ORIGINAL ARTICLE

Calcifying epithelial odontogenic tumor: a case series from South America

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Abstract:

Objective: To describe a case series of calcifying epithelial odontogenic tumor (CEOT) from South America. **Methods:** This study analyzed cases from Brazil (n=7), Venezuela (n=4), and Colombia (n=1), providing their clinicopathologic, radiographic, and treatment characteristics. **Results:** The series comprised seven (58.3%) females and five (41.7%) males, with a mean age at diagnosis of 33.4 years (range: 19–61). Clinically, 66.7% of patients presented with swelling, and the duration of the lesions ranged from 24 to 120 months. The mandible was involved in 91.7% (n=11) of cases, predominantly in the posterior region (83.4%). Radiographically, 83.4% of lesions were expansive, 75% displayed mixed density, 58.4% were associated with an impacted tooth, and 50% showed root resorption. The median lesion size was 45 mm. Histopathologically, 10 (83.3%) cases were classified as "classic" CEOT, while two (16.7%) were identified as the clear-cell variant. Curettage was performed in 50% of cases and surgical removal (en bloc) in 33.3%. No recurrences were observed in nine (81.8%) cases over a median follow-up of 36 months. **Conclusion:** These findings align with existing literature, highlighting a predominance of young adult females who often present with asymptomatic swelling. The importance of vigilant long-term follow-up post-treatment is emphasized.

Keywords: Jaws; Odontogenic tumors; Oral diagnosis; Oral medicine; Oral pathology.

INTRODUCTION

Calcifying epithelial odontogenic tumor (CEOT), also known as the Pindborg tumor, was first described by Pindborg in 1958[1]. However, a bibliometric review by Ide et al.[2] revealed that Heinz had initially documented the tumor in a German dental journal in 1932. CEOT is a rare benign epithelial odontogenic neoplasm[3],

Statement of Clinical Significance
This study analyzed 12 South American cases of calcifying epithelial odontogenic tumors, highlighting its mandibular predominance, frequent occurrence in young adult females, and the importance of vigilant long-term follow-up to prevent recurrences.

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accounting for approximately 0.03% of oral and maxillofacial lesions and 1.7% of all odontogenic tumors[4]. Given its rarity and histopathological overlap with other odontogenic tumors, the differential diagnosis of CEOT remains an important consideration[3].

Typically, CEOT presents as an asymptomatic lesion with slow, expansive growth, primarily affecting the posterior mandible[4–6]. The extraosseous (peripheral) variant, which is exceedingly rare, manifests as a nodular lesion confined to the soft tissue, most commonly in the gingival mucosa[4–7]. Despite its benign nature, CEOT can exhibit locally aggressive behavior[8], with approximately nine cases of malignant transformation reported in the literature[4,5].

Recent epidemiological data indicate that most published CEOT cases originate from Asia[4]. Apart from Brazil, which has reported a few case series[4,9] and isolated case reports[4,5], literature on this odontogenic tumor from South America is scarce. The aim of the present study was to describe the clinical, radiographic, and histopathological features, as well as the management, of a series of CEOT cases from Brazil, Venezuela, and Colombia as part of a collaborative South American effort.

MATERIAL AND METHODS

The series consisted of a convenience sample of 12 CEOT cases collected from the archives of three oral pathology and medicine services: Universidade Federal de Santa Catarina in Florianópolis, Brazil (n=7), Universidad Central de Venezuela in Caracas, Venezuela (n=4), and Universidad Nacional de Colombia in Bogotá, Colombia (n=1). The study was approved by the local research Ethics Committees (No. 18-23/57829 and No. 42095715.1.0000.0121), and Material Transfer Agreements were established to formalize the collaborative framework. Participants provided written informed consent in accordance with the Declaration of Helsinki.

Clinicodemographic information was obtained from patient records, including sex, age, lesion duration, anatomical location, symptoms (e.g., pain and swelling), radiographic characteristics (e.g., radiolucent, mixed, or radiopaque; unilocular or multilocular), changes in cortical bone (e.g., expansion or perforation), association with teeth (e.g., impaction, tooth displacement, or root resorption), lesion size, treatment methods, and recurrence data.

A 4-μm-thick section was cut from each paraffin block and stained with hematoxylin and eosin. Under polarized light, the amyloid-like material exhibited apple-green birefringence with Congo red staining. Diagnoses were made by experienced oral pathologists at each institution, based on the criteria outlined in the 5th edition of the World Health Organization (WHO) Classification of Odontogenic and Maxillofacial Bone Tumors[10]. Although a formal calibration process was not performed, a standardized diagnostic approach was adopted, and all cases were reviewed collaboratively. Microscopically, the diagnostic criteria included the identification of sheets or islands of polyhedral epithelial cells with well-defined borders and intercellular bridges, nuclear pleomorphism without significant mitotic activity, and the presence of extracellular eosinophilic amyloid-like material, often associated with concentric calcifications (Liesegang rings)[10]. Cases involving synchronous CEOT or hybrid odontogenic lesions were excluded from the study.

Data were organized using Microsoft Office Excel 2019 (Microsoft®, Redmond, WA, USA) and analyzed descriptively using GraphPad Prism version 8.0.0 for Windows (GraphPad Software, San Diego, CA, USA).

RESULTS

This series comprised seven (58.3%) females and five (41.7%) males, with a mean age of 33.4 (±11.1) years (range: 19–61). The most affected age group was the third decade of life (41.7%). The mandible was involved in 91.7% (n=11) of cases, predominantly in the posterior region (n=9; 75%) (Table 1).

Clinically, swelling was observed in eight (66.7%) patients, four (33.3%) patients were asymptomatic, and one (8.3%) patient reported pain. The duration of the lesions was documented in five cases, ranging from 24 to 120 months, with a median of 100 months.

Radiographically, cortical bone expansion was observed in 10 cases, while cortical perforation was noted in two cases. The internal characteristics of the lesions were mixed (n=9; 75%) or radiolucent (n=3; 25%). An impacted tooth was detected in seven cases, root resorption in six, and tooth mobility in one. Lesion sizes ranged from 10 to 70 mm, with a median size of 45 mm. Figures 1 and 2 illustrate the clinical and radiographic features of the CEOT cases.

Histopathologically, CEOT was characterized by soft tissue with varying amounts of calcifications. The epithelium component displayed variable architectural patterns, with cells typically being polyhedral and possessing abundant eosinophilic and well-defined cytoplasm. Intracellular bridges were observed between polyhedral cells. The nuclei were characteristically pleomorphic, with frequent giant and hyperchromatic nuclei. Calcifications, often arranged in concentric rings (Liesegang rings), were also observed (Figure 3).

Table 1. Clinicoradiographic features, histopathologic types, and treatment modalities of calcifying epithelial odontogenic tumors

Case	Sex	Age (years)	Evolution time (months)	Anatomical location	Symptoms	Radiographic aspects	Size (mm)	Histopathologic type	Treatment	Recurrence/ follow-up (months)
#1	F	37	24	Right mandible (second premolar region)	Asymptomatic	Osteolytic lesion with internal radiopaque foci, well-defined margins, and associated with tooth mobility	15	Classic	Surgical removal (*en bloc*)	NI
#2	F	30	120	Mandible (molar region)	Asymptomatic	Mixed lesion with expansion, associated with an impacted tooth but without tooth resorption	50	Classic	Curettage	No (36 months)
#3	F	31	100	Mandible (molar region)	Asymptomatic	Mixed lesion with expansion, associated with an impacted tooth but without tooth resorption	55	Classic	Curettage	No (38 months)
#4	F	29	NI	Mandible (molar region)	Swelling	Mixed lesion with expansion, associated with an impacted tooth and tooth resorption	28	Classic	Surgical removal (*en bloc*)	No (24 months)
#5	F	28	NI	Mandible (molar region)	Swelling	Mixed lesion with expansion, associated with an impacted tooth and tooth resorption	25	Classic	Curettage	No (120 months)
#6	F	19	NI	Mandible (molar region)	Asymptomatic	Mixed lesion with expansion, associated with an impacted tooth but without tooth resorption	17.5	Classic	Curettage	No (36 months)
#7	M	36	NI	Mandible (canine region)	Swelling	Mixed lesion with expansion and cortical bone perforation but without tooth resorption	10	Clear cells	Curettage	No (6 months)
#8	M	23	NI	Mandible (molar region)	Swelling	Mixed lesion with expansion, associated with an impacted tooth but without tooth resorption	65	Classic	Curettage	No (24 months)
#9	F	61	24	Left maxilla	Swelling and painful	Radiolucent lesion, expansion, and root resorption	40	Clear cells	Hemimaxillectomy	Yes
#10	M	28	NI	Right posterior mandible	Swelling	Multilocular radiolucency with expansion and root resorption	50	Classic	Hemimandibulectomy	No
#11	M	50	120	Posterior mandible	Swelling	Multilocular radiolucency with radiopaque foci, cortical bone perforation, and root resorption	70	Classic	Hemimandibulectomy	Yes (3 times)
#12	M	29	NI	Posterior mandible	Swelling	Multilocular radiolucency with expansion, root resorption, and an impacted tooth	50	Classic	Hemimandibulectomy	No

Note: F, female; M, male; NI, not informed.

Stromal amyloid protein was present as small, rounded to irregular, homogenous masses of lightly eosinophilic hyaline material that exhibited apple-green birefringence under polarized light with Congo red staining. Ten (83.3%) cases exhibited the "classic" pattern, while two (16.7%) exhibited the clear-cell variant, intermixed with other CEOT cells displaying clear cytoplasm.

Treatment included curettage in six (50%) cases, hemimandibulectomy/hemimaxillectomy in

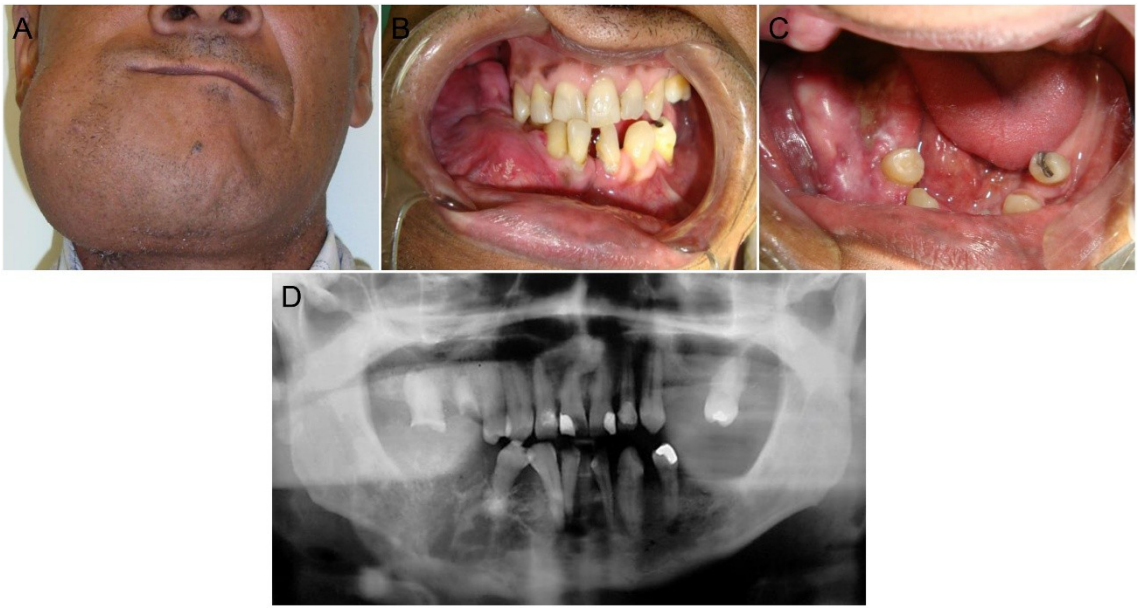

Figure 1. Clinical and radiographic features of a calcifying epithelial odontogenic tumor. **(A)** Swelling in the right cheek and mandible, measuring approximately 7.0 cm at its largest diameter. **(B, C)** Intraoral view exhibiting a firm tumor with ulcerated areas affecting the alveolar ridge and vestibular sulcus. Note the rotation of the right second premolar. **(D)** Panoramic radiograph revealing a multilocular radiolucency with radiopaque areas associated with the right second premolar.

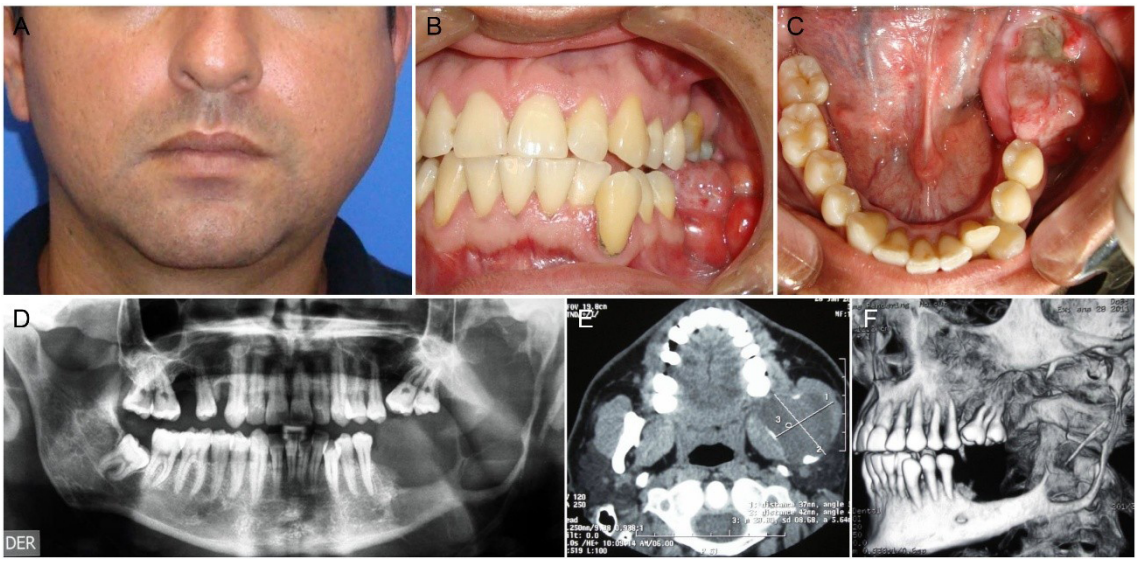

Figure 2. Clinical and radiographic features of a calcifying epithelial odontogenic tumor. **(A)** Swelling in the left cheek and mandible, measuring 3.0 cm at its largest diameter. **(B, C)** Intraoral view showing a lobulated, ill-defined tumor with ulcerated areas in the alveolar region of the molars, extending to the vestibular sulcus. **(D)** Panoramic radiograph revealing a multilocular radiolucent lesion involving the left mandibular ramus, coronoid process, and extending close to the condyle. **(E)** Axial view of computed tomography reveling an expansive and destructive hypodense lesion measuring 3.0 cm at its largest diameter. **(F)** Three-dimensional reconstruction revealing an ill-defined, scalloped border with a hypodense "soap bubble" appearance.

four (33.3%), and surgical removal (*en bloc*) in two (16.7%). Recurrence occurred in two cases, one of which recurred three times. Outcome data were unavailable for one case. Follow-up information was available for seven cases, with durations ranging from six to 120 months and a median follow-up time of 36 months.

DISCUSSION

Data from the present CEOT series revealed a higher prevalence among women in their third decade of life, partially aligning with a large multicenter Brazilian study that reported a higher occurrence in women during their fourth decade of life[4]. Conversely,

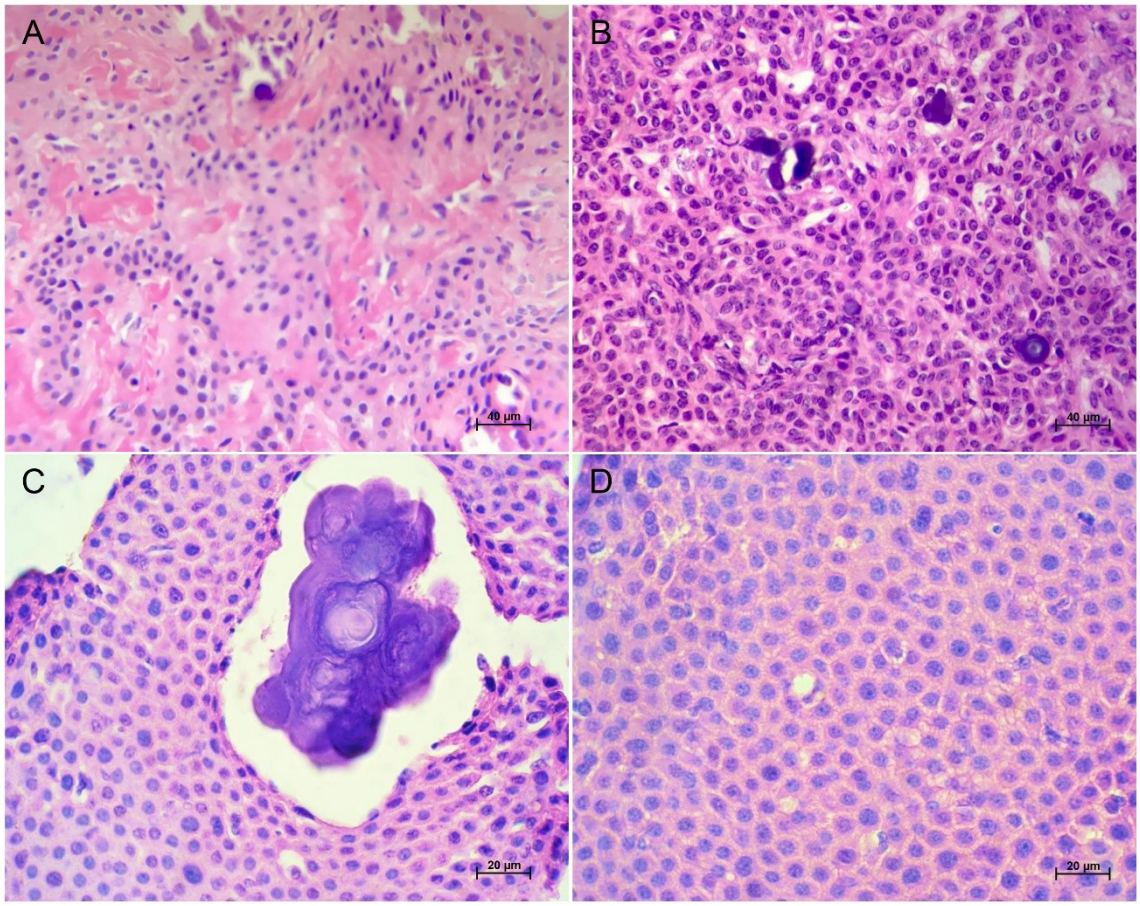

Figure 3. Histopathological features of a calcifying epithelial odontogenic tumor. **(A)** Amyloid material interspersed with nests of epithelial cells. **(B, C)** Calcified material interspersed with polyhedral eosinophilic cells, displaying eosinophilic cytoplasm, prominent intercellular bridges, and **(D)** hyperchromatic nuclei (hematoxylin and eosin staining: 200× and 400× original magnifications).

an American study reported a mean age of 40 years with an equal sex distribution[6]. A systematic review, however, identified the third and fourth decades as the most frequently affected age groups[5]. Mandibular involvement in our study was 91.7%, markedly higher than previously documented rates, which range from 58.7%[5] to 65.6%[4]. This pronounced predilection for the mandible is also observed in other odontogenic tumors, such as ameloblastoma, which shows a mandibular involvement rate of 94.2% among South African individuals[11].

The pathogenesis of CEOT likely involves a combination of odontogenic epithelial origin, amyloid deposition, calcification, and dysregulation of cellular signaling pathways. Next-generation sequencing has identified mutations in *PTEN*, *MET*, *JAK3*, and *CDKN2A* in CEOT, while single-gene studies have reported mutations in *PTCH1* and ameloblastin[12,13]. Nonetheless, these molecular findings have not yet been shown to impact clinical outcomes or influence treatment decisions.

CEOT can present in two clinical variants: intraosseous (central) and extraosseous (peripheral)[4]. The intraosseous variant, which accounted for all cases in our series, is characterized by a slow-growing, expansive mass within the jawbones, often leading to cortical bone expansion, tooth displacement, and root resorption[4]. Epidemiological data suggest that fewer than 400 cases of CEOT have been published in the literature[4-6]. In contrast, the extraosseous variant is less common and typically less aggressive, with approximately 30 cases reported hitherto[4-6].

Radiographically, most of our cases exhibited mixed radiodensities. Approximately 75% of reported cases in the literature present as mixed-density lesions due to tumor calcifications[5], often clustered around a tooth, producing the characteristic "driven snow" appearance[14]. Imaging may also reveal fine, sparse trabeculation, cortical expansion of the affected jaw, and inferior displacement of the inferior alveolar nerve canal when the posterior mandible is involved[14]. Another series reported an equal

distribution of radiolucent and mixed-density lesions, each accounting for 48% of cases[4]. These lesions are predominantly unilocular or multilocular, with well-defined or diffuse margins. In approximately half of the cases, an unerupted tooth — most commonly a mandibular third molar — is associated with the lesion[5]. Such features are essential in differentiating CEOT from other odontogenic tumors, such as ameloblastoma, which usually presents as a multilocular radiolucency without calcifications, and from other lesions like adenomatoid odontogenic tumor, ameloblastic fibroma, and dentigerous cyst[15]. While adenomatoid odontogenic tumor primarily affects the anterior maxilla and ameloblastic fibroma occurs in younger individuals, CEOT can be difficult to distinguish from early pericoronal presentations of a dentigerous cyst[15]. Calcifying odontogenic cyst should also be considered due to its radiographic similarity to CEOT, particularly in posterior mandibular cases[16].

Histopathologically, CEOT demonstrates a variety of architectural patterns, often accompanied by cellular pleomorphism and mitotic activity[9]. A classification system based on histomorphologic patterns has been proposed to account for this variability in presentation[17]. CEOT may exhibit isolated or combined patterns with polyhedral epithelial cells, calcifications, and amyloid deposits serving as its defining features[18,19]. Numerous histologic variants have been described, including those with clear cells[8], Langerhans cells[20], clear Langerhans cells[21], non-calcifying Langerhans cell-rich patterns[22], cystic/microcystic patterns[23,24], cementum/bone-like material[25], and myoepithelial cells[26]. Two cases of the clear-cell variant were documented here, corroborating findings from an earlier Brazilian multicenter study, which detected this variant in four out of 32 (12.5%) cases[4].

The presence of calcifications in Liesegang rings and amyloid deposits is a key feature that distinguishes CEOT from other odontogenic tumors, such as ameloblastoma, which lacks these characteristics, and odontogenic myxomas, which exhibits a mucoid stroma devoid of calcifications or amyloid[15,27]. Immunohistochemically, CEOT shows amyloid positivity for CD138 and amyloid A, while the calcifications are positive for CK5, CD138, and amyloid A, further reinforcing its distinction from other neoplasms[9]. Accurate differentiation from intraosseous squamous cell carcinoma is also crucial to avoid overtreatment. Additionally, the clear cell variant of CEOT requires careful distinction from clear cell odontogenic carcinoma and metastatic clear cell carcinoma[8]. The amyloid nature of the eosinophilic material in CEOT is confirmed by positive Congo red staining and thioflavine T fluorescence[28]. Odontogenic ameloblast-associated protein has also been identified in CEOT amyloid deposits, with dystrophic calcifications frequently forming large masses within these deposits[18,29]. Moreover, CEOT has been reported as a hybrid lesion that co-occurs with other odontogenic neoplasms, particularly adenomatoid odontogenic tumor, with approximately 26 documented cases[30].

The most common surgical treatments for CEOT include enucleation, excision, or curettage, which are typically used for smaller tumors[5]. In our series, most cases were managed conservatively using curettage or surgical removal (*en bloc*). However, larger or more aggressive tumors required more extensive procedures, such as hemimandibulectomy or hemimaxillectomy. The recurrence rate observed in this study was 16.7%, higher than the 11.6% reported elsewhere[5], potentially due to insufficient surgical margins or the inherently aggressive behavior of certain lesions. As this was a retrospective study based on archival records from multiple institutions, some variability in the completeness and quality of clinicoradiographic data was unavoidable. To mitigate these limitations, only cases with detailed histopathological documentation and representative imaging were included, and standardized diagnostic criteria were applied consistently across centers. Given the risk of malignant transformation, careful long-term follow-up remains essential[4].

CONCLUSION

CEOT is a rare but significant odontogenic tumor that requires careful clinical, radiographic, and histopathological evaluation for accurate diagnosis and treatment planning. Given its potential for local aggressiveness and recurrence, a surgical approach combined with long-term follow-up is essential. This series provides valuable data on the epidemiological, clinical, and pathological features of CEOT in a South American population, highlighting the importance of early diagnosis and individualized management strategies.

AUTHORS' CONTRIBUTIONS

JAAA: conceptualization, methodology, supervision, validation, writing – review & editing. JVLV: data curation, investigation, writing – original draft. MEZB: data curation, investigation, writing – original draft. MVD: investigation, resources, writing – review & editing. CPPV: investigation, resources, writing – review

& editing. JPRM: investigation, resources, writing – review & editing. ARP: investigation, resources, writing – review & editing. ILC: data curation, investigation, writing – original draft. JRDP: data curation, investigation, writing – original draft. GCS: conceptualization, methodology, supervision, writing – review & editing. IBV: conceptualization, methodology, supervision, writing – review & editing. KMF: investigation, resources, writing – review & editing. RLCAJ: investigation, resources, writing – review & editing. RG: investigation, resources, writing – review & editing. JRT: conceptualization, data curation, formal analysis, investigation, methodology, project administration, writing – original draft, writing – review & editing. BABA: conceptualization, data curation, formal analysis, methodology, supervision, validation, visualization, writing – review & editing.

CONFLICT OF INTEREST STATEMENT

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Competing interests: The authors have no conflicts of interest to declare.

Ethics approval: The study was approved by the local research Ethics Committees of the participating institutions (No. 6024262).

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