Hairy Polyp, a Clinicopathologic Study of Four Cases

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Abstract Hairy polyps (HPs), dermoids or teratoid tumors are rare tumors of naso-oropharyngeal region which commonly present at or shortly after birth. The etiology and classification of these tumors is still debatable and categorized by different authors differently. HPs have female predominance and usually present with respiratory and feeding problems. Microscopically, the polyp is covered by skin with underlying mesenchymal core. The aim of this study is to describe the clinicopathological features of Hairy polyps on a cohort of cases. We reviewed the surgical pathology database of our institution for last 10 years and retrieved four cases of hairy polyps. The age of patients ranged from 1 month to 18 years (mean = 12 years), with a female to male ratio of 1:3. Two of our cases presented at birth and two cases in late teens. Two of the HPs were located in nasopharynx, one on soft and hard palate and one on lower lip. One case was associated with bifurcation of tongue. Size of the polyps ranged from 2.3 to 4.5 cm (mean = 3 cm). Histologically, all HPs were lined by skin and the underlying core consisted of adnexal structures, adipocytes, skeletal muscle, smooth muscle and seromucinous glands. Lymphoid aggregates, cartilage and bone were seen in one case each. Our series highlights the diverse nature of this entity in terms of age of presentation and location. HP at lower lip and associated bifurcation of tongue has not been previously reported. We observed a male predominance in contrast to the published literature. However, number of cases is too few to read a definite conclusion on this point. The etiology is still controversial and includes congenital malformation and activation of pluripotent stem cells.

Keywords Hairy polyp · Dermoids · Male predominance · Bifurcation of tongue · Lower lip

Introduction

Hairy polyps (HP) are benign tumors of neonates and infants [1, 2]. HP can arise from any of the naso-oropharyngeal structures (including tonsils, eustachean tube and tongue) as a skin covered polypoidal mass [2–11]. Owing to its bi-germinal layer origin, it is characterized by skin (ectoderm) covering a (mesodermal) core of adipocytes, smooth muscle and cartilage [12]. Depending upon the size and location, these lesions may be asymptomatic or present with respiratory distress, difficulty in feeding, etc. [4–11]. Almost all patients recover uneventfully after surgical excision [1].

Materials and Methods

We retrieved four cases of HPs from the surgical pathology database of Aga Khan University Hospital reported between January 2002 and November 2012 through “Integrated Laboratory Management System (ILMS)” software. Since this was a retrospective study and did not involve actual patients, approval from the Hospital Ethics Committee was not sought. Clinical information regarding age, sex, location and the presenting complaints was obtained from the pathology reports. Microscopic glass slides were reviewed by two pathologists and were analyzed for presence of keratinized stratified squamous lining epithelium of polyp and presence of different components in the polyp core. We tried to get the follow up of patients through telephone numbers provided on their requisition slips but unfortunately none could be contacted.

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Results

The ages ranged from 1 month to 18 years. Two of the patients presented in their late teens, i.e. 17 and 18 years. Out of the two patients presenting at birth, one was operated at the age of 12 years. Male to female ratio was 3:1. Two polyps were located in nasopharynx, one on soft and hard palate and one on lower lip. Only one case was associated with other congenital malformation. The size of the polyps ranged from 2.3 cm to 4.5 cm. Grossly, the polyps were covered by tan brown skin bearing few hair shafts. Microscopically, all the polyps were covered with keratinized stratified squamous epithelium (Fig. 1a). Adnexal structures were seen in all cases; adipose tissue in three; skeletal muscle, smooth muscle and seromucinous glands in two cases (Fig. 1b). Lymphoid tissue, cartilage and bone formation was seen in one case each (Fig. 2a, b and Table 1).

Discussion

Hairy polyps (HP) are rare but the most common congenital, benign naso-opharyngeal tumors [1] which usually present at birth or in early infancy and are quite uncommon after first year of life [1, 2]. So far 180 cases have been reported in literature. Yilmaz et al. [2] have described 5 cases with age more than 1 year, the oldest being 58 years. Out of 4 cases reported at our institution, 2 cases presented in late teens. Out of 2 cases presenting at birth, one was operated in early teens while the other was operated at the age of 1 month. HPs are six times more common in females, [3] while 3 of our cases were males, which is in contrast to the findings of other series in which a female predominance was noted [2, 3].

HPs present as skin covered polypoidal masses which can arise from nasopharynx or oropharynx but are also

reported to arise from a number of surrounding structures including soft palate [2, 4, 13], hard palate [5], tonsils [6–8], eustacheantube [9], middle ear cavity [10] and tongue [11]. The polyps in our series show diverse distribution, two of them were located in nasopharynx, one in soft and hard palate and one in lower lip. HPs cause diverse symptoms depending upon their size and location. Large polyps cause obstruction and commonly result in respiratory distress and feeding difficulties. Other symptoms include hearing loss, otorrhea, vomiting, earache, bleeding, snoring and recurrent ear infections [4–11].

The pathogenesis of this entity is still debatable. HPs are usually isolated but can be seen in association with a number of congenital malformations including cleft palate, uvula and external ear agenesis, ankyloglossia, choanal atresia, facial hemihypertrophy, left carotid artery atresia, osteopetrosis [1, 6, 14], persistent nasopharyngeal membrane, 1st & 2nd branchial arch malformations, etc. and are therefore, considered as a developmental abnormality [6, 13, 15]. One of the cases in our series was associated with congenital abnormality i.e. bifurcation of tongue, which has not been reported in literature. Most of the authors categorize HP as teratomas, being composed of mature components derived from two germ layers i.e. skin, neural tissue from ectoderm and adipose tissue, muscle and cartilage from mesoderm. These germ layers are thought to arise from pluripotent stem cells in this region which escape normal regulators in embryonic life and therefore present at or soon after birth [12, 16]. HPs are also termed as dermoids due to their characteristic and consistent outermost skin component and variable mesodermal component which might be inconspicuous in some cases. Some authors designate HP as choristoma due to the presence of normal tissue in aberrant location [17, 18]. The term “teratoid tumors” used for these lesions, is a misnomer as teratoid is defined as bi-trigeminal lesion with less differentiated components, which is not observed in hairy

Fig. 1 **a** Low power examination shows a polypoid lesion covered by skin and underlying adnexae. The core is composed of adipose tissue and skeletal muscle fibers (H&E, $\times 40$ magnification). **b** Transition of

respiratory mucosa to stratified squamous mucosa. Subepithelial seromucinous glands are seen (H&E, $\times 100$ magnification)

Fig. 2 Skin with underlying proliferation of pilosebaceous glands and hyaline cartilage (H&E, ×40 magnification). **b** Mesenchymal derivatives including bone, skeletal muscle and adipose tissue (H&E, ×40 magnification)

Table 1 Summary of gross and microscopic features of Hairy polyps (n = 4)

No.	Age	Sex	Location	Size (cm)	Associated Malformation	Components of polyp
1.	17 years	Female	Nasopharynx	2.5	None	Skin adnexae, adipose tissue, skeletal and smooth muscle, seromucinous glands and lymphoid tissue.
2.	18 years	Male	Nasopharynx	3	None	Skin adnexae, adipose tissue, skeletal and smooth muscle, and seromucinous glands.
3.	12 years	Male	Soft and hard palate	4.5	Bifurcation of tongue	Skin adnexae and seromucinous glands.
4.	1 month	Male	Lower lip	2.3	None	Skin adnexae, adipose tissue, cartilage and mature bone.

polyps [15]. In our opinion, these lesions represent developmental abnormalities and malformations which arise during fetal period. The view is supported by the fact that these lesions commonly present shortly after birth and the associated malformations usually represent failure of closure of various plates in naso-oropharyngeal region during organogenesis. Furthermore, like the structures in naso-oropharynx, HPs are also derived from ectoderm and mesoderm. Lack of any endodermal derivative in these lesions negates the possibility of their origin from the pluripotent stem cells of this region.

The differential diagnosis is mainly based upon location as well as age and includes choanal atresia, meningocele, encephalocele, glioma, craniopharyngioma, rhabdomyosarcoma, neuroblastoma and Rakhe's cleft cyst [19, 20]. Microscopically, these differential diagnosis do not pose significant challenge except for closely related entities such as teratomas, hamartoma and dermoid cysts. Teratomas are ruled out by the presence of endodermal derivatives. Hamartomas are ruled out by the presence of single germ layer (usually mesodermal) derivative and dermoid cysts by presence of keratin flakes. MRI is most helpful in locating and characterizing these tumors on the basis of presence fat in the mesenchymal cores [21]. Excision results in complete and uneventful recovery. So far, two cases with recurrence have been reported. Metastasis has so far not been reported [1, 10].

Conclusion

Our series highlights the diverse nature of this entity in terms of age of presentation and location. HP at lower lip and associated bifurcation of tongue has not been previously reported. We observed a male predominance in contrast to the published literature. However, number of cases is too few to read a definite conclusion on this point. The etiology is still controversial and includes congenital malformations and activation of pluripotent stem cells.

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