Case Report

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Cytodiagnosis of an Unusual Adult Parotid Lymphangioma

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Abstract

Lymphangioma is an uncommon benign congenital tumour of lymphatics that is seen in children and rarely in adults. Lymphangiomas primarily involving the parotid gland is an extremely uncommon occurrence in adults. It is commonly diagnosed on histopathology. There are very few case reports in English literature, on role of fine needle aspiration cytology (FNAC) in diagnosing these lesions. Herein, we report on the cytological findings of a primary parotid lymphangioma in a 33-year-old male patient. Cytological smear studied reveal small, round lymphocytes without atypia with occasional benign appearing salivary gland epithelium against proteinaceous background. In our case, the cytological finding along with computed tomography (CT) findings allowed the diagnosis of lymphangioma. The cytological diagnosis is as accurate as histopathology. Hence lymphangioma of parotid gland can be accurately diagnosed on FNAC in outpatient department, which is simple, inexpensive and routine procedure for rapid diagnosis.

Keywords: Lymphangioma; Parotid Gland; Cytology.

Introduction

Lymphangioma is a benign congenital tumour that is more commonly seen in children below the age of two years and rarely in adults. In adults lymphangiomas may occur either spontaneously, or in response to infection or trauma [1].

The presentation of adult lymphangiomas is usually asymptomatic, has no gender predilection and diagnosed at the late stage due to painless mass that grows slowly. It is rarely seen in parotid gland and when it occurs, the gland is usually incorporated by lymphanioma of surrounding tissue [2] .

To our knowledge less than 100 adult cases lymphanioma have been reported in the English literature [1]. Lymphanioma primarily involving the parotid gland is an extremely uncommon occurrence in adults.

Hence herein we report a case of an unusual adult primary parotid lymphangioma on the left side in a 33-year-old male patient, presented with swelling size measuring 8 X 5 X 4cms over parotid region since 8 months. The diagnosis was made on fine needle aspiration cytology (FNAC) and presented here, in view of limited data in the literature on role of FNAC in diagnosing such lesions.

Case Report

A 33-year-old male presented to the Surgical Out Patient Department with a slowly enlarging mass on the left side of the parotid region since 8 months. He complained of mild discomfort and pain during mastication. There was no history of trauma, infection, trismus, dysphagia or dyspnoea. Local examination revealed a single, soft, cystic, fluctuant, mobile swelling measuring 8 X5 X 4cms over left mandibular region just below the ear lobe (figure 1 and 2). Computed tomography (CT) revealed an ill-defined

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heterogeneous lesion with variable degrees of enhancement involving cheek, mandibular, and masticator spaces on the left side of the space. Clinical diagnosis of cystic lesion of parotid was made. Fine needle aspiration cytology (FNAC) of the swelling was performed under all aseptic precautions. A 20 ml of straw- yellow coloured fluid was aspirated which reduced the swelling size. Smear studied from the centrifuged deposits were air dried and alcohol fixed. Air dried smears were stained with May- Grunwald – Giemsa stain. Alcohol fixed was stained with Papanicolaou and haematoxylin and eosin stains (Hand E). Smear studied show mature red blood cells, mature small, round lymphocytes without mitosis or atypical cells and few of the lymphocytes show degenerative changes against proteinaceous background. (Figure 3 and 4). Hence, final diagnosis of benign cystic lesion suggestive of lymphangioma was rendered and adviced for excisional biopsy.

Gross

The excised cystic mass measures 7x6x3cms with multilocular appearance filled with yellowish fluid (figure 5).

Histopathological Features

Multiple sections of the cyst reveal large irregular spaces filled with eosinophilic proteinaceous material and spaces lined by endothelial cells. The cystic spaces are separated by intervening fibrous tissue and lymphoid aggregates (figure 6). The excision was complete and there was no evidence of recurrence till the final review of the case.

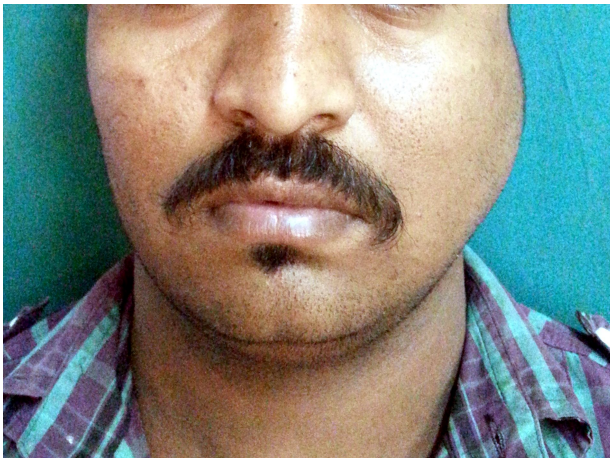

Fig. 1: Solitary ill-defined swelling over the parotid region.

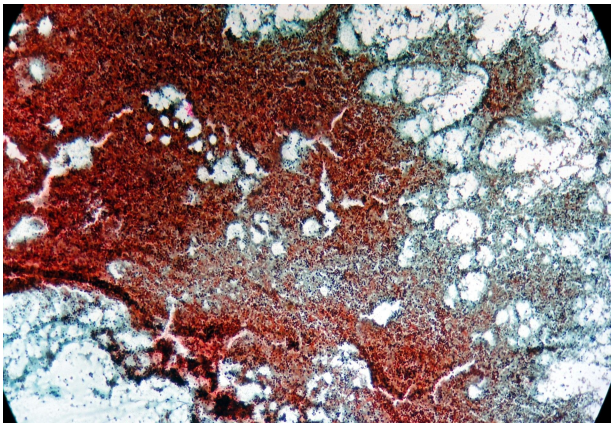

Fig. 3: Cytological smear shows scattered lymphocytes against proteinaceous background (Pap, x100)

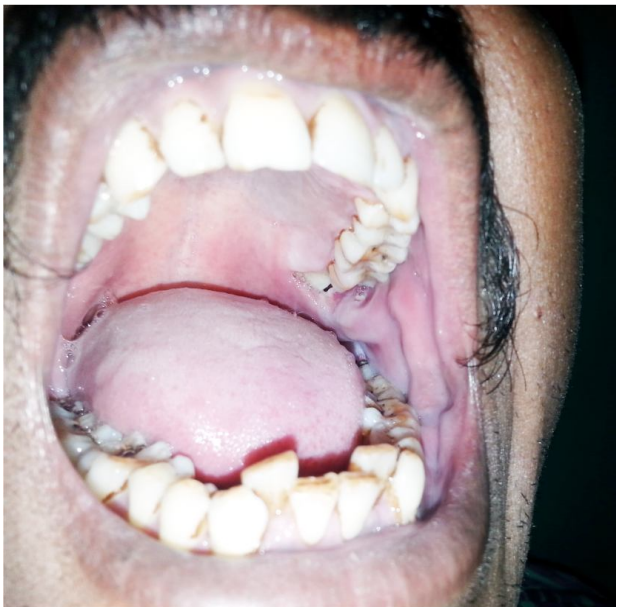

Fig. 2: Intraoral swelling of the parotid gland.

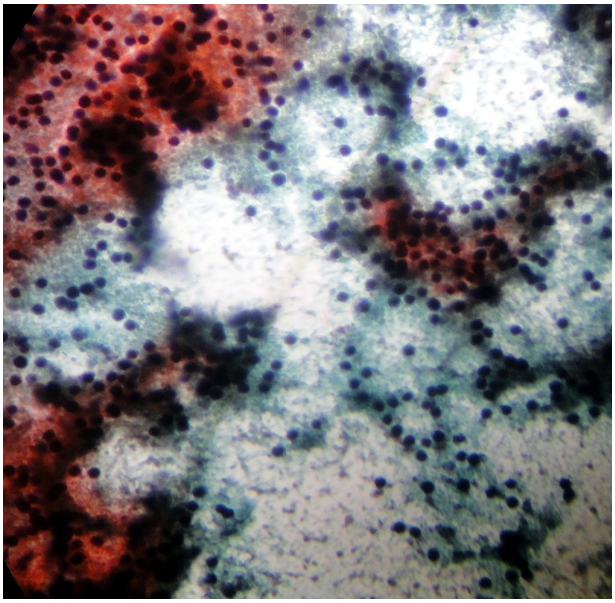

Fig. 4: Smear shows matured, round lymphocytes without atypia/mitosis (Pap, x400)

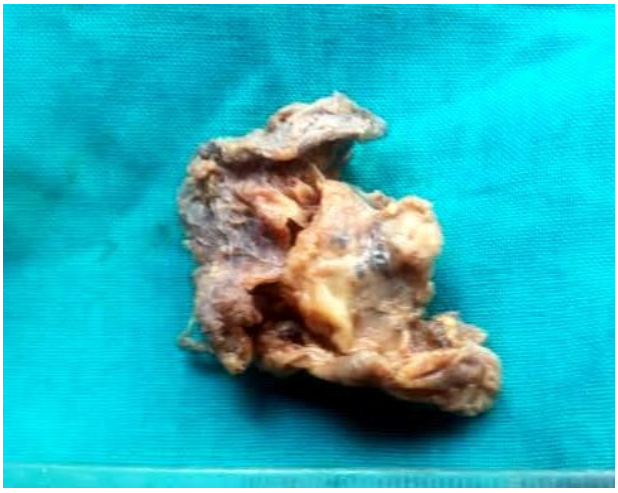

Fig. 5: Cut section of the tumour shows multilocular appearance filled with yellowish fluid.

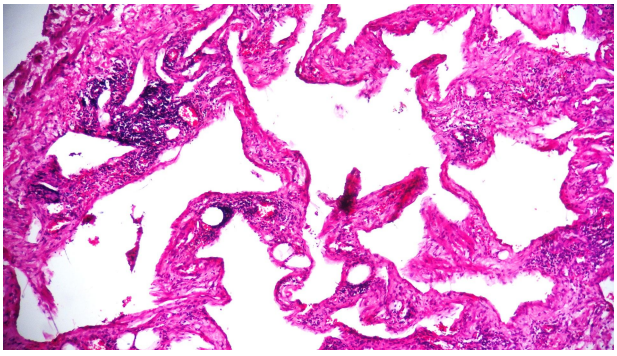

Fig. 6: Section shows cystic spaces lined by endothelial cells with aggregates of mature lymphocytes (H and E, x400)

Discussion

Lymphangioma is a congenital malformation of the lymphatic tissue that fail to communicate normally with the lymphatic system [3]. The common sites of lymphangioma are posterior triangle of neck (75%), axilla (20%), mediastinum (5%), groin, reteroperitonial space and Pelvis [4].

In 1828, Redenbacher first described a lymphangioma lesion. Lymphangiomas constitutes 4% of all vascular tumors in children [5]. Lymphangiomas are classified as lymphangioma simplex, cavernous lymphangioma and cystic lymphangioma or cystic hygroma based on the size of lumen of the lymphatic vessel [6].

The clinical differential diagnosis of cystic lesions of parotid gland in adult includes benign lymphoepthelial lesions, branchial cleft cysts, chronic siloadenitis, cystic warthin's tumour, cystic pleomorphic adenoma, cystic mucoepidermoid carcinoma and lymphoma.

The cytological features of lymphoepthelial lesions show lymphoid cells with few degenerated spindle metaplastic epithelial cells. Branchial cleft cysts show anucleated and nucleated squamous epithelial cells against inflammatory background, chronic siloadenitis show sheets of ductal epithelial cells with reactive changes and few squamous metaplastic cells admixed with chronic inflammatory cells. Cystic warthin's tumor show mild cellularity consisting of small cohesive group of bland oncocytic epithelial cells admixed with lymphocytes. Wherein, cystic pleomorphic adenoma, cystic mucoepidermoid carcinoma practically reveals acelluar smears containing cells positive for malignancy.

Although the lymphangioma is a benign lesion it may cause significant morbidity because of its large size, critical location or due to proclivity to become secondarily infected. Factors which predict good prognosis include head and neck location, size <5cms and macro cystic architecture.

The treatment modalities for lymphangiomas are surgical resection, radiation therapy, cryotherapy, electrocautery, sclerotherapy steroid administration, ligation and radiofrequency tissue ablation technique [7]. The common post-operative complications encountered are recurrence which is most common, joint contracture, hypertrophy of the scar and fluid accumulation [8]. A study however recommends a two-year post surgical follow-up to detect any recurrence [1]. In our case, the excision was complete and there was no recurrence up to three-year post surgical follow-up.

Conclusion

Lymphangiomas in adult are uncommon and rarely present as parotid swelling. Though the case posed a diagnostic challenge, it was accurately diagnosed by FNAC. Here we emphasized the diagnosis of lymphangiomas by preoperative FNAC and imaging in the management of this lesion in our article.

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