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Case Report

Naso-oropharyngeal hairy polyps in two age groups: two case reports

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Abstract: *Objectives:* Hairy polyps are rare, with an incidence of 1 in 40,000 live births. They are almost exclusively observed in neonates and infants and are extremely rare in adults. We report two cases of naso-oropharyngeal hairy polyps in two age groups (adult and neonate) and described the differences in the clinicopathological features of the polyps between the two age groups. *Methods: Case 1* A 62-year-old man with a hairy polyp occupying the orifice of the right eustachian tube who presented with a 1-week history of right aural fullness and hearing impairment. *Case 2* A 1-month-old girl with a hairy polyp originating from the upper pole of the right tonsillar fossa who presented with intermittent cyanosis since birth, particularly during feeding in a supine position. *Results:* Both patients recovered satisfactorily after the surgery and were disease-free at the most recent follow-up. The differences in the clinicopathological features between the two age groups (adult and neonate) with naso-oropharyngeal hairy polyps were discussed herein. *Conclusions:* Our study made two pertinent contributions. First, we reported the seventh case of naso-oropharyngeal hairy polyp in adults in English-language literature in the past three decades. Second, our study highlights differences in the clinicopathological features of naso-oropharyngeal hairy polyps based on the age of presentation.

Keywords: Hairy polyp, choristoma

Introduction

Hairy polyps, first described by Brown Kelly in 1918, are rare, with an incidence of 1 in 40,000 live births [1]. They are almost exclusively observed in neonates and infants and are extremely rare in adults [2]. These polyps are mature congenital ectodermal and mesodermal tissue-aggregates that are often covered with thin hair, presenting as polypoid masses in the naso-oropharyngeal region commonly at or shortly after birth [3, 4]. The mesenchymal components often include fat and cartilage, whereas the outer ectodermal layer consists of a keratinizing squamous epithelial layer with adnexal structures, such as hair follicles-hence the term *hairy* polyp (choristoma) [5]. Hairy polyps can arise from the nasopharynx or oropharynx; however, they can also arise from numerous surrounding structures, including the hard palate, middle ear cavity, tongue, and lower lip [4]. However, their etiology is relatively less known, and whether they are developmental malformations or primitive teratomas is debatable [3]. In this study, we report two cases of naso-oropharyngeal hairy polyps in two age groups (adult and neonate) and described differences in the clinicopathological features between the two age groups.

Case report

Case 1

A 62-year-old man presented at our hospital with a 1-week history of right aural fullness and hearing impairment. A nasopharyngoscopic examination revealed a mass occupying the orifice of the right eustachian tube (**Figure 1A**). It was firm and hairy, and it did not bleed on touch. The mass was subsequently excised under general anesthesia by using a combination of endoscopic and transoral approaches. During surgery, the pedunculated mass was observed to arise from the right eustachian tube (**Figure 1B**). Histopathology revealed a pol-

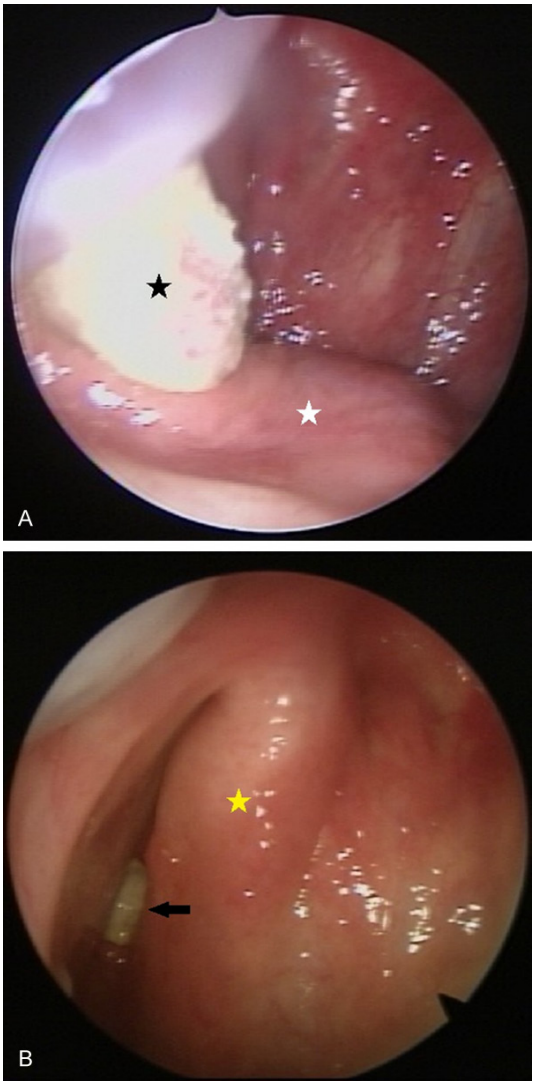

Figure 1. Case 1. A. On nasopharyngoscopic examination, a firm and hairy mass (black asterisk) occupying the orifice of the right eustachian tube was observed. (white asterisk: soft palate) B. Nasopharyngoscopy revealed a mass (arrow) arising from the right eustachian tube (yellow asterisk: posterior tubal cushion).

ypoid lesion covered with keratinizing squamous epithelium and adnexal structures, including hair follicles and sebaceous glands. The features indicated a hairy polyp (**Figure 3A**). The patient recovered satisfactorily after the surgery and was disease-free at the most recent follow-up, 30 months after surgery.

Case 2

A 1-month-old girl presented at our hospital with intermittent cyanosis since birth, particularly during feeding in a supine position. These episodes were managed by changing her position to a prone and upright position. A physical examination revealed the presence of a pedunculated mass originating from the upper pole of the right tonsillar fossa (**Figure 2A**). The mass was mobile, and it moved between the oropharynx and oral cavity during swallowing. No other abnormality was observed. Computed tomography revealed a hypodense tubular mass, approximately 3.5 cm long, in the right oropharynx (**Figure 2B**). The mass was subsequently excised under general anesthesia via a transoral approach. Grossly, the proximal end of the mass was covered with mucosal epithelium, whereas the distal end was covered with hairy skin (**Figure 2C**). Histopathological analysis revealed that the oral mucosa harbored a polypoid lesion covered with keratinizing squamous epithelium and subcutis with underlying hyaline cartilage and a central core of fibrovascular adipose stroma. The features suggested a hairy polyp (**Figure 3B**). The patient recovered satisfactorily after the surgery and was disease-free at the most recent follow-up, 32 months after surgery.

Discussion

Hairy polyps are almost exclusively observed in neonates and infants and are extremely rare in adults. Only seven cases of naso-oropharyngeal hairy polyps in adults have been reported in the English-language literature in the past three decades [2]. A female preponderance of 3.5 times has been observed in neonates and infants with hairy polyps [3]. However, in adults with naso-oropharyngeal hairy polyps, a nearly equal sex-distribution has been observed [2].

Hairy polyps cause various symptoms depending on their size and location. Naso-oropharyngeal hairy polyps are increasingly recognized as a major, often-missed cause of respiratory distress and feeding difficulty in neonates and infants. Such hairy polyps in neonates can result in life-threatening asphyxia [3, 4]. Unlike in neonates, the presentation in adults is less obvious and less intense [2].

The pathogenesis of hairy polyps remains debatable. Hairy polyps in neonates are considered as developmental malformations associated with the development of the first and second pharyngeal arches owing to their endoscopically documented anatomical associa-

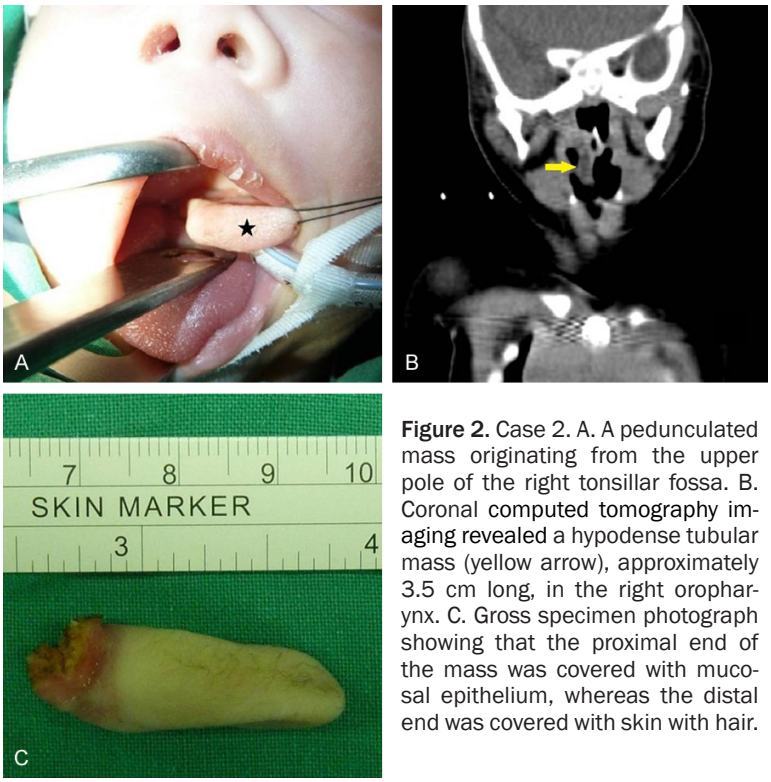

Figure 2. Case 2. A. A pedunculated mass originating from the upper pole of the right tonsillar fossa. B. Coronal computed tomography imaging revealed a hypodense tubular mass (yellow arrow), approximately 3.5 cm long, in the right oropharynx. C. Gross specimen photograph showing that the proximal end of the mass was covered with mucosal epithelium, whereas the distal end was covered with skin with hair.

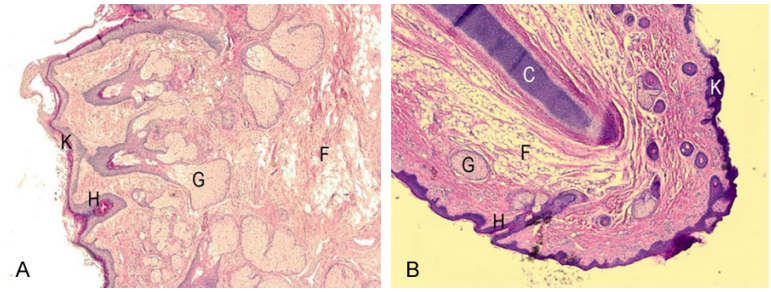

Figure 3. A. Histopathological section of hairy polyp of patient 1 revealing a polypoid lesion, which was covered with keratinizing squamous epithelium and adnexal structures (hematoxylin and eosin stained [H&E] 40×). B. Histopathological section of hairy polyp of patient 2 revealed a polypoid lesion, which was covered with keratinizing squamous epithelium and subcutis over underlying hyaline cartilage and a central core of fibrovascular adipose stroma (H&E 40×). (K: keratinizing squamous epithelium, H: hair follicle, G: sebaceous gland, C: hyaline cartilage, F: fibrovascular adipose stroma).

tions with the eustachian tube and tonsillar pillars [3]. Hairy polyps in neonates are often associated with congenital malformations, including cleft palate, uvula, and external ear agenesis, ankyloglossia, choanal atresia, facial hemihypertrophy, left carotid artery atresia, osteopetrosis, persistent nasopharyngeal membrane, and first and second branchial arch malformations [4]. Hairy polyps are not associated with congenital syndromes nor do people exhibit genetic predisposition to these polyps [6].

However, the occurrence of hairy polyps in the later stages of the lives of previously asymptomatic adults remains unexplained. Moreover, no congenital or developmental anomaly has ever been associated with adults. These choristomatous lesions could occur because of delayed pluripotent cell morphogenesis, where stem cells have either escaped local governing influences because of some inciting factors (trauma) or have been misdirected or trapped on their way to their predestined target (missed target hypothesis). Focal neoplasia could be an alternative explanation. Hairy polyps in adults appear to represent focal neoplastic proliferations or benign teratomas rather than merely developmental aberrations, as widely believed [3].

In neonates and infants, radiological imaging is particularly useful for planning the surgical removal of hairy polyps that are otherwise obscured because of the endotracheal tube as well as for evaluating associated otolaryngological complications. However, radiation exposure is a concern. Hairy polyps exhibit characteristic radiological imaging features, including the presence of fat in the polypoid mass. In general, the differential diagnosis for fat-containing masses in the nasospharnx is limited to teratomas and hamartomas, which tend to be heterogeneous, and lipomas and their variants, such as chondrolipomas or fibrolipomas, which are extremely infrequent in the nasopharynx and eustachian tube and tend to occur in adults rather than in neonates [5].

Despite the theoretical and speculated differences in origin, the management and prognosis of hairy polyps in adults do not differ significantly from those in neonates. Excision results

Table 1. Differences in the clinicopathological features of naso-oropharyngeal hairy polyps between two age groups

	Adults	Neonates
Incidence	Seven cases in the past three decades have been reported	1 in 40,000
Sex-distribution	Equal	3.5-times female preponderance
Symptom	Less obvious and less intense	Respiratory distress and feeding difficulty
		Life-threatening asphyxia
Congenital malformation	Not associated with congenital malformation	Often associated with congenital malformations
Pathogenesis	Missed target hypothesis	Developmental malformation
	Focal neoplasia hypothesis	

in complete and uneventful recovery [2, 4]. No case of malignant transformation has yet been reported [1, 6]. A case of recurrent hairy polyp-over a period of 6 years-due to incomplete excision has been reported, indicating the slow-growing nature of these polyps [7]. However, the confined space of the eustachian tube and proximity to the internal carotid artery can pose an operative challenge, which can be addressed by performing preoperative imaging as well as staging [5]. The differences in the clinicopathological features of naso-oropharyngeal hairy polyps between the two age groups (adult and neonate) are summarized in **Table 1**.

In conclusion, our study makes two pertinent contributions. First, we report the seventh case of naso-oropharyngeal hairy polyps in an adult in the English-language literature in the past three decades. Second, our study highlights important differences in the clinicopathological features of naso-oropharyngeal hairy polyps according to the age of presentation.

Disclosure of conflict of interest

None.

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