Acta Otorrinolaringol Esp. 2016;67(2):e9–e12

Acta Otorrinolaringológica Española

www.elsevier.es/otorrino

CASE STUDY

Naso-Oropharyngeal Choristoma (Hairy Polyps) in Adults: A New Case, and Review of the Literature☆,☆☆

Coristomas naso-orofaríngeos (pólipos pilosos) en adultos: un nuevo caso y revisión de la literatura

Mainak Dutta,[a,*] Soumya Ghatak,[b] Soham Roy,[c] Ramanuj Sinha[a]

[a] *Department of Otorhinolaryngology and Head-Neck Surgery, Medical College and Hospital, Kolkata, 88, College Street, Kolkata 700073, West Bengal, India*
[b] *Department of Otorhinolaryngology and Head-Neck Surgery, R. G. Kar Medical College and Hospital, Kolkata 700004, West Bengal, India*
[c] *Department of Otorhinolaryngology-Head and Neck Surgery, Children's Memorial Hermann Hospital, University of Texas Medical School at Houston, Houston, TX, USA*

Received 7 August 2014; accepted 9 September 2014

Clinical Case

A 42-year-old woman presented with progressive difficulty in swallowing, and snoring since 4 years, with right-sided nasal obstruction for 2 years. She had a persistent sense of obstruction behind her tongue; on examination, a large, smooth-walled globular mass was seen occupying the oropharynx extending up to the tongue-base and vallecula (Fig. 1). It was firm, non-tender, did not bleed on touch, and was free on all sides except superiorly, clinically resembling a choanal polyp. On diagnostic naso-endoscopy, the pedunculated mass was seen attached to the right Eustachian tube orifice and the adjacent epipharyngeal surface of soft palate, hanging from nasopharynx into the oropharynx obstructing the aerodigestive tract. Computed tomography (CT)-scan was non-contributory; the paranasal sinuses were clear, with no breach in the bones and skull-base. The lesion was excised under general anesthesia by combined naso-endoscopic/trans-oral approach. Grossly, the pear-shaped mass measured about 5 cm × 3.5 cm × 3 cm, was firm, bosselated, and appeared heterogeneous in consistency. Histopathology revealed stratified squamous epithelium with epidermal appendages (hair follicles, sebaceous glands) along with well-organized, mature cartilage nests and smooth-muscle fibers with a fibro-adipose core, without any evidence of dysplasia (Fig. 2a and b). The features suggested bigerminal choristoma (hairy polyp) of the naso-oropharynx. The symptoms of the patient ameliorated

☆ Please cite this article as: Dutta M, Ghatak S, Roy S, Sinha R. Naso-orofaríngea coristomas (Hairy pólipos) en adultos: un nuevo caso y revisión de la literatura. Acta Otorrinolaringol Esp. 2016;67:e9–e12.

☆☆ This paper has been presented as a Poster in the Annual Meetings of The Triological Society (TRIO) at the 116th Combined Otolaryngology Spring Meetings (COSM) on 10th–14th April 2013, held at Orlando, Florida, USA. Links: http://www.researchposters.com/Posters/COSM/COSM2013/2-224.pdf and http://www.triomeetingposters.org/wp-content/uploads/2013/04/2-224.pdf.

* Corresponding author.
E-mail address: duttamainak@yahoo.com (M. Dutta).

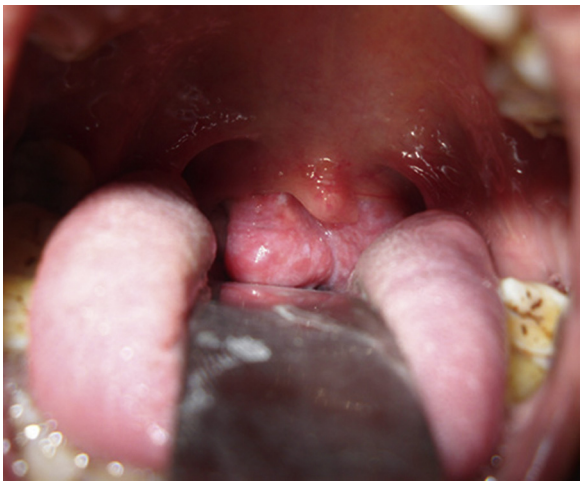

Figure 1 A large fleshy mass could be seen in the oropharynx of this 42-year-old woman. It is free on all sides except superior, and seems to originate from the nasopharynx.

following excision of the mass; she recuperated well and was disease-free on 2-year follow-up.

Discussion

Hairy polyps are mature congenital ectodermal and mesodermal tissue-aggregates, often covered with thin hair, presenting as polypoid masses at anatomically aberrant sites (choristoma).[1] They are almost exclusively seen in neonates and infants and can result in life-threatening asphyxia. Hairy polyps, however, are exceedingly rare in adults[1]; a PubMed/MEDLINE® search with the keywords ''hairy polyp'', ''choristoma'', ''adult'' and ''naso-oropharynx'' revealed only 6 such cases in the last 3 decades (Table 1).[2–6] Unlike in neonates, the presentation in adults is insidious and less dramatic, clinically apparent even beyond the fifth decade in some cases,[2–4] rising questions whether they should as a rule be considered as developmental malformations.

Hairy polyps in neonates are often associated with congenital disorders (like branchial arch anomalies)[1,6]; they are linked with development of the first and second pharyngeal arches owing to their endoscopy-documented anatomic associations with the Eustachian tube and tonsillar pillars.[1] However, their occurrence late in the lives of previously asymptomatic adults remains unexplained.[1] Also, no congenital/developmental anomaly has ever been associated with adults.[1] These choristomatous lesions could occur due to delayed pluripotent cell morphogenesis where stem-cells have either escaped the local governing influences due to some inciting factors (trauma), or have been misdirected or trapped on way to their pre-destined target (''missed target hypothesis'').[1] However, focal neoplasia could be an alternative explanation.[1] We have carried out an analysis on the origin of hairy polyps based on an extensive literature review.[1] Though the accumulated data there was based on clinical facets irrespective of age-groups and did not delve in-depth in the molecular aspects, we believe that neoplasia should be considered to explain the pathogenesis of hairy polyps in adults. Being characteristically bigerminal, they have traditionally been considered as ''dermoid''. However, the consideration of hairy polyp as teratoma–a true neoplasia–is based on the changing concept of teratoma itself, whereby the classical belief of it being a trigerminal lesion is argued. Teratoma is presently considered a neoplastic mass composed of ''any two germ layers'',[6,7] or even as multiple tissue-aggregate non-indigenous to their anatomic location,[8] making tissue composition irrelevant. Accordingly, terms like ''bigerminal teratoma''[9] and ''benign teratoma''[10] have been attributed to describe hairy polyps. Our description of hairy polyp as *bigerminal choristoma*, therefore, fits in with the evolving concept of teratoma in terms of composition and location. We believe that hairy polyps in adults constitute a distinct entity that has seldom, if at all, been explored before. It is to be acknowledged that their origin remains controversial with no single acceptable theory, and detailed molecular/cytogenetic analysis is required for a suitable explanation. But clinically, hairy polyps in adults seem to represent focal neoplastic

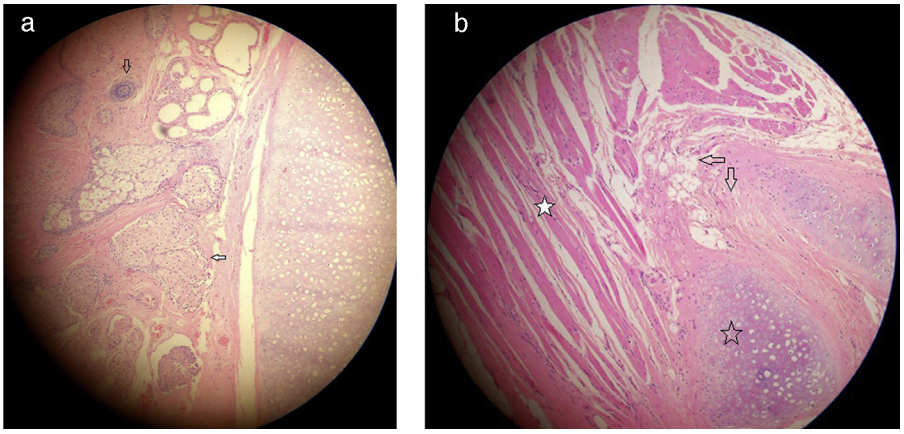

Figure 2 Histopathology showed mature tissue elements of ectodermal and mesodermal origin, including (a) stratified squamous epithelium, skin adnexa (hair follicle, hollow arrow; sebaceous glands, solid arrow), and (b) fibro-adipose tissue (arrows), cartilage (hollow star) and muscle fibers (solid star). (Hematoxylin & Eosin, ×400).