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Case Report

Pathology Section

Lymphangiomatous Polyp of Palatine Tonsil in A Child Presenting with Dysphagia and Dysarthria

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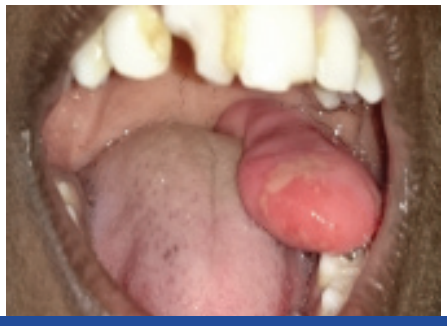
ABSTRACT

Pedunculated lymphangiomatous polyps of the tonsil are rare benign lesions that have been described in literature using varied nomenclature. Majority of the cases have been reported in adults with varying clinical symptoms. We report a case of lymphangiomatous polyp of left palatine tonsil in a 14-year-old male child who presented with dysphagia and dysarthria. Clinical examination revealed a large pedunculated polyp arising from upper pole of left tonsil. Patient underwent left tonsillectomy with excision of the polyp. Based on histopathological features a diagnosis of pedunculated lymphangiomatous polyp was made. We discuss the clinical and histopathological features of this lesion with differential diagnosis and short review of literature.

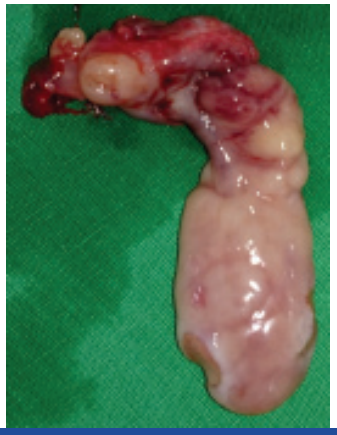
Keywords: Hamartoma, Lymphangioma, Pedunculated polyp, Tonsil

CASE REPORT

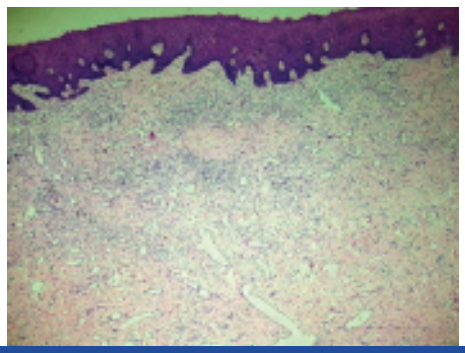
A 14-year-old boy presented with one year history of mass in the oral cavity which was initially small and progressively increased in size. He complained of difficulty in speaking and swallowing since four months. There was no history of respiratory difficulty sore throat or bleeding. Patient was undergoing psychiatric treatment for behavioral problems and is on resperidone therapy. Otorhinological examination of oral cavity revealed a smooth pinkish white mobile nontender firm pedunculated mass arising from superior pole of left tonsil and extending over the anterolateral surface of the tongue [Table/Fig- 1]. Patient could voluntarily swallow and regurgitate the mass. Rest of the oral cavity, nasopharynx and larynx were normal. There was no evidence of cervical lymphadenopathy. Systemic examination and all other routine investigations were normal. A left tonsillectomy with tonsillar mass excision was done under general anesthesia and the specimen was sent for histopathological examination. Grossly the left tonsil measured 2.5x1.5cms. A polypoid large smooth mass was seen arising from the superior pole of the tonsil attached to it by a small stalk measuring 7x2x2cms [Table/Fig- 2]. Cut section was homogenous white tan and the mass had a firm consistency. Microscopic examination of hematoxylin and eosin stained sections showed a polypoid mass lined by nonkeratinising stratified squamous lining. Subepithelium showed a dense collection of lymphocytes along with few lymphoid follicles [Table/Fig-3]. Stroma of the polyp showed many dilated lymphatic and blood vessels admixed with fibrocollagenous and adipose tissue [Table/Fig- 4].No epitheliotropism was seen. Rest of the tonsil showed features of reactive hyperplasia. Based on the histological features a diagnosis of pedunculated lymphangiomatous polyp of the left palatine tonsil was given. Patient had an uneventful postoperative course and is doing well one month after the surgery



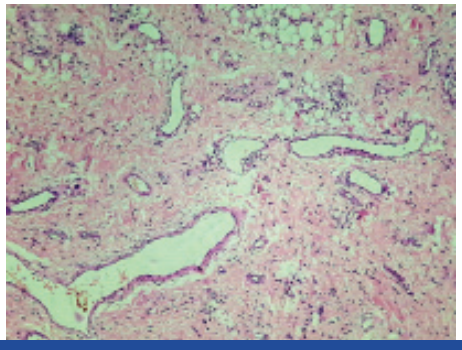
[Table/Fig-1]: Clinical photograph showing a large polypoid mass arising from the left tonsil and extending over the tongue



[Table/Fig-2]: Gross photograph showing a smooth pinkish white mass arising from the superior pole of the left tonsil



[Table/Fig-3]: Microscopic photograph showing a polyp lined by stratified squamous epithelium along with dense lymphocytic infiltrate and lymphatic channels in the subepithelium (HE x100)



[Table/Fig-4]: Microscopic photograph of stroma showing dilated lymphatic channels along with fibrous and adipose tissue (HE X400)

DISCUSSION

Lymphangiomatous polyps of palatine tonsils are uncommon lesions composed of varying proportions of lymphatic and blood vessels mixed with fibrous adipose and lymphoid tissue. Depending on the proportion of these mesenchymal components present in each case they have been described in literature using different terms like, lymphangiomatous polyp [1-3] lymphangioma [4] lymphangiectatic fibrolipomatous polyp [5] hamartomatous polyp [6] lipoma [7] fibrolipoma [8] and angiofibrolipoma [9] and others. Only 35 cases of lymphangiomatous polyps have been reported in world literature 10 being in children. All cases behaved in a benign fashion and no complications have been reported. The varying terminologies used and its rare occurrence has made it difficult for clinicians and pathologists to classify these lesions correctly. Hence, many authors believe that their true incidence may be higher than that reported in literature [1]. The pathogenesis of these lesions remains unclear. Chronic inflammation and obstruction of the lymphatic channels was earlier thought to be a possible mechanism. But this is unlikely because chronic tonsillitis is much more common than these polyps. Moreover, some patients do not give history of tonsillitis as in the present case [3]. Presently it is thought that these lesions are hamartomas rather than true neoplasms because the various elements found in polyps are also found in normal tonsil but are arranged in a haphazard fashion [1,3].

The cause could be an aberration during the development which may produce an hamartomatous focus with its own growth potential [4]. However, recently the cause of development of lymphangiomatous polyps is supposed to be a neoplastic proliferation of lymphatic vessels leading to a network of lymphatics that never achieve sufficient anastomosis with larger lymphatic vessels. This uncontrolled proliferation of lymphatic vessels is thought to be caused by a dysregulation of growth factors involved in lymphangiogenesis as Prox-1 and vascular endothelial factor (VEGF-C). Therefore, both lymphangiogenesis and angiogenesis may be contributing in the formation of these lesions [4]. Kardon et al., [1] have reviewed 26 cases of these lesions and described their clinical and histological features. The age range was between 3 to 13 y. Both sexes were equally affected and have the same age of presentation. Patients have presented with varied symptoms like mass in the oral cavity, dysphagia, tonsillitis, sore throat, lump in the throat, cough, respiratory difficulty, increased salivation, vomiting and blood in the sputum. All cases have shown unilateral involvement without side predilection. Our patient presented with dysarthria and dysphagia as the main symptom probably because of the large size of the lesion. Grossly these lesions are small in size with a range of 0.5 to 3.4cms and a mean size of 1.6cm.Our case presented with a size of 7cm which has not been reported earlier. They present as polypoid smooth white tan yellow homogenous masses which may be pedunculated or sessile. The main histological components of these polyps is an epithelial covering of squamous or respiratory type along with varying proportions of adipose tissue fibrocollagenous tissue blood vessels lymphatics and a lymphoid infiltrate Some cases may show follicle formation with presence of lymphocytes in the lumina of lymphatic channels and inside the epidermis. This is termed as epitheliotropism and was not seen in the present case. Barreto et al., [10] have studied the immuno profile of these lesions, even though immunohistochemistry is not necessary for their diagnosis. Vascular channels were reactive for factor VIII related antigen, CD31 and CD34.Lymphocytes within vascular channels and within the epithelium of the polyps were predominantly CD3 positive even though there was a polymorphous B cell and T cell immunophen notype expression. Based on the immunoprofile of vascular and stromal components both these authors suggested that tonsillar polyps were composed of disorganized connective tissue and lymphatic channels and can be considered as hamartomatous proliferations. Gunbey et al., [2] have reported a case in a child with dysphagia while Duggal et al., [11] have presented a case with cough and vomiting in a child with hamartomatous polyp.The differential diagnosis of these lesions include squamous papilloma, fibroepithelial polyp, juvenile angiofibroma and rarely epidermal cyst. Squamous papilloma is the commonest benign tumour of the tonsil with an exophytic surface epithelial proliferation arranged in multiple layers. It does not invade the underlying stroma and has no lymphocytic component. Lymphangioma is made of widely dilated vascular channels with luminal proteinaceous material and lymphocytes. Fibrocollagenous tissue adipose tissue and lymphoid tissue seen in the stroma of hamartomatous polyps is not seen in these cases. Juvenile angiofibroma usually arises from the nasopharynx and is an aggressive lesion. It has a cellular stroma composed of stellate and plump cells along with staghorn type thin walled vascular channels. Lymphangiomatous polyps in contrast have a paucicellular stroma. Epidermal cysts are lined by squamous lining and filled with keratinous material. Lymphangiomatous polyps have been treated with simple excision or tonsillectomy. No recurrence or malignant transformation has been reported in any case.

CONCLUSION

A very large lymphangiomatous polyp of tonsil represents an unusual presentation of a rare lesion in a child. This lesion needs to have a uniform terminology and should be considered a hamartoma rather than a true neoplasm.

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