Adult nasopharyngeal hairy polyp presenting with middle ear effusion

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Abstract
Hairy polyp is a relatively uncommon congenital mass mainly seen in naso-oropharynx. It has been also called as choristoma owing to its bimerial composition and unexpected location. It is almost always seen at birth and in the early infancy period. Adult presentation is very exceptional and only 5 adult cases have been reported as far as we know. We present a rare case of nasopharyngeal hairy polyp in a 69 year old woman admitted with hearing impairment due to middle ear effusion. We also review the relevant literature. The lesion was totally removed via combined naso-endoscopic and trans-oral approach. After 1 year of follow up there was no sign of recurrence neither in the endoscopic evaluation nor in the magnetic resonance imaging. Although nasopharyngeal hairy polyp frequently leads ear problems exclusively in the infancy period, the presented case is the 1’st adult hairy polyp case with middle ear effusion. Also it is the 2’nd oldest one so far.

Key words: Choristoma, Eustachian tube, Nasopharynx, Hairy polyp, Adult

Introduction
Hairy polyp (HP) is a congenital benign mass which was first described by Brown-Kelly in 1918 [1]. It typically consists of mature ectodermal and mesodermal elements [2]. Due to this composition and improbable location it also has been named as choristoma [3]. It is mainly seen at birth or in the infancy period and may originate from any sub-region of naso-orofarenks [4,5]. HP of infancy commonly present with respiratory distress, feeding difficulties and less frequently with middle ear effusion. Intensity of the symptoms depends on the site of the involvement and size of the lesion [5,6]. Adult presentation of HP is very rare and according to our knowledge only 5 cases has been reported so far. In adults main reported symptoms were epistaxis, nasal obstruction and dysphagia [3]. Middle ear effusion was not reported in any of the adult HP cases up to now. We present the 6th adult case in which the main symptom was hearing impairment due to middle ear effusion

Case
A 69 year old woman was referred with a history of hearing loss. She also reported that she had fullness sensation in her left ear for 2 weeks. Oral amoxicillin had prescribed by a general practitioner with the diagnosis of acute otitis media. However, her symptoms worsened and hearing loss with pain in the left ear during swallowing has begun 5-days prior to admission. When thoroughly questioned, the patient was also complained about snoring and left sided nasal obstruction. She stated that snoring had started 6 months ago and had been progressed up to the time of referral. She was otherwise healthy, with no other major medical problems. On oropharyngeal examination a pale, smooth and pedunculated mass hanging from the nasopharynx just posterosuperior to the left palatopharyngeal arch and uvula was detected (Fig. 1).

Right otoscopic examination was normal. In left otoscopic examination tympanic membrane was hyperemic with effusion in the cavum tympani. Transnasal rigid endoscopic nasopharyngeal evaluation revealed the pedunculated skin-covered mass originating from the lateral nasopharyngeal wall. Both nasal passages were otherwise normal. Pure tone audiogram confirmed a mild conductive hearing loss on the ipsilateral ear.

Tympanometry was Type-B for the left and Type-A for the right ear which was compatible with the otoscopic findings. Magnetic resonans imaging (MRI) with intravenous gadolinium diethylenetriaminepentaacetic acid (Gd-DTPA) was done. It revealed a well circumscribed mass extending from inside the left eustachian tube to the nasopharynx and oropharynx measuring 30x7x18 milimeters (Fig. 2).
Figure 1: Oropharyngeal view which reveals the hanging polypoid mass just behind the uvula and the palatopharyngeal arch

Figure 2: MRI with intravenous contrast. (a) Coronal view revealing a well-circumscribed mass (arrow) which is peripherally enhanced after intravenous contrast. (b) The same lesion (arrow) in axial view in which dilatation in the orifice of the left eustachian tube can be noticed

Surgical excision was done under endoscopic view by dissecting the peduncle from the orifice of the eustachian tube under general anesthesia. After freeing the peduncle the mass was taken out transorally. A concomitant paracentesis to the left ear was also done. There was no major bleeding.

Histopathology demonstrated a 35X15X10 mm polypoid lesion covered by stratified squamous epithelium with associated seromucinous glands and lymphoid follicles which was compatible with the diagnosis of HP.

After 1 year of follow up there was no sign of recurrence neither in the endoscopic evaluation nor in the MRI. The middle ear effusion was also totally resolved.

Discussion

Although the classification of germinal-cell originated tumors of naso-oropharynx was first done by Arnold in 1870 [7], the term of “hairy polyp” was first used by Brown-Kelly in 1918 for a benign nasopharyngeal congenital mass having both ectodermal and mesodermal components [1,2]. It was called as hairy because of its outer layer which composed of mature epidermis that frequently has a hairy appearance. In Arnold’s classification; HP had defined as “choristoma” which describes a lesion mistakenly separated from its mother tissue [3,7]. He also had described it as a type of dermoid due to its mature bigeminal composition [7].

HP is relatively rare and has an incidence of 1 in 40,000 live births and have a tendency to occur in female newborns [8,9]. Due to this presentation, HP is typically defined as a disease of early infancy and is very exceptional after the first year of life [10]. Only 5 adult cases have been reported up to now, the oldest being 71 years [4,11]. We believe that our case is the 6th adult HP and also the 2nd oldest one so far.

HP of naso-oropharynx mostly originates from lateral nasal wall followed by the tonsils, palatal arches and soft palate [4,5]. Almost two-thirds of lateral pharyngeal wall HPs originate from the eustachian tube [3]. In our case it was also originated from the eustachian tube resulting with middle ear effusion. HP of infancy usually presents with feeding difficulties, drooling, respiratory distress, hemoptysis, coughing, otitis, hearing loss, vomiting and recurrent ear infections [5] while in adults it commonly presents with snoring, recurrent epistaxis, dysphagia and cough [3]. In our case the main symptoms were hearing loss and otalgia which were presumably caused by middle ear effusion. Considering the adult cases, middle ear involvement has not been reported before. In the literature the adult HPs were presented mainly with symptoms associated to nasal obstruction and swallowing difficulties [3].

Differential diagnosis of HP; including teratoma, hamartoma and dermoid cyst can sometimes be challenging due to similar histopathological findings. Teratomas can be differentiated by trigeminal origin and the observation of endodermal derivatives while hamartomas can be identified by the presence of single germ cell layer [11]. When regarding the dermoid cysts; they have the typical keratin flakes. In this presented case none of these above mentioned findings were seen, instead there was a bigeminal structure consisting of ectodermal and mesodermal components with an epidermal lining which was compatible with HP. The mesodermal inner core which mainly composed of fat can also be noticed in the MRI (Fig. 2). Whereas teratomas exhibit more blended appearance of germ cell layers and may sometimes have bone or teeth fragments which may be hyperdense in MRI unlike in our case.

Main treatment modality for HP is total surgical excision. Malignant potential, metastasis or recurrence after complete removal has not been reported so far [8]. Although it can be removed trans-nasally or trans-orally; in our opinion the best approach is combined naso-endoscopic and trans-oral approach which provides better visualization and control [12]. Thus, we used the combined endoscopic approach in which we first excise the peduncle from the eustachian tube under
endoscopic view followed by removal of the mass transorally.

Naso-oropharyngeal HPs typically seen in female neonates with left sided predominancy while they are extremely rare in adult population [10,13]. We present the 6th case of adult nasopharyngeal HP with an uncommon presentation. We achieved total removal of the lesion by a combined naso-endoscopic and trans-oral approach. We believe that endoscopic guidance is essential for total removal to prevent recurrence in adult HPs

**Conflict of Interest:** The authors declare no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

**References**