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CALCIFYING EPITHELIAL ODONTOGENIC TUMOR INVOLVING THE MAXILLARY SINUS: REVIEW OF THE LITERATURE

CALCIFICAÇÃO DO TUMOR EPITELIAL ODONTOGÊNICO ENVOLVENDO O SEIO MAXILO: REVISÃO DA LITERATURA

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Abstract
Calcifying epithelial odontogenic tumors (CEOT) account for approximately 1% of odontogenic tumors. The occurrence of this tumor in the maxillary sinus is rare, and there are few scientific reports of an invasion of this tumor to this location. This article aimed to perform an update on the cases of this type of tumor in which there was extension to the maxillary sinus. A total of 317 studies, 13 articles were selected according the inclusion criteria. Ages ranged from 15 to 51 years and both sides were affected. There was no predilection for sex. The treatment of choice was surgical excision. A CEOT is uncommon and involvement with maxillary sinus is extremely rare.

Keywords: Calcifying epithelial odontogenic tumor, pindborg tumor, maxillary sinus.

Resumo
Tumores odontogênicos epiteliais calciantes (TOEC) são responsáveis por aproximadamente 1% dos tumores odontogênicos. A ocorrência desse tumor no seio

maxilo é rara, e há poucos relatos científicos de uma invasão desse tumor a este local. Este artigo teve como objetivo realizar uma atualização sobre os casos desse tipo de tumor em que houve extensão do seio maxilo. Um total de 317 estudos, 13 artigos foram selecionados segundo os critérios de inclusão. As idades variaram de 15 a 51 anos e ambos os lados foram afetados. Não havia predileção por sexo. O tratamento escolhido foi a excisão cirúrgica. Um TEOEC é incomum e o envolvimento com seios maxilos é extremamente raro.

Palavras-chave: Tumor epitelotogênico calcificador, tumor pindborg, seio maxilo.

1. Introduction

Calcifying epithelial odontogenic tumor (CEOT) or Pindborg tumor was described by Danish oral pathologist, Dr. Jens Jorgen Pindborg in 1955, as a distinct pathological entity. It is a benign, locally invasive odontogenic tumor of epithelial origin (intra or extraosseous), accounting for less than 1% of all odontogenic tumors. Intraosseous tumors affect the mandible more than the maxilla at a proportion of 2:1. When a CEOT affects the maxilla, it rarely extends into the maxillary sinus. There is predominance in the majority of cases occurring between the third and fifth decades of life. There is no predominance with respect to sex. Although it is a benign neoplasm, it may exhibit an aggressive pattern. Among the possible treatments, small intraosseous lesions indicate conservative tumor enucleation with curettage and removal of healthy bone adjacent to the lesion. Larger lesions require resection of the tumor beyond the tumor-free margins.[1-13] This study describes an update of CEOT with maxillary sinus extension.

2. Materials and methods

Two of the authors (J.C.P.D. and E.D.R.) performed the selection of articles independently. Searches were performed in the PubMed/MEDLINE and Scopus databases for articles published without time restriction. The key words used in this study were: ‘‘Calcifying epithelial odontogenic tumor or CEOT or Pindborg tumor and Maxillary sinus’’.

Among the inclusion criteria were studies that referred to the topic addressed, belonged to reports of clinical cases and presented CEOT with extension to the maxillary sinus. The following exclusion criteria were applied: investigations in which the tumor did not reach this location, retrospective studies and literature reviews.

The studies were first classified according to the inclusion and exclusion criteria. After performing searches in the selected databases, a careful analysis was performed to identify any cases of disagreement between the authors. Studies were selected based on their titles and abstracts. After the first selection stage, the selected articles were analyzed based on their full content.

3. Results

The searches performed in the databases led to the retrieval of 317 articles in total: 272 from PubMed/MEDLINE and 45 from Scopus. After the removal of 29 duplicate articles, 288 studies were selected for analysis based on their title and abstract, and in accordance with the inclusion and exclusion criteria. Following the full-text review, 13 studies were analyzed and form the basis of this review.

Table 1: Scientific articles presenting CEOT with extension to the maxillary sinus.

Author/year	Sex	Age	Location and extension	Treatment
Mitra et al.[1] 2016	-	47	Right maxillary sinus, nasal cavity and nasopharynx	Surgical excision
Rani et al.[2] 2016	Female	48	Maxilla and right maxillary sinus	Surgical excision
De Castro et al.[3] 2012	Female	29	Maxilla and left maxillary sinus	Surgical excision
da Rosa et al.[4] 2011	Female	33	Maxilla and left maxillary sinus	Hemimaxillectomy
Friedrich and Zustin[5] 2011	Male	49	Maxillary sinus, nasal cavity	Surgical excision
Angadi and Rekha[6] 2011	Male	30	Right Maxilla	Surgical excision
Mohtasham et al.[7] 2008	Male	18	Maxilla and right maxillary sinus	Hemimaxillectomy
Gopalakrishnan et al.[8] 2006	Male	15	Left maxillary sinus	Surgical excision
Bridle et al.[9] 2006	Female	30	Right maxillary sinus	Surgical excision
Lee et al.[10] 1992	Female	27	Maxilla and left maxillary sinus	Hemimaxillectomy
Baunsgaard et al.[11] 1983	Male	51	Maxilla and left maxillary sinus	Surgical excision
Stimson et al.[12] 1968	Male	35	Maxilla and left maxillary sinus	Hemimaxillectomy
Gon[13] 1965	Female	35	Maxilla and right maxillary sinus	-

4. Discussion

CEOT is a rare benign neoplasm with approximately 200 cases reported in the literature, involvement in the maxillary sinus is rare, with only 13 clinical cases described. The age range was between 15 and 51 years; highest prevalence was observed in the group of subjects aged 30 to 50 years. Both sides of the maxilla were affected and there was no predilection for sex.[1-13]

CEOT in the maxillary sinus is generally aggressive and require extensive surgeries because they present rapid growth when compared to mandibular cases, and they have the capacity to invade surrounding vital structures, affecting the morbidity of these patients.[2,4,7,10,12] Among the treatments reported, the surgical excision was the most conservative approach for the surgical removal of the lesion (Table 1).[1-3,5,6,8,9,11]

Long-term regular follow-up is required to observe for potential recurrence.[1,2,13] Mass in the maxillary sinus usually exhibits abnormal eye signs and nasal obstructive symptoms.[1,9]

CEOT although benign, can present aggressive characteristics, invade important structures and have recurrence. Its occurrence in the maxillary sinus is extremely rare, with only 13 cases reported in the literature. Both sexes were affected with patient ages ranging from 15 to 51. The treatment of choice was surgical excision.

5. Conclusion

The CEOT represents a rare epithelial odontogenic tumor that has a marked predilection for the mandible, with few cases described in the maxilla and even more rarely, specifically in the maxillary sinus. The scarcity of calcified deposits combined with the anatomical location in the maxillary sinus emphasizes the particularity of this pathology now presented. Thus, aggressive surgical intervention is suggested as a treatment option for these cases.

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