

Non-calcifying epithelial odontogenic tumor: a rare case report and literature review

Hela Zouaghi¹, garma maroua¹, afef slim¹, abdellatif chokri¹, Manel Njima², and jamil selmi¹

¹University of Monastir Faculty of Dental Medicine of Monastir

²Fattouma Bourguiba University Hospital of Monastir

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Abstract

This work aimed to report a rare case of a non-calcifying Pindborg Tumor involving the right mandible. The absence of calcifications in the calcifying epithelial odontogenic tumor presents a diagnostic challenge and prognostic implications. A literature review of the sixteen reported clinical cases of this scarce variant was performed.

Introduction

Calcifying epithelial odontogenic tumor is a benign lesion that Thoma and Goldman first described, and then in 1958, it was described in detail by Jens Pindborg¹. It's classified by the world health organization among the benign odontogenic epithelial tumors². Since this description, many cases have been reported in the literature. The mean age of occurrence is the fourth decade with no gender predilection and it has a mandibular preference^{1,3}. A scarce non-calcifying variant was described. The absence of calcifications can make a diagnostic challenge.

In this work, a rare clinical case was reported, and a literature review of the different reported cases of the non-calcifying epithelial odontogenic tumor was conducted.

Case report

A 40-year-old female patient was referred to the oral surgery department for the management of a radiolucent lesion associated with the right mandibular impacted wisdom tooth of incidental discovery. Her familial and past medical history was non-contributory. She was asymptomatic.

The extraoral examination was normal. The intraoral examination showed a well-circumscribed mandibular sessile nodule distal to tooth 47, extending on both vestibular and lingual gingiva and measuring approximately 1 cm in diameter. It was covered by normal mucosa and firm on palpation. Tooth 48 was clinically absent (Figure 1). Radiographic examination using panoramic radiograph showed a unilocular expansile radiolucent homogenous image on the right mandibular angle, associated with an impacted wisdom tooth 48. A cone beam computed tomography was required to precisely determine lesion expansion and limits. The lesion measured 2 cm × 1.5 cm × 1 cm and its limits were well-defined. The cortical bone and the mandibular canal were respected. (Figure 2)

Clinical and radiological findings let suspected many diagnoses, such as odontogenic cysts, odontogenic keratocyst, dentigerous cyst, ameloblastoma and osteolytic lesion related to the brown tumor. Parathormone, vitamin D, phosphor and calcium serum levels were requested. There were within the normal limits except for the decreased vitamin D level.

Total enucleation of the lesion was performed under local anesthesia, with the impacted wisdom tooth extraction.

Histopathological examination revealed a nodule formed by trabeculae and clusters of polyhedral epithelial cells, focally united by intercellular bridges. They had an eosinophilic cytoplasm and regular finely nucleated nuclei. These epithelial cells were separated by interspersed hyaline fibrous tissue, little cellular by places, globular, taking the aspect of amyloid-like material (Figure 3). This substance showed brick red color on Congo red staining (Figure 4). The diagnosis of a non-calcifying epithelial odontogenic tumor was consequently confirmed.

The post-operative period was uneventful. The patient continues to be on regular follow-ups. No sign of recurrence was detected after six months follow-up.

Discussion

The calcifying epithelial odontogenic tumor (CEOT) or Pindborg tumor has been well documented in the literature since its first description. It is a rare lesion, the reported cases didn't exceed 400¹, and it represents less than 1% of all odontogenic tumors³. The non-calcifying variant is very rare; it is the least reported one¹. The radiological and histological features of the non-calcifying epithelial odontogenic tumor are distinguished from those of the usual calcified variant. Consequently, there was a diagnostic challenge in the reported case and the diagnosis of a Pindborg tumor was not suspected.

On radiographic examination, Pindborg tumor appears most commonly as a mixed radiopaque/ radiolucent image. Both multilocular and unilocular presentations were described. The association with an impacted tooth was frequently found. The present case was exempt from radiopacities. This radiolucent aspect is insufficient to diagnose a non calcifying variant since it was described in early diagnosed immature lesions, in which calcifications were found on histopathological examination. This radiolucent aspect was also described in some called cystic variant⁴. This radiolucent aspect may lead to the misdiagnosis of this variant as an odontogenic cyst, like in the reported clinical case.

The most distinctive microscopic feature of classical CEOT is the presence of sheets of polyhedral cells, amyloid globules and Liesegang ring calcifications in the tumor tissue⁵. According to the literature, non-calcifying cases described are primarily associated with the presence of Clear Cells and Langerhans Cells. This led authors to the definition of variants of this tumor: the Clear Cells variant and Langerhans Cells-rich variant¹.

The absence of calcification does not only pose a diagnostic problem, but also has been suggested to be an indicator of poor differentiation of the tumor. Consequently, the non-calcifying epithelial odontogenic tumor would have more risk of recurrence and requires radical treatment and a long-term follow-up.³

The international literature was reviewed; only 16 cases of non-calcifying epithelial odontogenic tumor were found from 1981 to 2021 and are resumed in table 1. No sex predilection was noted (7 females/ 9 males). Age ranged from 20 to 68 years old. The present reported case adds a female patient aged 40 years old. Ten of the cases were developed in the maxillary bone over seven cases in the mandible, including the present reported case. Histologically, 4 of the reported non calcifying variants contained clear cells, 5 contained Langerhans cells, 2 contained both clear and Langerhans cells, and 5 showed neither clear cells nor Langerhans cells like the reported clinical case. Treatment modalities varied between enucleation and partial resection with 1cm margins to prevent recurrence. Enucleation was preferred as a more conservative approach in the present case since the diagnosis of Pindborg tumor was unlikely suspected before histopathology, especially with the absence of calcification and the sporadic similar reported cases. Also, there were no clinical or radiological signs of aggressivity of the lesion.

Considering the prognostic implications that present the absence of calcifications, close follow-up appointments are mandatory to assess any possible recurrence early. In the literature, no cases of recurrence were reported over follow-up periods ranging from 6 months to 10 years.

Conclusion

The absence of calcifications in CEOT was reported in very few cases of Pindborg tumor. A careful diagnostic approach should be conducted to make the definitive diagnosis of this rare variant. Some authors suggested that it is a poor differentiated form with more unpredictable behavior. This possible aggressiveness still represents a controversial subject; however, no recurrence was reported till date. More case reports of non-calcifying epithelial odontogenic tumor are needed to investigate this entity better.

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Table 1: Review of the reported cases of non-calcifying CEOT

<i>Author</i>	<i>Year</i>	<i>Age</i>	<i>Sex</i>	<i>Location</i>	<i>Histopathology</i>	<i>Recurrence</i>
Aufdermaur et al. ⁶	1981	68	M	Mand	No calcification	No (1Year)
Asano et al. ⁷	1990	44	F	Max	No calcification Langerhans cells Clear cells	Non specified
Takata et al. ⁸	1993	58	M	Max (post)	No calcification Langerhans cells	Non specified
Hafian et al. ⁹	2004	61	M	Max (ant)	No calcification	Non specified
Wang et al. ¹⁰	2007	52	F	Max (ant)	No calcification Langerhans cells	Non specified
Wang et al. ¹¹	2007	38	M	Mand	No calcification Clear cells	Non specified
Wang et al. ¹¹	2007	39	F	Max	No calcification Clear cells	Non specified
Kaushal et al. ³	2012	57	M	Mand	No calcification	No (1Year)
Afroz et al. ¹²	2013	20	F	Max (ant)	No calcification Langerhans cells Clear cells	Non specified
Chen et al. ¹³	2014	40	F	Max (ant)	No calcification	No (5 Years)
Chen et al. ¹³	2014	58	M	Max	No calcification Langerhans cells	No (10 Years)

<i>Author</i>	<i>Year</i>	<i>Age</i>	<i>Sex</i>	<i>Location</i>	<i>Histopathology</i>	<i>Recurrence</i>
Tseng et al. ¹⁴	2015	24	M	Max	No calcification Langerhans cells	Non specified
Kurihara et al. ¹⁵	2017	60	M	Mand (ant)	No calcification Clear cells	No (3 Years)
Taneeru et al. ¹⁶	2017	27	F	Mand (post)	No calcification	Non specified
Santosh et al. ¹⁷	2019	43	F	Max (ant)	No calcification Langerhans cells	No (18months)
Patankar et al. ¹⁸	2021	43	M	Mand (post)	No calcification Clear cells	Non specified

Figure legends

Fig. 1: Intraoral aspect showing a lower well circumscribed gingival exophytic mass extending distal to the right second mandibular molar.

Fig. 2: Cone beam computed tomography showing a well-defined hypodense image on the right mandibular angle + impacted third molar

Fig. 3: Histological section stained with hematoxylin-eosin: Sheets and nests of polygonal epithelial cells with eosinophilic cytoplasm and large areas of amyloid-like material. (HEx100)

Fig. 4: Amyloid-like material in tissue exhibits a deep red color with Congo red stain.





