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Pleomorphic adenoma originating from parotid salivary gland in an 11-year-old boy: a case report

Adenoma pleomórfico proveniente da glândula salivar parótida em um paciente de 11 anos de idade: relato de caso

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Abstract

Introduction: Salivary gland tumors are rare and make up to 3% of head and neck tumors. Pleomorphic adenoma (PA) comprises 80-90% of benign parotid neoplasms. It presents several histological features due to the different compound with myxoid and chondroid matrix. Its main characteristic**s** are high recurrence rate and frequent malignant conversion. Symptoms are usually rare in most of the cases, and the only sign is asymptomatic swelling. **Case report**: The present report describes a rare case of Pleomorphic adenoma of the left parotid gland in an 11-year-old male.

Keywords: Salivary gland tumors. Pleomorphic adenoma. Myxoid and chondroid matrix. Malignant conversion.

Resumo

Introdução: *Tumores em glândulas salivares são raros, representando aproximadamente 3% de todos os tumores de cabeça e pescoço. O Adenoma Pleomórfico (AP) compreende cerca de 80-90% das neoplasias benignas de parótida. Esta lesão apresenta muitas características histológicas devido ao diferente componente matricial mixóide e condroide. Suas principais características consistem no elevado índice de recorrência e frequente evolução para quadros de malignidade. Relatos sintomatológicos são geralmente raros na maioria dos casos, sendo o único sinal o aumento de volume local assintomático.* ***Relato de caso:*** *O presente relato descreve um caso raro de Adenoma Pleomórfico na glândula salivar parótida do lado esquerdo em um paciente do sexo masculino de 11 anos de idade.*

Palavras-chave*: Tumores de glândulas salivares. Adenoma pleomórfico. Matriz mixóide e condroide. Evolução para malignidade.*

Introduction

Salivary gland tumors are rare and make up to 3% of head and neck tumors (1). Approximately 90% of the benign neoplasm of the major salivary gland is associated with the parotid gland. Pleomorphic adenoma (PA) comprises 80-90% of these benign parotid neoplasms. Such adenoma of the submandibular and sublingual gland is quite uncommon and comprises the rest (8-10%) of the group (2). It is generally considered as a benign tumor, even though this lesion has several histological features due to the different compound with myxoid and chondroid matrix.

The main characteristic**s** are high recurrence rate and frequent malignant conversion. Symptoms are usually rare or not significant in most of the cases, and the only sign is the asymptomatic swelling that slowly grows in the parotid region without involving facial nerve, whose function remains unchanged. Often, onset of facial nerve deficit, changes in consistency, more rapid growth, and pain are signs of malignant transformation (3,4).

The incidence of parotid tumor is about 2.4 in 100000/year. The right side involvement is more common than the left side, being more common in female than male (2:1). Its incidence is rare in children, and in adults is more commonly found between the ages of 50 and 60 years (5-7). The exact etiology is unknown, however, it has been noted that the incidence increases 15-20 years after exposure to radiation. Martinelli et al. (8) suggested that the simian virus (SV40) may play an etiologic role in the development of pleomorphic adenoma. The present report describes a rare case of pleomorphic adenoma of the left parotid gland in an 11-year-old male.

Case report

An 11-year-old male presented with a swelling in the left front region of the ear for two and half months. The swelling was initially small in size and gradually increased to the size of 2x2cm, not associated with pain, fever, loss of weight, loss of appetite, or related to meals. There was no history of exposure or contact with tuberculosis.

Clinical examination showed that spherical shape swelling was present measuring 2x2 cm in diameter in the left preauricular region (Figure 1). Swelling has ill-defined and irregular border, the surface was smooth and the skin over the swelling was normal in color. It was not tender on palpation and temperature was not raised. The consistency of swelling was firm and fluctuation was absent. Swelling was not fixed to overlying skin. There was a bilateral submandibular lymph node palpable, mobile, single in number, non-tender, and soft in consistency. Intraoral examination showed no abnormalities, and opening of Stenson's duct seemed normal. Other systemic examinations were unremarkable.

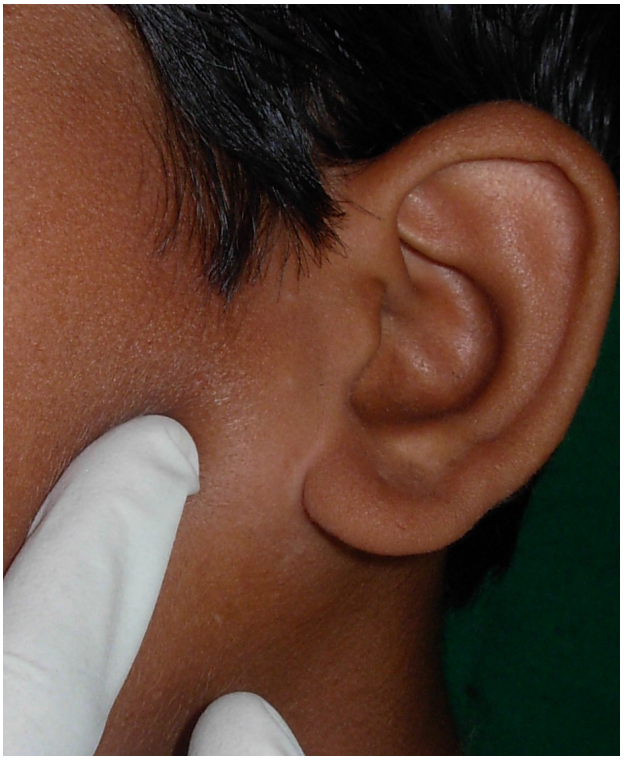

Figure 1 - Clinical extraoral photograph of swelling in the left preauricular region

When swelling is seen in front of the ear it is important to formulate a differential diagnosis, since this would help further evaluation of the condition and management of the patient. After considering all clinical findings, following entities were considered in differential diagnosis: pleomorphic adenoma, lymphadenitis, and fibroma. After that, the patient was investigated with complete hemogram, intra oral radiographs, orthopantomograph, ultrasonography, and fine-needle aspiration cytology.

Routine hematological investigations were within normal limit. Orthopantomograph shows no evidence of abnormalities. Ultrasonographic findings of swelling were very clear boundary rounded in shape hypoechoic lesion. The ultrasound architecture of the lesion was homogenous. Posterior echoes were unchanged, and ultrasound characteristics of tissues were solid. Ultrasonographic impression was benign parotid mass (Figure 2).

Fine-needle aspiration biopsy was performed on the patient using 25 gauze needles after locally anesthetizing the overlying skin with 1% xylocaine. The aspirated material was expressed on fully frosted glass slide and smeared with plain glass slide. The fully frosted glass slide were immediately fixed in 95% ethanol and stained by standard papanicolaou technique. The plain slide was air dried and stained by modified Wright stain. The needles were rinsed in carbowax solution, and cytocentrifuged smears were prepared. Fine-needle aspiration revealed cellular smears composed of a mixture of epithelial and stromal components. The epithelial cells were arranged in loose sheets and singly distributed displaying plasmacytoid nuclei with bland nuclear chromatin and dense cytoplasm. The stromal component is composed of fibrillary chondromyxoid substance admixed with spindle-shaped mesenchymal cells (Figure 3). These cytological features are consistent with pleomorphic adenoma.

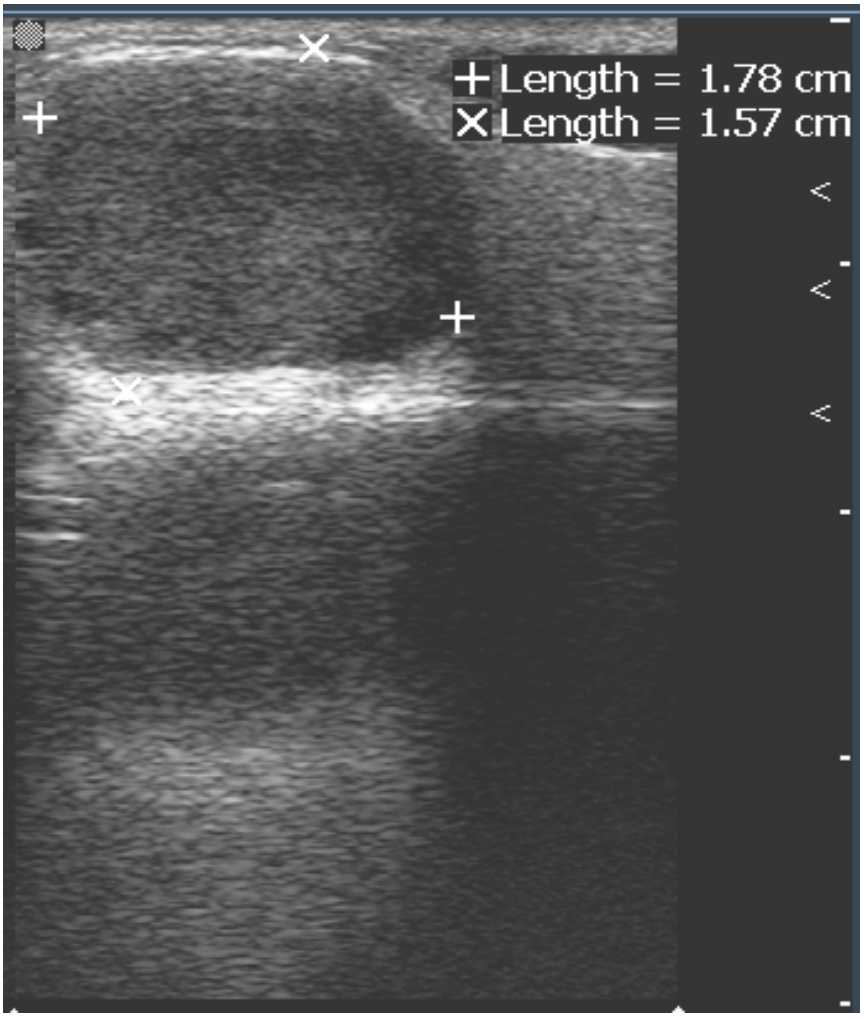

Figure 2 - Ultrasonographic image shows a hypoechoic and homogeneous solid lesion sugestive of benign parotid mass

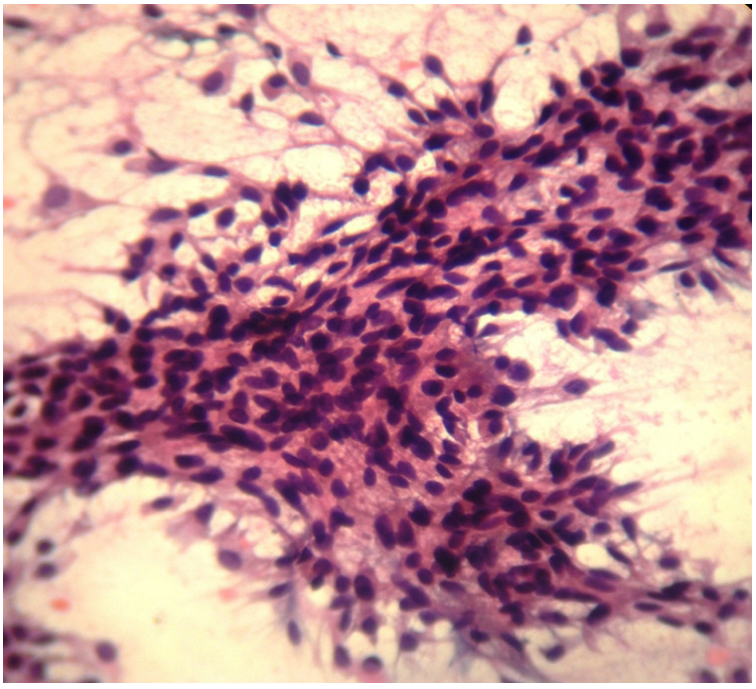

Figure 3 - The fine-needle aspiration cytology report shows admixed epithelial, myosepithelial, and mesenchymal component

The choice of treatment of pleomorphic adenoma of the parotid gland depends on the agressiveness of the tumor, the extension of the mass, and its relation with the facial nerve. The surgical treatment of benign tumor of the parotid gland can consist of enucleation, enucleoresection, and superficial or total parotidectomy with preservation of the facial nerve, where possible. The superficial parotidectomy and/or total parotidectomy with preservation of the facial nerve gives excellent results. In the present case, since the lesion is small and located in the superficial lobe of the parotid gland, superficial parotidectomy was performed with preservation of the facial nerve. Postoperative recovery was uneventful. The patient is followed for up to 24 months and free from the disease (follow up photograph were not available).

Discussion

Pleomorphic adenoma (PA) is characterized by great histological diversity, and myoepithelial cells are considered responsible for production of the extracellular matrix. It is the most common of all salivary gland neoplasms, accounting for 50-70% of benign parotid tumors. Salivary gland tumors are comparatively rare disorders in the paediatrics population, comprising only 1% of all the head and neck tumors and less than 5% of all the salivary gland tumors (9).

The term "pleomorphic" refers to both the histogenesis and the histology of the tumor (10). Pleomorphic adenoma is a benign salivary gland tumor with wide cytomorphologic and architectural diversity. The tumor has three components: an epithelial cell component; myoepithelial cell component; and a stromal (mesenchymal) component. The identification of these three components, which may commonly vary from one tumor to another, is essential to the recognition of pleomorphic adenoma. Fine-needle aspiration biopsy was done for the diagnosis of pleomorphic adenoma (11,12).

Imaging plays an important role, not in the diagnosis of the mass, but to accurately locate the intra or extra parotid location of these lesions. This has important implications for the surgical management of the patient. Parotidectomy with facial nerve preservation is the method of choice in treating benign lesions of the parotid gland (13). This technique is diagnostic, and the risk of recurrence is low in most cases, with a very low risk of permanent facial nerve injury. There is a significant risk of tumor recurrence, often multifocal, with incomplete excision (14). The standard approach to parotid mass is clinical examination followed by fine-needle aspiration.

An ultrasound examination combined with fine-needle aspiration is the preferred choice. This allows the correct location of the lesion and ensures adequate material is obtained for cytological examination. Lesions of the parotid are a diagnostic challenge to the radiologist. Knowledge of the parotid gland anatomy with use of imaging is essential in the accurate localization of these lesions (14). In the present case, since the lesion is small and situated in the superficial lobe of parotid gland, superficial parotidectomy was performed.

Recurrence is low in total parotidectomy compared with enucleation and superficial parotidectomy (7). There was up to 43% of recurrence after surgical resection, since pleomorphic adenoma does not have true capsule. They have pseudocapsule extending into surrounding normal gland tissue. During enucleation, while the main tumor mass is being resected, pseudopodes are left in the gland, giving rise to multiple nodular recurrences and risk of malignant changes (15).

Generally, malignant transformation can be suspected with a sudden increase in growth, pain, ulceration, spontaneous bleeding, and superficial and deep tissue invasion. The incidence of malignancy frequently shows a correlation between the length of the history of pleomorphic adenoma (23 years on average) and the development of carcinoma. Long standing tumors may show malignant change. Therefore, early diagnosis and treatment of pleomorphic adenoma is essential (16).

Conclusion

Pleomorphic adenoma is a benign tumor, most commonly involving parotid gland. Proper diagnosis with fine-needle aspiration cytology and careful surgical resection with preservation of facial nerve has proven to be highly accurate, and helps in better prognosis with least chances of recurrence.

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