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Clinical Report**ODONTOGENIC KERATOCYST OF MANDIBLE**

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Abstract: The Odontogenic Keratocyst is a developmental odontogenic cyst and deserves special attention because of its peculiar histopathologic features and biologic behavior. It is believed that the Odontogenic Keratocyst arises from the proliferation of remnants of dental lamina. It is usually asymptomatic, and solitary lesion, however, it may be associated with Nevroid Basal Cell Carcinoma Syndrome. This work aimed to present a case of a very extensive Odontogenic Keratocyst in a 28-year-old woman.

Keywords: odontogenic keratocyst, odontogenic cysts, basal cell carcinoma.

INTRODUCTION

Odontogenic Keratocyst was the term categorized by Philipsen (1956) as a distinct entity from other odontogenic cyst types. Its high recurrence rate, association with Nevroid Basal Cell Carcinoma Syndrome, typical histological features and aggressive biologic behavior [1], [2], [3], [4], [5], [6], [7], [8] places Odontogenic Keratocyst in a unique position within the spectrum of odontogenic lesions. [2]

Odontogenic Keratocyst may be found in patients who range in age from infancy to old age, nevertheless most of the cases (60%) with peak incidence between the second and fourth decades of life. [8] There is a male predilection. [7], [9], [10], [11], [12], [13] Odontogenic Keratocyst also may be seen in female gender. [8]

The mandible is involved in most of the cases with a marked predilection for the posterior body and ascending ramus. Large Odontogenic Keratocyst may be associated with pain, swelling, or drainage. [7], [8], [11]

Odontogenic Keratocysts tend to grow in an anteroposterior direction within the medullary cavity of the bone without producing bone expansion. Radiographically the Odontogenic

Keratocyst commonly is described as a unilocular radiolucency. [7], [11]

The histopathologic examinations of the Odontogenic Keratocyst typically displays thin and friable wall. The cystic lumen may contain a clear liquid that is similar to a transudate of serum, or it may be filled with a cheesy substance that consists of keratinaceous debris. Inflammatory infiltrate is not a common finding in this cyst. The epithelial lining consists of a uniform layer of stratified squamous epithelium, usually six to eight cells in thickness. The epithelium-connective tissue interface is often flat, and rete ridge formation is inconspicuous. Detachment of portions of the cyst-lining epithelium from the fibrous wall is usually observed. The luminal surface displays flattened parakeratotic epithelial cells, which present a wavy or corrugated appearance. The basal epithelial layer consists in a palisaded layer of cuboidal or columnar epithelial cells, which are often hyperchromatic. Small satellite cysts, cords, or islands of odontogenic epithelium may be observed within the fibrous wall. In the case when there is an inflammatory process, the typical features of the Odontogenic Keratocyst may be changed. [11], [12], [14]

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Fig. 1 - Large unilocular well-circumscribed radiolucency extending from the left mandibular second premolar to the contralateral right mandibular canine.

Fig. 2 - Significant root resorption associated with the apex of the left mandibular canine.

Most Odontogenic Keratocyst are treated by enucleation and curettage.^{[11], [15]} Bone grafting may be used after treatment of large Odontogenic Keratocyst to reduce risk of pathological break.^[16]

CASE REPORT

A 28-year-old woman was referred to the Oral and Maxillofacial Surgery Service, Emergency and Trauma Hospital, João Pessoa, Paraíba, Brazil for evaluation of a symptomatic mass in anterior portion of the mandible. The patient had noted the mass growing and mobility teeth approximately three months earlier and denied any history of previous trauma. Clinically, a hardened tumor could be observed, producing swelling and facial asymmetry at the anterior portion of the mandible.

The patient complained of pain during swallowing and bleed at teeth brushing. It was observed mobility of the all associated teeth with the lesion. Radiographic examination showed a unilocular circumscribed radiolucency that extended since the left mandibular second premolar to the contralateral right mandibular canine. Buccal cortical expansion and thin and resorption associated with the apex of the left mandibular canine was observed [Fig. - 1], [Fig. - 2].

There was clinical suspect that may be an Odontogenic Keratocyst; therefore it was done a punching which obtained a clear liquid that was similar to a transudate of serum inner the cystic lumen. During the treatment that consisted by enucleation and curettage, there was breaking of the cortical bone. After removal the specimens were sent to histopathologic examination.

The histological sections revealed a cystic formation with wall of the connective tissue with a few cells, dilated vessels and hemorrhage. The cystic lumen was partially filled a clear liquid that is similar to a transudate of serum. The epithelium-connective tissue interface shown flat and rete ridge formation was imperceptible. The epithelial lining was composed of a uniform layer of stratified squamous epithelium, with six cells thickness. The luminal surface presented flattened parakeratotic epithelial cell, which display a corrugated appearance [Figure - 3]. The basal

epithelial layer consisted in a palisaded layer of columnar epithelial cell with hyperchromatic nuclei [Figure - 4]. Areas of the mixed inflammatory cell infiltrate in the connective circumjacent tissue were observed.

The histopathologic features were compatible with Odontogenic Keratocyst. Up to now the patient did not show any evidence of recurrence or manifestations of the Nevroid Basal Cell Carcinoma Syndrome.

DISCUSSION

The Odontogenic Keratocyst is a developmental odontogenic cyst that may be considered a special entity for its specific histopathologic features and biological behavior.^{[1], [2], [3], [6], [7], [8], [11]}

Different from the mentioned studies^{[7], [11]}, which report that the Odontogenic Keratocyst has a male predilection, this case was detected in a female.^[8] The mandible is more often involved than the maxilla, and the lesions occur mostly in the molar - angle-ascending ramus area.^{[9], [11], [17]} Our case showed occurrence in the anteroposterior portion of the mandible, crossing the midline accord with the findings of a few authors.^{[7], [11]} This case represented a large Odontogenic Keratocyst associated with mobility teeth, pain during swallowing and bleed at teeth brushing.

Radiographically, this Odontogenic Keratocyst showed a unilocular circumscribe radiolucent area. Expansion of the bone resulting in facial asymmetry. This fact may be important in differential clinical and radiologic diagnosis because large dentigerous and radicular cysts are often associated with bony expansion.^[11] Resorption of the roots of the teeth adjacent to the lesion is little common.^[11] Nevertheless, in this case there was resorption associated with the apex of the left mandibular canine.

The histopathologic features of this case were basically identical to those previously reported.^{[11], [12], [14]} The Odontogenic Keratocyst typically display a thin friable wall. The cystic cavity contained a clear liquid that is like to a transudate of serum or a

Fig. 3 - The thin parakeratotic epithelial lining displays a parakeratinized surface with corrugated appearance. The cystic lumen is partially filled with a liquid. (HE - 100x).

Fig. 4 - The basal epithelial layer presents hyperchromatic, columnar cells. The thin epithelial lining shows a parakeratinized surface with corrugated appearance (HE - 400x).

material that consists of keratinaceous debris. Inflammatory infiltrate is uncommon.^{[7], [11]} In this case were observed areas of the mixed inflammatory cells infiltrate in the connective circumjacent tissue, beyond numerous vessels that sometimes showed engorged with red blood cells, and areas of erythrocyte extravasations. The epithelial lining consisted of a uniform layer of stratified squamous epithelium, often six to eight cells in thickness. The epithelium-connective tissue interface shown flat and rete ridge formation was imperceptible.

The luminal surface presented flattened parakeratotic epithelial cells, which display a corrugated appearance. The basal epithelial layer consists in a palisaded layer of columnar epithelial cell with hyperchromatic nuclei. The Odontogenic Keratocyst was treated by enucleation and curettage. During the treatment that consisted by enucleation and curettage, there was breaking of the cortical bone that represented a significant fact happened in this case. Although previous report mentioned high recurrence rate associated with Odontogenic Keratocysts and its possible association with Nevroid Basal Cell Carcinoma Syndrome^[11], up to now, the patient of the present case did not show any evidence of recurrence or manifestations of the Nevroid Basal Cell Carcinoma Syndrome.

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Clinical Report

LARYNGEAL CYST- CASE REPORT

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Abstract: Laryngeal cysts are rare, generally benign lesions which can affect all age groups. We report a case of huge supraglottic cyst. A 19 years old female presented with hoarseness and airway obstruction secondary to large cystic mass involving the entire right side of supraglottis attached to right aryepiglottic fold. Cyst was dissected by laryngofissure approach. On regular follow up for 3 years there is no recurrence of the lesion.

Keywords: Laryngeal cyst, supraglottis, laryngofissure

INTRODUCTION:

Cysts of larynx are usually benign. Interference with normal laryngeal function, specifically respiration, has resulted in death in infants and adults. The majorities of cysts seen in adulthood are asymptomatic or occur with mild symptoms such as a lump in throat or voice alterations. Neonates and infants generally present with stridor. These patients require careful evaluation since the smaller airway manifests a much greater potential for obstruction. The most common site of laryngeal cysts is on the lingual surface of epiglottis but the lesions may occur within the endolarynx and can involve aryepiglottic folds. Differentiation from malignant tumour as well as supraglottitis must be made during evaluation. Treatment is dependent on size, anatomical location and degree of respiratory compromise. Tracheostomy may be necessary to ensure and protect a patient airway prior to definitive treatment of the problem. Main stay of treatment is surgical excision by various ways. We report successful management of huge supraglottic cyst by laryngofissure method.

CASE REPORT:

The present case is a 19 years old girl, who presented with complaints of change of voice since two months and difficulty in respiration since one month. On indirect laryngoscopic examination there was huge globular cyst like lesion involving right side of supraglottis and rest of laryngopharynx was not seen. Neck was normal and rest of ENT examination revealed no

abnormality. Direct laryngoscopy was done which revealed wide based cystic lesion in supraglottis attached to right aryepiglottic fold. CT larynx both plain and contrast (Scan 1) was done which showed well defined hypodense cystic lesion of size approximately 3.2 x 3.1 cm in supraglottis with smooth enhancement of cyst wall and effacement of right aryepiglottic fold. Cyst was operated under general anaesthesia by laryngofissure approach (Fig 1). Cyst was found to be arising from right aryepiglottic fold, cyst was dissected completely and was sent for histopathological examination. Tracheostomy was done at the end of procedure anticipating postoperative oedema. Size of cyst was 3 x 3 cm and on aspiration was found to contain thin mucinous fluid and histopathological report was simple cyst.

Post operatively patient was put on antibiotics and steroids in tapering doses for 10 days and was decanulated at the end of three weeks. On regular follow up of patient, there is no evidence of recurrence of cyst.

DISCUSSION:

Cysts of larynx are rare lesions. They are routinely identified on mirror laryngoscopy and may go untreated if asymptomatic, their incidence is unknown. In 1938, New and Erich reported 35 cases of cysts of larynx in a series of 722 benign laryngeal tumours. The most common locations of laryngeal cysts are the lingual surface of epiglottis, the free margin of false cords, arytenoids and aryepiglottic folds, pyriform fossa and the ventricle. Laryngeal cysts are found at any age, although they occur less frequently