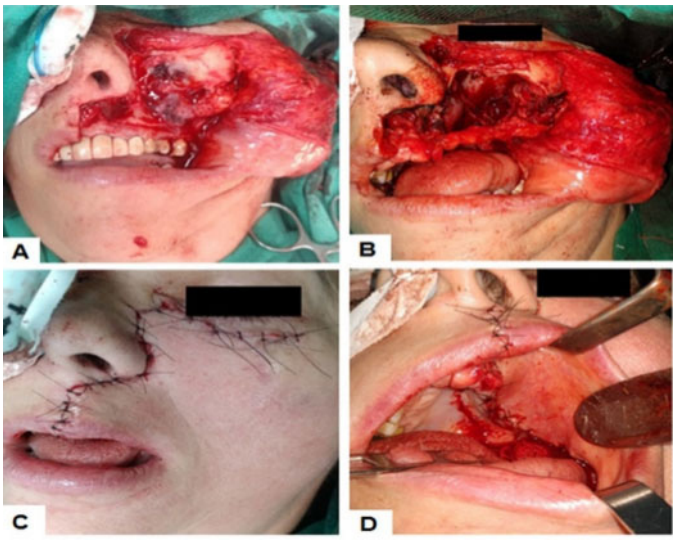

Figure 2: Surgical treatment of the tumor. (A) Surgical exposure of the tumor using the Weber Ferguson's access with Diefenbach's modification. (B) Surgical field and fat pad flap. (C) Extraoral and (D) intraoral sutures in planes.

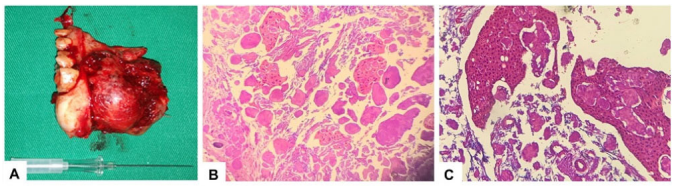

Figure 3: Tumor size and histopathological characteristics. (A) Surgical part of the tumor after resection measuring 50×50×30 mm. (B) Histopathologic slides showing (C) multiple concentric Liesegang ring calcifications and focal epithelial cells surrounding amorphous eosinophilic material along with focal calcifications.

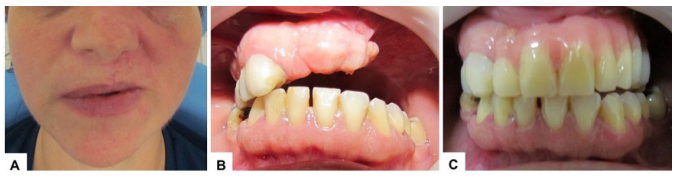

Figure 4: Clinical postoperative follow-up. (A) Extraoral and (B) intraoral appearance after five months, showing satisfactory healing. (C) Temporary dental prosthesis for oral rehabilitation.

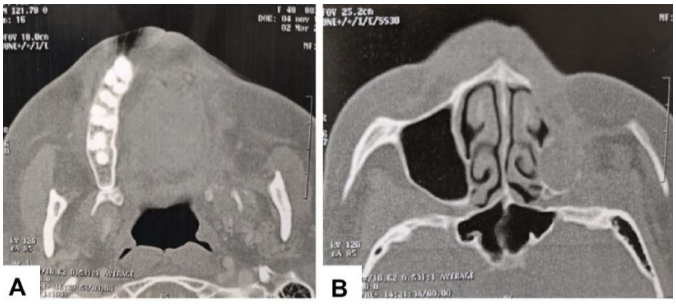

Figure 5: Imaging postoperative follow-up. (A and B) 10-month helical tomography in the axial plane showing no tumor recurrence.

DISCUSSION

Calcifying epithelial odontogenic tumor is a rare benign neoplasm, first reported in 1946 by Kurt H. Thoma and Henry M. Goldman [4] and implemented as a separate entity by Jens J. Pindborg in 1958 [2]. Two more recent reviews on CEOT showed less than 400 cases in the literature, showing the rarity of this tumor [2, 4].

The peculiarity of the present case is based on the intraosseous development of the lesion in the posterior region of the left maxilla, with involvement of the maxillary sinus, palatine process, left lateral wall of the nasal cavity and orbital floor. Other authors reported cases of CEOT in the posterior region of the left maxilla with an extension similar to the present case [7–9]. However, the incidence of CEOT in a maxilla with sinus extension is rare. The presence of facial edema, pain, nasal obstruction, respiratory difficulty, headache, epistaxis, and proptosis are common signs and symptoms of tumors with this extension [6]. From a comprehensive review of the literature in the PubMed/MEDLINE, Science Direct, and Scielo databases, it was possible to identify 29 cases

of CEOT with extension to the maxillary sinus, as shown in Table 1.

A large age range for the occurrence of CEOT has been observed, ranging from young involvement (4 years) to older ages (92 years) [2, 10]. However, Chrcanovic and Gomez [4], based on an analysis of 339 cases of CEOT reported in the literature, observed a higher incidence of neoplasia between the third and fourth decades of life, with a mean age of 38.1 years. In terms of gender prevalence, a similar distribution between the genders has been observed, with a small predilection for females (1.3:1) [2, 4]. In cases of involvement of the maxillary sinus, an age range between 14 and 69 years was observed, with an average of 37.7 years and distribution between women and men in a proportion of 1:1. In the present case report, an unusual incidence of maxillary intraosseous CEOT with sinus extension was presented in a 49-year-old female patient.

The histopathological findings of CEOT are based on sheets, cords, and nests of polyhedral neoplastic epithelial cells with well-defined borders, distinct intercellular bridges, usually with pleomorphism, and rare typical mitosis. Due to morphological aspects, these neoplastic cells present with abundant and eosinophilic cytoplasm, with a prominent and hyperchromatic nucleus. The presence of amyloid protein, which tends to coalesce and calcify, giving rise to the so-called calcified concentric rings of Liesegang is a common finding [3, 5]. Three variations of CEOT architectural patterns have been reported in the literature: clear cell variant, microcystic variant, and CEOT with Langerhans cells [2]. The clear cell variant has been associated with greater aggressiveness of the

Table 1: List of articles in the literature with reports of CEOT of maxillary sinus involvement

Author	Geographic location	Age/gender	Anatomical location	Dental association
Gon (1965) [35]	Africa	35/F	Right jaw	Yes
Stinson et al. (1968) [21]	North America	35/M	Left jaw	Yes
Baunsgaard et al. (1983) [25]	Europe	51/M	Left jaw	Yes
Lee et al. (1992) [26]	North America	27/F	Left jaw	Yes
Bouckaert et al. (2000) [18]	Africa	54/M	Right jaw	No
Patinõ et al. (2005) [11]	Europe	24/F	Right jaw	–
Shekarkhar et al. (2005) [36]	Asia	62/F	Right jaw	No
Gupta et al. (2006) [20]	Asia	26/M	Recurrence in left jaw	No
Bridle et al. (2006) [22]	Europe	30/F	Right jaw	Yes
Gopalakrishnan et al. (2006) [27]	North America	15/M	Left jaw	Yes
Mohtasham et al. (2008) [19]	Asia	18/M	Right jaw	No
Nakano et al. (2008) [28]	Asia	33/F	Left jaw	Yes
Bousdras et al. (2009) [29]	Europe	64/F	Left jaw	Yes
Maria et al. (2010) [6]	Asia	25/M	Left jaw	Yes
Friedrich et al. (2011) [30]	Europe	49/M	Bilateral maxilla	No
da Rosa et al. (2011) [23]	South America	33/F	Left jaw	No
Kamath et al. (2012) [8]	Asia	35/M	Recurrence in left jaw	Yes
Sahni et al. (2012) [13]	Asia	52/M	Right jaw	No
Müller et al. (2012) [31]	Europe	36/M	Right jaw	No
Somayaji et al. (2013) [12]	Asia	34/F	Recurrence in right jaw	No
Carrero et al. (2014) [16]	Europe	69/M	Left jaw	Yes
Foroughi et al. (2015) [9]	Asia	34/F	Recurrence in left jaw	No
Rani et al. (2016) [14]	Asia	48/F	Right jaw	No
Mitra et al. (2016) [37]	Asia	47/M	Right jaw	No
Priya et al. (2016) [32]	Asia	28/F	Left jaw	Yes
Munteanu et al. (2016) [5]	Europe	45/F	Left jaw	No
Gruber et al. (2019) [33]	South America	26/M	Right jaw	Yes
Singh et al. (2020) [34]	India	14/F	Left jaw	Yes
de Arruda et al. (2020) [7]	South America	45/F	Recurrence in left jaw	Yes
Present case	South America	49/F	Left jaw	No

F: female, M: male.

neoplasm, although a recent systematic review does not support this relationship [4]. Cases of tumor malignancy have been reported, being extremely rare, with less than 10 cases reported [2]. In the presented clinical case, the histological findings confirmed the conventional characteristics of the tumor.

The radiographic manifestations of the tumor differ in their evolution [3], with irregular or delimited presentation, unilocular or multilocular radiolucent appearance, with the multilocular aspect being less frequent (30%) [2, 4]. Radiopacities are detectable in the vast majority of cases, although radiolucent areas free from calcification can also be observed [2]. Authors have characterized the radiographic findings as images similar to "soap bubble," "honeycomb," "snowstorm," and "driven snow" [7, 11, 12]. The association of the lesion with dental elements is present in half of the cases [4]. In the present report, a non-delimited unilocular appearance with mixed aspects of calcification, without dental association, was observed, being similar to the characteristics observed by other authors in their respective reports [13, 14].

The radiographic findings of the neoplasm can be confused with other lesions, such as dentigerous cyst, ameloblastoma, adenomatoid odontogenic tumor, glandular odontogenic cysts, ameloblastic fibroma, keratocystic odontogenic tumor, calcifying odontogenic cysts, and odontogenic myxomas [2, 3, 15]. The combination of clinical, radiographic, and histopathological analyzes is essential for the correct diagnosis and treatment development [1].

The treatment of CEOT varies from conservative approaches of enucleation and curettage to more radical treatments of mandibulectomy and maxillectomy, being individualized for each lesion [2, 4, 10]. Treatment is based on the size and location of the tumor, histological findings, the patient's health conditions, the surgeon's skill and experience levels, and post-surgical rehabilitation methods [4]. Some findings such as the presence of cortical perforation and bone erosion, invasion of the maxillary sinus, tooth displacement, and root resorption also help in the choice of therapy. These manifestations show signs of aggressiveness, requiring a more invasive approach [2, 4].

Enucleation with healthy tissue margins is the most indicated in cases of mandibular CEOT [10]. In intraosseous and extraosseous cases, the treatment of enucleation and excision or curettage is the most used, respectively [4]. Carrero et al. [16] recommend performing enucleation followed by curettage in cases of small lesions in the mandible and indicate the most invasive approach of resection with a minimum safety margin of 1 cm of healthy bone in cases of lesions larger than 4 cm, recurrences and in maxillary involvement. For Franklin and Pindborg [17], the use of radical treatments, such as wide resections and hemisections, is unjustifiable in the most cases. However, in CEOT located in the posterior region of the maxilla, the need for more radical therapeutic approaches has been indicated, as they behave in an infiltrative and expansive manner, and may damage nearby vital structures [16]. In a report by Bouckaert et al. [18], is presented a case of CEOT extending to the left maxillary sinus, bilateral orbital cavity, ethmoidal sinuses, frontal bone and brain, with associated proptosis and ocular dystopia, facial edema, and body imbalance, which demonstrates the infiltrative capacity of this neoplasm. The treatment of choice and the characteristics of the 29 cases of maxillary CEOT with sinus involvement described in the literature, together with the present case, are shown in Table 2.

Table 2: Clinical-pathological and therapeutic characteristics of CEOT cases with sinus involvement reported in the literature

Author	Tumor size	Extension beyond the maxillary sinus	Histological findings	Treatment	Follow-up (years)	Recurrence
Gon (1965) [35]	NR	–	Conventional	Curettage	NE	NE
Stinson et al. (1968) [21]	45×34 mm	Left nasal cavity and ethmoidal and sphenoid sinuses	NR	Partial maxillectomy from Weber Ferguson's access and maxillofacial prosthesis	7 years	No
Baunsgaard et al. (1983) [25]	1–2 cm fragments	Left nasal cavity	Conventional	Enucleation from Caldwell-Luc's access	2.5 years	No
Lee et al. (1992) [26]	25×35×4 mm	Compression of the lateral wall of the nasal cavity	Conventional	Partial maxillectomy and maxillofacial prosthesis	7 years	No
Bouckaert et al. (2000) [18]	NR	Bilateral orbital cavity, ethmoid sinuses, frontal bone and brain	Conventional	Not performed due to patient refusal. Tumor debulk planning via Weber Ferguson's access with temporal extension	NE	NE

Table 2: *(Continued)*

Author	Tumor size	Extension beyond the maxillary sinus	Histological findings	Treatment	Follow-up (years)	Recurrence
Patinõ et al. (2005) [11]	35×25 mm	Zygoma and hard palate	Conventional	Partial maxillectomy with iliac crest graft	4 years	No
Shekarkhar et al. (2005) [36]	NR	Lateral wall of nasal cavity and hard palate	Conventional	Partial maxillectomy	NR	NR
Gupta et al. (2006) [20]	NR	Orbital and oral cavity and infratemporal fossa	Conventional	Initial: surgical excision Posterior: partial maxillectomy	NR	Initial after 1.5 years. Posterior not reported
Bridle et al. (2006) [22]	NR	Lateral nasal wall, orbital floor and infratemporal fossa	Conventional	Partial Maxillectomy with Le Fort l surgical approach	1.3 years	NR
Gopalakrishnan et al. (2006) [27]	43×38×39 mm	Nasal cavity and inferior meatus compression	Cystic variant	Enucleation from Caldwell-Luc's access	1 year	No
Mohtasham et al. (2008) [19]	35×20×05 mm	–	Conventional	Not performed due to patient leakage. Partial maxillectomy planning	NE	NE
Nakano et al. (2008) [28]	NR	Hard palate	Conventional	Surgical excision with safety margin	6 years	No
Bousdras et al. (2009) [29]	NR	Nasal cavity and hard palate	Conventional	Surgical excision and upper dental prosthesis	3 years	No
Maria et al. (2010) [6]	42×16×28 mm	–	Conventional	Surgical excision with safety margin	0.5 years	No
Friedrich et al. (2011) [30]	40×40×40 mm	Nasal cavity floor, hard palate and soft tissue	Conventional	Surgical excision with safety margin and prosthetic obturator	5 years	No
da Rosa et al. (2011) [23]	NR	Lateral wall of the nasal cavity, masseter, orbicularis oris and medial pterygoid muscles	Conventional	Partial maxillectomy from Weber Ferguson's access	2 years	No
Kamath et al. (2012) [8]	NR	Lateral wall of nasal cavity, orbital floor and pterygoid plate	Conventional	Initial: enucleation Posterior: surgical excision with safety margins	6 years	Initial after 3 years. Posterior no recurrence.
Sahni et al. (2012) [13]	30×20×15 mm	Lateral wall of nasal cavity	Conventional and clear cell variant	Partial maxillectomy with Le Fort l surgical approach	3 years	No
Müller et al. (2012) [31]	15 cm^3	Lateral wall of nasal cavity and orbit floor	Conventional	Transoral enucleation and curettage	4 years	No
Somayaji et al. (2013) [12]	25×25×50 mm	Nasal cavity, ethmoid sinuses, hard palate and orbital floor	Conventional	Initial: curettage Posterior: partial maxillectomy and palatal prosthesis	9 years	Initial after 1 year. Posterior no recurrence.

Table 2: *(Continued)*

Author	Tumor size	Extension beyond the maxillary sinus	Histological findings	Treatment	Follow-up (years)	Recurrence
Carrero et al. (2014) [16]	40×30 mm	Zygoma	Conventional	Surgical excision and temporal muscle flap reconstruction	8 years	No
Foroughi et al. (2015) [9]	Uncertain	Orbit floor, ethmoid sinuses and nasal cavity	Conventional	Initial: surgical excision Posterior: partial maxillectomy and ethmoidectomy from Weber Ferguson's access and prosthetic obturator	8 years	Initial after 8 years. Posterior no recurrence.
Rani et al. (2016) [14]	50×40 mm	Oral communication	Conventional	Enucleation	1 year	No
Mitra et al. (2016) [37]	45×32 mm	Nasal cavity and nasopharynx	Nasal polyp variant	Polypectomy	NR	NR
Priya et al. (2016) [32]	36×33×32 mm	–	Pigmented variant	Enucleation	1.5 years	No
Munteanu et al. (2016) [5]	4 cm in diameter	Hard palate and vestibular mucosa	Conventional and few clear cells	Partial maxillectomy from Weber Ferguson's access with Diefenbach's modification	1 year	No
Gruber et al. (2019) [33]	50×30 mm	–	Conventional	Enucleation	5 years	No
Singh et al. (2020) [34]	NR	Lateral wall of nasal cavity	Conventional	Enucleation from Caldwell-Luc's access	NR	No
de Arruda et al. (2020) [7]	Uncertain	Pterygoid process, sphenoid, orbital cavity and nasal cavity	Clear cell variant	Initial: enucleation Posterior: partial maxillectomy. Second recurrence: orbital exenteration not performed due to patient refusal	NE	First recurrence after 5 years and second recurrence after 4 years
Present case	44×24×32 mm	Lateral wall of the nasal cavity, hard palate and orbital floor	Conventional	Partial Maxillectomy from Weber Ferguson's access with Diefenbach's modification	Under monitoring	No

NE: Not evaluated, NR: Not reported

The procedures of partial maxillectomy and surgical resection with a safety margin were the initial approach in 34.4% and 24.1% of the cases, respectively. The refusal of treatment and the abandonment of therapy by the patient before its completion have also been described [7, 18, 19]. This can be associated with the invasiveness of the indicated treatments. Treatments from surgical excision, enucleation, and curettage were the initial approach of five recurrent cases [7–9, 12, 20]. These recurrences were subsequently treated using more invasive approaches of partial maxillectomy and surgical excision with a safety margin.

Use of transfacial accesses has been characterized as a common approach in cases of large partial maxillectomies [5, 9, 13, 18, 21–23]. These approaches provide a better view of the surgical field and facilitate the approach to lesions in the middle third of the face [24]. The Weber Ferguson's access was used by four authors, as in the case presented, and was in the treatment plan of one of the cases of abandonment by the patient [5, 9, 18, 21, 23]. Weber Ferguson's access promotes good exposure of the anterior and lateral walls of the maxilla, nasal bone, piriform cavity, zygomatic bone, and masseter muscle [24]. Differently, two authors used the Le Fort 1

surgical approach [13, 22]. This method is well described for obtaining access to the nasopharynx, skull base, and upper anterior cervical spine, in addition to the maxillary, ethmoidal and sphenoid sinuses, nasal and pterygopalatine fossa, and medial portion of the infratemporal fossa, with the main advantage of obtaining a good visual field, presence of low morbidity and absence of facial scars [22, 24]. Selection of the appropriate technique depends on the anatomical location of the tumor, which justifies the use of different approaches.

The CEOT recurrence rate has been reported between 15% and 30%, occurring, in most cases, due to inadequate management [5]. In the literature review performed, lesion recurrence was not observed in 18 cases [5, 6, 11, 13, 14, 16, 21, 23, 25–34], but reported in five cases [7–9, 12, 20], with a time variation between the initial approach and the recurrence of 1–8 years. Recurrence information for four cases [22, 35–37] has not been reported or evaluated by the authors. Chrcanovic and Gomez [4], in a recent review of the literature, observed an overall recurrence rate of 12.6%, being higher in cases of tumor malignancy (42.9%).

Excision and curettage were associated with a higher recurrence rate [4]. All CEOTs with sinus involvement found in this case report, which were treated from partial maxillectomies, did not present any report of recurrence, although in some cases the follow-up was carried out in reduced or non-existent periods. The follow-up of the patient for five to ten years is indicated by most authors [11, 29]. In the cases observed in this case report, follow-up was carried out between six months to nine years, not being reported or evaluated by eight studies [7, 18–20, 34–37]. An approximate period of 50 months is the average between the time of the initial occurrence and its recurrence. However, a range between 2 and 360 months was observed [4]. This demonstrates that the absence of tumor signs for five to ten years is not evidence of non-recurrence. Thus, in the present case report, the use of an extensive invasive surgical resection approach is justified by the best therapeutic results described in the literature from this approach. The patient has 20 months of postoperative follow-up, with no sign of tumor recurrence. The patient's follow-up will be carried out along with the reconstructive and rehabilitative procedures.

CONCLUSION

This case report describes a rare incidence of CEOT with extension to the maxillary sinus, being the 13th case with these characteristics reported in the literature and the fourth in the South American continent, according to the review carried out. The treatment of the present case from maxillectomy with a safety margin, associated with a transfacial approach, promoted a satisfactory prognosis, with no sign of tumor recurrence after 20 months. The long-term follow-up of the patient will be essential for the early detection of possible recurrences.

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