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'Hairy polyp' of the pharynx in association with an ipsilateral branchial sinus: evidence that the 'hairy polyp' is a second branchial arch malformation

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Abstract
The pharyngeal 'hairy polyp', or 'dermoid' has caused considerable debate as to its origin since the original classification proposed by Arnold in 1870.[1] This classification implies that the hairy polyp is either a teratoma or sequestration dermoid cyst. Many papers contest this view, in favour of a developmental malformation. We describe the first case of a hairy polyp in association with an ipsilateral branchial sinus as further supportive evidence of a developmental malformation of the second branchial arch.

Key words: Branchial Region; Choristoma; Nasopharynx

Introduction

The hairy polyp, originally classified by Arnold in 1870[1], is typically a pear or sausage-shaped, grey/white, pedunculated mass arising from the oro-nasopharynx, ranging from 0.5 to 6 cm in length. It is the commonest congenital nasopharyngeal mass with 137 cases reported.[2,3] Histologically, the surface is composed of keratinizing stratified squamous epithelium with normal skin appendages (hairs, sebaceous and sweat glands). Beneath the dermis the mass is composed of fibro-adipose tissue and may contain elements of cartilage and muscle.

They usually present shortly after birth, or in the first year of life, with feeding or respiratory difficulties. If small they may be found incidentally in adults.

There has been considerable debate and confusion regarding the origin of these lesions. They are most commonly classified with teratomas or dermoid cysts. The term hamartoma has also been used to describe hairy polyps, but is incorrect because its hairy skin is not native to the area where the polyp arises.[4] True hamartomas of the tonsil have been described.[5–7] as have true hamartomas of the nasopharynx.[8] Gundrum[9] proposed using the term 'choristoma', which means a mass of histologically normal tissue in an abnormal location. Although appropriate, and a term which has proved popular, this is purely descriptive, and gives no indication of aetiology.

Case report

An otherwise normal, term, female child was referred with a five-day history of nasal regurgitation and a tendency to gag while feeding. When the child vomited the father noticed a fleshy mass presenting in the mouth. Apnoeic episodes occurred when the infant lay in the supine or prone positions, the best position for breathing being the right lateral. No stridor was present and the cry was normal. Further examination revealed a sinus at the anterior border of the left sternomastoid, at the junction

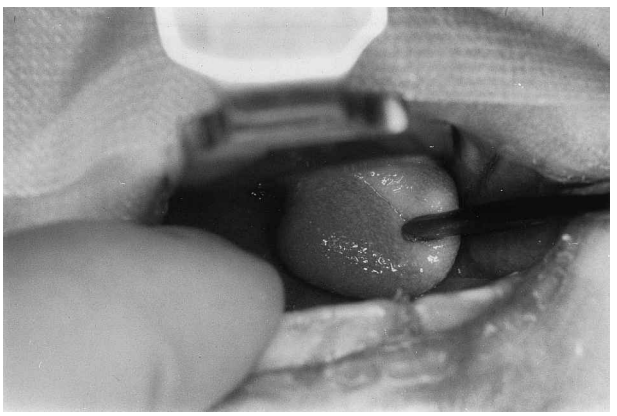

Fig. 1
The polpy as seen in the oropharynx prior to removal.

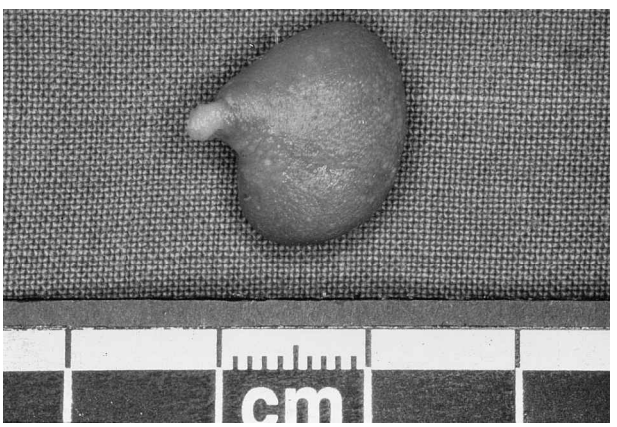

Fig. 2
The keratinized, hair-bearing polyp excised at its thin base.

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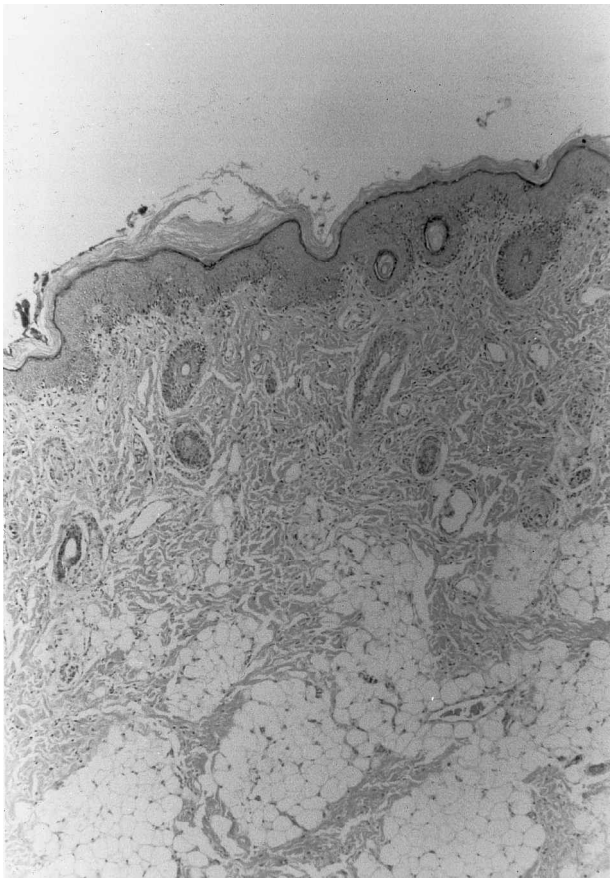

FIG. 3

Polyp showing stratified squamous keratinizing epithelium, skin appendages, and a fibro-fatty core (H & E; ×40)

of the upper two thirds and lower third. A tract was palpable, disappearing beneath the anterior border of the muscle. The nasasl breath test was normal. Ear examination was normal. A cleft palate was excluded, but no obvious intraoral mass could be visualized.

The child underwent an examination under anaesthetic in the supine position. A grey/white mass was seen within the nasopharynx, which proved to be mobile, pedunculated, and arising from the upper pole of the left tonsil (Figure 1). The lesion was removed at its base (Figure 2).

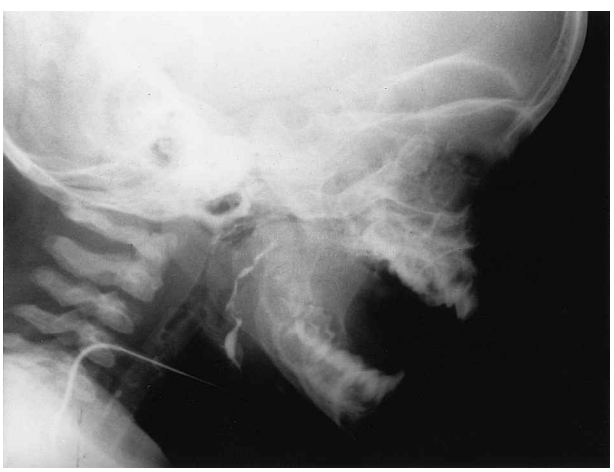

FIG. 4

Lateral view of sinogram. The tract can be seen extending to the upper pole of the tonsil.

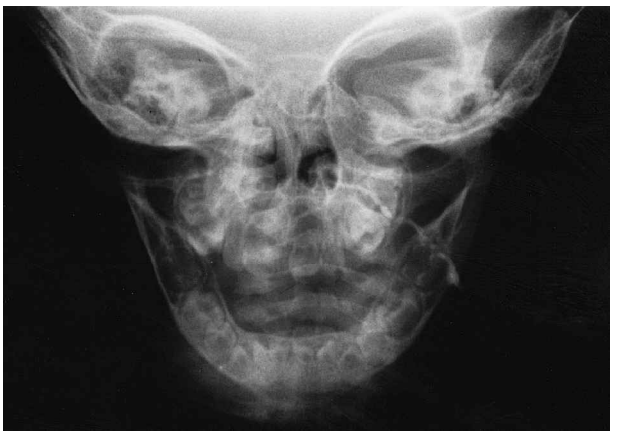

FIG. 5

A-P view of sinogram showing the left-sided tract.

Histological examination found the polyp to be 1 × 1.5 cm, composed of a fibro-fatty tissue core, covered by keratinizng squamous eithelium with normal skin appendages. These features are of the 'so-called' hairy polyp. The fatty core, stratified squamous epithelium, keratin, sebaceous glands and hair follicles are demonstrated in Figure 3.

The child made an uneventful recovery, with complete resolution of all symptoms, and was discharged home. Two months later the sinus tract became the source of recurrent infection. A sinogram was performed to demonstrate its extent (Figures 4 and 5), where it can be seen ascending as far as the upper pole of the left tonsil – the site where the hairy polyp was found.

Discussion

Arnold first described the hairy polyp in 1870.[1] He described four categories of mass arising from the nasopharynx, namely epignathi, teratomas, teratoids and 'dermoids' (or hairy polyps). Implications of this classification have been, firstly, that 'hairy polyps' are the lowest order of teratoma, and secondly (because of Arnold's use of the word 'dermoid' to describe their covering of skin) that hairy polyps are 'dermoid cysts'.

Teratomas, teratoids and epignathi

Epignathi are extremely rare, and probably represent a large, well-differentiated teratoma. They have also been considered to represent a maldeveloped twin, parasitic foetus, or foetus-in-foetu. They are too rare for any definite assessment of their origin.

The distinction between teratoids and teratomas is somewhat artificial. Willis[10] proposed that these two groups should be considered as one. Both are considered neoplastic (and may be either benign or malignant) because they show progressive and uncoordinated growth. They are thought to arise from totipotential cells. These cells are only present in adults in the gonads. Half of all teratomas are adult gonadal teratomas. The early embryo develops from totipotential cells, which can give rise to congenital teratomas (the remaining half) presenting in infancy at various sites, the commonest being sacrococcygeal (40 per cent), ovary (37 per cent) and head and neck (six per cent).[11]

Teratomatous lesions (teratomas and teratoids) typically contain elements from all three germ layers, although in recent decades this definition has become less stringent with the acceptance of examples that are composed of only bi-germal elements. On section, they consist of solid and

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cystic areas, with cysts lined by epithelium and containing epithelial products (squames, sebum and hair, sometimes teeth), and solid areas containing any mesoderm-derived tissue (e.g. muscle, bone). Teratomas have an organized structure, containing recognizable organs or parts of organs. Teratoids are less well differentiated, lacking this organized pattern, tend to be larger, and are more likely to be malignant.

Hairy polyps are composed of skin (ectodermal origin), and subcutaneous tissue, often with cartilage and muscle (mesodermal origin). There is a case, therefore, to consider the hairy polyp as a limited form of teratoma, and indeed many authors take this view.[11–14]

Dermoid cysts

Dermoid cysts are derived from a single germ layer, and are cysts, containing desquamated epithelial products. They may be congenital (sequestration dermoid cysts), due to incomplete breakdown of epithelium at embryological fusion lines, or acquired (implantation dermoid cysts) due to traumatic implantation of epithelium, most commonly on the lower lip and digits of the hand.

A few case reports describe the hairy polyp as a dermoid cyst. Although Arnold did not refer to the 'hairy polyp' or 'dermoid' as a dermoid cyst, some authors have taken this to be the case considering them to be sequestration dermoid cysts between the first and second branchial arch, i.e. first pharyngeal pouch,[15] or during fusion of the palatine processes,[16] to account for their location.

Hairy polyps

The ectodermal element of hairy polyps is confined entirely to its covering of hairy skin, i.e. on its surface. There are no ectodermal inclusion cysts within the mesoderm. This differentiates them from both teratomas and dermoid cysts.

In determining the site incidence of head and neck teratomas Tapper[11] included the hairy polyp, describing the oro-nasopharynx as a common site; the commonest being the anterior or lateral neck, followed by the face, oro-nasopharynx, and orbit. Hairy polyps are by far the most common congenital mass of the oro-nasopharynx, and only occur at this site. If you exclude hairy polyps then true teratomatous lesions of the oro-pharynx are extremely rare.

The sex incidence of these two lesions also differs. Teratomas have equal sex incidence,[12] whereas the hairy polyp affects females six times more commonly than males. Hairy polyps have not been associated with skull base defects, have no malignant potential, do not show progressive growth (they may be found incidentally on examination in adults), and do not recur after excision. All these features are seen commonly with teratomas.

Hairy polyps are usually single, but three cases of bilateral polyps have been reported.[4,17,18] As one of these authors points out, the bilateralism seen in these cases supports the theory of a developmental malformation rather than a teratomatous origin.[4]

With regard to dermoid cysts, true sequestration dermoid cysts of the head and neck do occur at head and neck fusion lines (e.g. the root of the nose, or submental region in the midline). Although they could occur in the nasopharynx they have not been described. The only descriptions of dermoid lesions here are those of the hairy polyp.

Congenital malformations

Another explanation for the aetiology of the hairy polyp is persistence of part of the nasopharyngeal membrane.[19] Certainly, polyps are located at the inferior limit of this membrane, but they are not found around the superior limit of the posterior choana, and as yet, have not been described in association with choanal atresia, which one might expect.

The largest series of hairy polpys is that of Brown-Kelly[20] in 1918, who reported his own case and reviewed a further 49 from the English, French and German literature. He found that 10 per cent of these patients had a cleft of the hard or soft palate. Haddard[21] reported a further two cases associated with cleft palate in 1990. These clefts conceivably occur secondary to the polyp intervening between the palatal shelves, preventing their closure.

Equally as common in Brown-Kelly's[20] review were associated malformations of the first and second branchial arches. These included absence of the external ear, microtia, low set ears, and hemifacial microsomia. The association of external malformations with a polyp in the pharynx is more difficult to explain. These are the only other commonly associated anomalies, which suggests the hairy polpy may itself be a malformation of the same arches. In fact, hairy polyps arise on the pharyngeal surface of the first and second branchial arches (surrounding or within the eustachian tube, or between it and the upper pole of the tonsil). Our case adds a new second arch abnormality to this list, in support of this theory.

Heffner *et al.*[22] presented further evidence in 1996. They reviewed the pathology of eight hairy polyps and four pharyngeal teratomas available at their institute. They noted a striking gross and histological similarity between the hairy polyp and foetal auricles (which normally develop from the first and second arches). Polyps containing cartilage (most of their series) had the cartilage arranged as an elongated or curved plate of uniform thickness, the same thickness as that of the infantile pinna, many of which contained large amounts of elastin. This arrangement was not found in the teratomas containing cartilage. External cutaneous accessory auricles arise in front of the tragus to below the ear, as far as the level of the hyoid bone, representing the ectodermal surface of the first and second arches. These accessory auricles would never be considered teratomas, but they are histologically identical to the hairy polpy, which arises on the pharyngeal surface of the same arches. Heffner proposed that hairy polyps were displacements of first or second arch tissue with 'auricular hillock' potential, and that these lesions represented 'pharyngeal accessory auricles'.

Conclusion

Teratomas, teratoids, and probably epignathi, represent similar lesions with different levels of differentiation. Hairy polyps were included in Arnold's original classification, which described nasopharyngeal masses, because they are common at this site. Many case reports have appeared in the literature since, distinguishing the hairy polyp as a separate entity. Furthermore, the use of the word dermoid is misleading, and should be dropped in favour of the more descriptive and individual term, hairy polyp.

The strongest case for the aetiology of the hairy polyp is a developmental abnormality, and the commonest associated malformations are those relating to the first and second branchial arches. We add one further case report to the literature, associated with another, not previously described, second arch abnormality.

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