МЕДИЦИНСКИЕ НАУКИ

AMELOBLASTOMA ASSOCIATED WITH DENTIGEROUS CYST

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ABSTRACT

Ameloblastoma is a benign, locally aggressive tumour, with an unicystic variant that is very difficult to be differentiated from odontogenic cysts, because of their similarity in the clinical manifestation and X-ray examination. The morphological similarities between these processes make for a more difficult histological diagnosis. We present a case of a 32-year old male, admitted in the Maxillofacial surgery clinic in a University hospital for surgical treatment, because of a swelling in the left mandibular vestibule. A cystic formation, histologically diagnosed as an epithelial one, is removed. Eight months later, the patient is admitted once again, with the same symptoms. The biopsy result from the second operation is a plexiform unicystic ameloblastoma. What is being discussed is the connection between the two pathological processes and the difficulties with giving the correct morphological diagnosis.

Key words: unicystic ameloblastoma, plexiform ameloblastoma, dentigerous cysts.

Introduction

Dentigerous cyst is the second most common odontogenic cyst. It represents 20% of the epithelium lined jaw cysts and is usually seen in teenage and adolescent years, although it can occur over a wider age period. By definition, a dentigerous cyst occurs in association with an unerrupted tooth, most commonly around permanent mandibular third molars (wisdom teeth). Dentigerous cysts can grow large enough to produce a painless bone expansion. They present with a non-keratinized squamous lining that is relatively uniform in thickness, unless there is an associated inflammation. The epithelium is seen to overline a fibrovascular connective tissue stroma/wall. The epithelium can show a variety of epithelial differentiation, including columnar or cuboidal changes [8]. Rarely, secondary neoplasms could arise from dentigerous cysts, most notably ameloblastoma. Approximately 50% of ameloblastomas arise from the epithelial lining of a dentigerous cyst [4]. Ameloblastoma is a benign, locally aggressive tumor (25-35% recurrence) of odontogenic epithelium. The mandible is the most common site for most types of ameloblasomas. In approximately 2/3 of the cases it occurs along the posterior side of the mandible. Clinically, it can present as a painless swelling or expansion of the jaw. It is hard to distinguish between unicystic ameloblastoma and odontogenic cysts, both clically and using an X-ray [5, 7]. Histologically, the follicular pattern of ameloblastoma is the most common one. The plexiform subtype is the next most common pattern. The tumour epithelium is seen to form irregular plexiform masses or network of strands. The stroma is usually scanty. Microcyst formation can occur in the stroma. Traditional surgical treatment of ameloblastoma requires segmental resection with wide margins, while dentigerous cysts - only enucleation and curettage.

Case report

A 32-year old male is admitted in the Maxillofacial surgery clinic of UMHAT "St. George" – Plovdiv at the end of February, 2019, with a swollen and painful left region of the lower jaw. The symptoms date back a few months, but for the past 3-4 days there has been a significant increase in the swelling, now with a severe pain. The clinical examination describes facial asymmetry, caused by a restricted, painful swelling around the left part of the mandible. The mouth opens normally, the teeth are unaffected. There is expansion of the mandibular vestibule, with signs of destruction. When punctured, there is a cloudy, brownish matter, pouring out that area. Antibiotic therapy is prescribed and on the next day, a surgical removal, under general anesthesia is performed. A mucoperiosteal flap is formed, with a complete excision of the pathologic tissue and stitching of the flap. Drainage is put in.

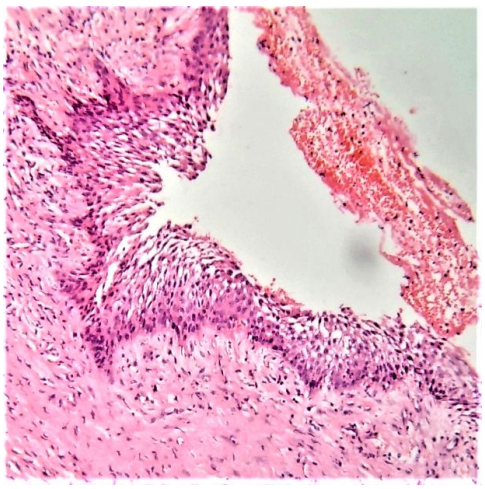
Eight months later (end of September, 2019) the patient is admitted once again at UMHAT "St. George"- Plovdiv with the same symptoms (severe oedema and pain) in the area of the previous operation. The clinical examination again shows expansion in the left vestibular area of the mandible. Under general anesthesia is performed a resection of the alveolar ridge, along with the newly formed tumour formation.

Material and metods

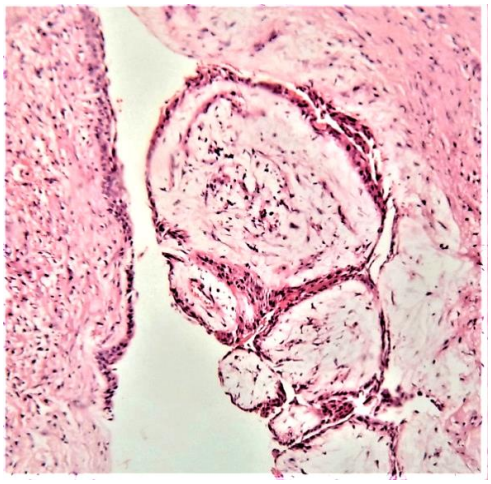
The histologic specimens were fixated with 10% neutral formalin and embedded in paraffin. The cut sections were 4 mkm thick and stained with hematoxylin and eosin (H-E).

Results

The biopsy from the first operation (№ 4070-72/2019) shows fragments from a benign epithelial cyst. These fragments, almost entirely, are lined with nonkeratinizing multilayered epithelium (fig.1), one area showing a plexiform structure, layered with unilayered cuboid epithelium, without atypia (fig.2).

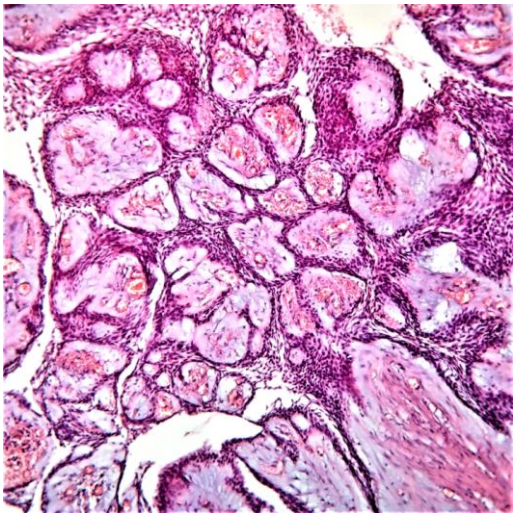


Wall of an odontogenic cyst, lined with multilayered epithelium - x 40, (fig.1)



Wall of an odontogenic cyst with a plexiform structure (right) - x 40, (Fig.2)

The biopsy from the second operation (№21953-55/2019) depicts the presence of unicystic type of ameloblastoma, with a predominant plexiform pattern (fig.3).



Ameloblastoma, Plexiform pattern - x 40, (Fig.3)

Discussion

The unicystic type comprises 10-15% of all ameloblastomas. This macroscopic form has an identical clinical and radiographic appearance with dentigerous cysts [1, 3]. The only way to differentiate between these two pathologic processes is by revising the biopsy sample from the operation. Moreover, ameloblastomas have a high recurrence rate, which means that the surgery requires the resection of a larger portion from the bone [5]. During a routine histological examination of a surgical biopsy, in the wall of a dentigerous cyst we may observe the presence of nests

of odontogenic epithelium with a palisade arrangement of basaloid cells [3]. There have been reported cases of ameloblastic changes in the mucosa of dentigerous cysts, however, without ameloblastoma developing in the following 7 years [6]. To set the diagnosis ameloblastoma on a biopsy sample, all signs of ameloblastic differentiation must be present.

For ameloblastoma to develop in a dentigerous cyst, what is characteristic is an initial intracapsular proliferation of odontogenic epithelium in the form of anastomosing fibres with thickening of the stroma around them [1]. What follows is intralumenal formation of a plexiform variant of the tumour. The odontogenic epithelium in the soft tissues in the wall of the cyst is arranged in a double-layered plexiform parent. Each epithelial line needs to have hypechromatic nuclei that are vacuolized, as well as characteristic reverse polarization away from the basement membrane. The stroma is presented by stellate reticulum-like cells, suprabasal cells composed of loosely arranged angular cells, which confirms the diagnosis – plexiform ameloblastoma [2].

In the case we are reporting, not all morphologic criteria are met, in order for us to set the diagnosis ameloblastoma on the biopsy sample from the first operation. In the wall of the epithelial cyst are present single plexiform structures, layered with cuboid epithelium, which does not change the initial diagnosis [3, 8]. Meanwhile, in the biopsy from the second operation is seen the typical morphologic picture for ameloblastoma, only 8 months after the first one. The short period of time, during which the tumour developed and grew in the place of the removed epithelial cyst, supports the presence of areas with tumour parenchyma in the cyst.

Conclusion

In order to assume there is a development of an ameloblastoma in the wall of a dentigerous cyst, not all morphological signs of the tumour must be seen. The presence of even one structure, characteristic for ameloblastoma, requires more attention in the follow-up of the patient, so we could prevent the tumour from forming, or, at least, find it in an early stage of its development

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КАТЕПСИН В КАК ВОЗМОЖНЫЙ ПРЕДИКТОР ГЕСТОЗА.

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АННОТАЦИЯ

Вопросы профилактики и прогнозирования осложнений беременности, определение тактики диагностических и терапевтических подходов при ведении гестации, в родах, послеродовом периоде, несмотря на достигнутые в этой области медицины успехи, далеки от разрешения.

В данной статье проведено исследование, одной из сторон проблемы патогенетических механизмов развития гестоза. Определено, что данный симптомокомплекс получает развитие преимущественно во второй половине беременности, на фоне различных степеней перинатального риска.

Установлено, что активность катепсина В – лизосомального фермента класса гидролаз повышается как в группе контроля при физиологическом течении беременности, так и при увеличении срока гестации,