INTRODUCTION

Inverted papilloma is a benign tumour that is characteristic by invagination of nasal and paranasal sinus mucosa epithelium into stroma. Although characterized as a benign tumor, it demonstrates a high recurrence rate and has the capacity to expand by local extension into vital structures. Concurrent squamous cell carcinoma (SCC) has been found in 3-10% of cases (Rafii et al., 2007; Roodsari, Naraghi & Pourkaveh, 2014; Topdag, Mutlu & Ila, 2014).

Inverted papilloma of the sinonasal tract is an uncommon lesion in adults and is even less common in children. It is may be found during every period of life. It often increases at the age of 50-60 years, and the proportion of males to females is 3 : 1. While most commonly found in adults, inverted papilloma can occur in children as well. The true incidence of inverted papilloma is therefore difficult to determine. The condition is most probably under-diagnosed, as it may coexist or develop alongside simple inflammatory polyp. It is possible to miss an inverted papilloma coexisting with simple inflammatory polyps, if biopsy sample collect inflammatory polyps only. Inverted papilloma is one of the most frequently seen benign lesions that originate from nasal and paranasal sinus mucosa epithelium stroma (from the Schneiderian membrane). Its incidence rate is 0.5-1.5 per 100,000 people in a year (Rafii et al., 2007; Anari & Carrie, 2010; Roodsari, Naraghi & Pourkaveh; Topdag, Mutlu & Ila, 2014).
The manner of presentation is similar to sinusitis and nasal polyposis, although unilateral nasal obstruction is the most common presenting symptom. Other clinical feature include epistaxis, epiphora and headache (Limaye, Mirani & Raz, 1989; Anari & Carrie, 2010; Visvanathan, 2010).

In its histology, the epithelium, inverted into connective tissue, is observed, and it is generally an exophytic and polypoid lesion. Different from inflammatory polyps, it is pinky and has a vascular appearance macroscopically, and it does not have goblet cells, koilocytes, or eosinophils in its structure microscopically (Limaye, Mirani, Raz, 1989; Topdag, Mutlu & Ilia, 2014).

The aim of surgical treatment of sinonasal inverted papilloma is to remove the whole disease. The method of surgical treatment that offers the best operative exposure and least morbidity is dictated by the extent of the disease and the surgeon's technical ability (Sautter et al., 2007; Anari & Carrie, 2010).

Here we present a case of pediatric inverted papilloma coexisting with inflammatory polyp of a 10-year-old girl.

CASE REPORT

A 10-year-old girl came to ENT Department of Haji Adam Malik General Hospital on November 13th 2015 with the mass filling right nasal cavity as a chief complain. This mass initially small and gradually grow bigger for 3 years (Fig. 1). She also complained nasal obstruction. There was history of nose bleed, facial pain and headache. She did biopsy on the mass and the result was benign lesion, histopathology represent an inverted papilloma (Fig. 2). On November 23rd 2015 she came with dyspnoe as a chief complain. We referred her to Division of Pharyngo-
laryngology for the management of airway obstruction and they did a tracheostomy procedure.

Fig. 1 The mass filling right nasal cavity

Fig. 2 Histopathology of biopsy

The physical examination of the ear was normal. In the left nasal cavity we found a mass filling whole left nasal cavity, easily bleed when we touched. In oropharynx we found the soft palate was pushed downward by the mass.

CT Scan examination show that there was soft tissue mass filling left maxillary sinus, nasal cavity, choana, nasopharynx and oropharynx,
pushed nasal septum to the right side and there was destruction of medial wall of left maxillary sinus (Fig. 3). She underwent some examination e.g. blood laboratory test and chest x-ray, the result was in normal limits.

![Axial Nasopharyngeal CT Scan](image)

We diagnosed this patient with pediatric inverted papilloma by clinical history, physical examination and also biopsy of the mass. We planned resection with rhinotomy lateral or transpalatal approach.

The surgery was done on December 8th 2015 in H. Adam Malik General Hospital Operating Theatre. Surgery was done under general
anesthesia. We planned resection with rhinotomy lateral or transpalatal approach, but at the end we decided did a polypectomy.

Patient was placed in a supine position with a slight elevation of head end. After desinfection, we evaluated the patient then we decide to did a transoral and transnasal tumor removal. The oral cavity was kept open by using mouthgag, then we released the mass with finger from the nasopharynx area while the assistent pushed the mass from the nasal cavity with the finger until mass detach and the mass was taken out from mouth. We saw a polypoid mass, half of this polypoid mass showed vascular appearance (Fig 4.). We put on a bellocq tampon for two days.

![Fig. 4 Polypoid mass with vascular appearance partially](image)

After the surgery patient was treated for 3 days and was given antibiotic, anti haemorrhagic, analgetic and steroid (for one day) intravenously. The patient was discharge from the hospital on the 3rd postoperative day (fig. 5). The post operative pathology result was evaluated as inverted papilloma coexisting with inflammatory polyp (Fig. 6).
DISCUSSION

The World Health Organization (WHO) defines inverted papilloma as a benign epithelial tumour composed of well differentiated columnar or ciliated respiratory epithelium having variable squamous differentiation. The WHO classifies inverted papilloma as a subgroup of Schneiderian papillomas. Schneiderian papillomas are papillomas arising in the sinonasal tract, which is lined with schneiderian epithelium or squamous
epithelium, ectodermally derived respiratory mucosa. This distinctive epithelium can give rise to three histologically unique types of papillomas: exophytic, inverted and oncocytic papillomas. In 1991, Reingertz was the first to histologically describe appearance of inverted papillomas (Steiger, 2007; Anari & Carrie, 2010; Vorasubin et al., 2013; Zimmer & Carrau, 2014; Mallen & Suh, 2016).

Sinonasal inverted papilloma is a benign lesion that occurs in the nasal cavity and paranasal sinuses. Associated clinical problems include a tendency towards local destruction, recurrence and malignant transformation into squamous cell carcinoma. The term inverted papilloma describes the histological appearance of the epithelium, inverting into stroma, with a distinct and intact basement membrane that separates and defines the epithelial component from the underlying connective tissue stroma (Limaye, Mirani, Raz, 1989; Karkos et al., 2009; Anari & Carrie, 2010; Dhingra 2013).

Inverted papilloma patients typically present with nonspecific signs and symptoms, including unilateral nasal obstruction, nasal polyps, epistaxis, rhinorrhea, hyposmia and frontal headache. Accordance with previous statement, our patient complained about nasal obstruction, epistaxis and headache. These tumours are rare in children. Even it is rare, we should considered in the differential diagnosis of unilateral lesions of the sinonasal tract. In over 1000 cases seen at Armed Forces Institute of Pathology, there were only 4 pediatric cases (Limaye, Mirani, Raz, 1989; Karkos et al, 2009; Anari & Carrie, 2010).

Inverted papilloma usually appears large, firm, and gray in color with a multinodular, polypoid, uneven surface. Accordance with the mass appearance of our patient. Histologically, inverted papilloma shows markedly thick inverted or endophytic growth of nonkeratinizing transitional cells. The thick epithelium undergoes squamous maturation and inverts
into stroma with a distinct basement membrane that separates the epithelium from the underlying connective tissue stroma. Surface keratinization and a granular cell layer are uncommon. Numerous intraepithelial microcysts containing cell debris, macrophages and mucin are present (Anari & Carrie, 2010; Vorasubin et al., 2013).

The most common nasal sites involved by inverted papilloma, in order of descending prevalence, are: lateral nasal wall, ethmoid cells, maxillary sinus, and, less often, the frontal and sphenoid sinuses and nasal septum. Inverted papilloma originating medially (i.e. from the septum or turbinates) comprises 34 per cent of reported cases; that originating laterally (i.e. from the sinuses and lateral nasal wall) makes up the rest (66 per cent). Inverted papilloma of our patient originating laterally, this is accordance to the lateral nasal wall represents the most common site of origin (Anari & Carrie, 2010; Visvanathan et al., 2010).

The exact aetiology of inverted papilloma is not fully understood. However, human papilloma virus (HPV) is currently thought to be the leading cofactor in the pathogenesis of papillomas. Human papilloma virus is associated with both the inverted and exophytic types of Schneiderian papilloma, but the oncocytic type seems not to be related to HPV infection. Human papilloma virus infection occurs in benign inverted papilloma as an early event during the multistep tumourigenesis process. Other genetic insults may be required, creating a cumulative effect, in order for inverted papilloma to progress from benign (i.e. grades I and II) through dysplastic (grade III) to carcinomatous (grade IV). Chronic inflammation may be involved in the pathogenesis of inverted papilloma. Tobacco has been associated with inverted papilloma but the causal relationship is unproven. Occupational exposure to different types of smoke, dust and aerosol has also been speculated to have an effect (Anari & Carrie, 2010; Topdag, Mutlu & Ilia, 2014).
The hallmark of inverted papilloma on CT is the unilateral opacification of a contiguous nasal cavity and sinus mass. Lobulated margins are another CT indicator. The inverted papilloma is homogeneous with a soft tissue density and enhances heterogeneously with contrast. As the tumour enlarges, the adjacent bone may become thinned, bowed, eroded or sclerotic. Bone remodelling is observed most commonly at the medial wall of the maxillary sinus, followed by the lamina papyracea. The nasal septum is preserved until late in the disease course. There is a high correlation between the origin of the inverted papilloma and the presence of focal hyperostosis on the pre-operative CT, this may facilitate pre-operative prediction of tumour origin. When evaluating a CT for focal hyperostosis, two patterns of localised bone thickening are noted: (1) 'plaque-like bone thickening', seen mainly when focal hyperostosis involves the lateral wall of the nasal cavity; and (2) 'coneshaped bone thickening', seen only in the walls of the paranasal sinuses or the bony septum (Karkos et al., 2009; Anari & Carrie, 2010).

Treatment of sinonasal inverted papilloma aims to remove the disease completely at the first attempt, and also to create post-operative anatomy which allows easy post-operative endoscopic surveillance. It is very important to identify the site of attachment of the tumour pedicle in order to ensure full resection. The method of choice depend on the extent of the disease, the skill of the surgeon and the technology available. Option comprise : (1) Endonasal approach (2) Limited external approach (3) Radical external approach (4) combination endonasal and external approach. Aggressive surgery treatment is suggested by some authors, since our patient was childhood we tried to very non-aggressive method (Limaye, Mirani, Raz, 1989; Sham et al., 2009; Anari & Carrie, 2010).
CONCLUSION

We reported a case of pediatric inverted papilloma in a 10-year-old girl with a mass filling right nasal cavity as a chief complaint. She also complained about nasal obstruction. From the history, physical examination and pre-operative biopsy we diagnosed patient with pediatric inverted papilloma. We managed this patient with transnasal and transoral polypectomy with a good result.

REFERENCES


