Bilateral Naso-sinusal Glandular Hamartoma: A Case Report

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Bilateral Naso-sinusal Glandular Hamartoma: A Case Report

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Abstract

Differential diagnosis of unilateral nasal mass may be a troublesome question, and also unusual conditions such as glandular hamartoma, consisting in non-neoplastic or inborn malformation, must be considered. We here describe a 78-year-old patient with an unilateral nasal mass, associated to a bilateral naso-sinusal polyposis, which was initially considered to be an inverted papilloma, (on the basis of the preliminary biopitic examination), but that actually resulted a bilateral naso-sinusal glandular hamartoma at definitive histology, after surgical excision.

Introduction

Differential diagnosis of unilateral nasal mass may be a troublesome question, and also unusual conditions such as non-neoplastic or inborn malformation, must be considered between all the possible causative factors [1], in order to address the proper therapeutic plan.

We here describe a 78-year-old patient with an unilateral nasal mass, associated to a bilateral naso-sinusal polyposis, which was initially considered to be an inverted papilloma, but that actually resulted, after surgical excision, a bilateral naso-sinusal glandular hamartoma.

Case Report(s)

A 78-year-old caucasic man referred to our clinic for a long-time persistent and progressive bilateral nasal obstruction. He also complained subjected hyposmia, ageusia and purulent anterior nasal discharge worsened in the last 5 months. No headache, epistaxis, or visual impairment were reported. The patient was unsuccessfully treated with macrolides for the previous seven days, and a standard maxillo-facial TC-scanning, performed under his personal physician request, showed a complete opacification of both nasal cavities, maxillary and ethmoid sinusus, and an incomplete obliteration of frontal sinuses, with partial erosion of the bony structures. The patient denied any previous naso-sinusal surgical treatment, allergies, asthma, or other bronchopulmonary diseases. His medical history was suggestive for chronic vasculopathy, essential hypertension, and quiescent HCV-related hepatopathy. The patient had moreover cardiac pace-maker for atrial fibrillation, and a positive history for previous anaphalaxys secondary to contrast medium.

Rhinoscopy revealed the presence of a wide perforation of the anterior cartilagineous nasal septum, with complete obliteration of the nasal cavities by whish, glistening, polypoid masses; in addition to this on the left nasal cavity there was a fleshy, blackberry-like and rosy neoformation. Biopsy of the left nasal mass, performed in local anesthesia (made elsewhere), was suggestive for an inverted squamous cell papilloma. Surgical removal by means of a transanal functional endoscopic sinus surgery was therefore planned. In general anaesthesia a vegetating mass anchored to the lateral left nasal wall, just over the middle turbinates and occluding both the nasal cavities, was completely removed by means of a centripetal technique: after debulking of the lesion by means of piecemeal removal and cavitation, dissection was performed into a subperiosteal layer comprising the ethmoidal and the nasal fossa. Nasal turbinates were only partially recognizable, and only the tail of the left middle turbinate was preserved. Bilateral maxillary antrostomy and ethmoidotomy were also performed, completing evacuation of the pathological material that was sent for definitive histological examination. Surgical procedure also included an accurate milling with a dedicated adamantine cutter of the anchoring surface of the mass. Intraoperative multiple frozen sections of the residual margins resulted negative for malignancy.

Histopathological findings

On gross examination, multiple brown polypoid fragments were excised from each nasal fossa, measuring from 0.5 to 4 cm (on right side) and from 0.5 to 3 cm (left side).

On histopathological examination, the fragments showed a glandular proliferation of round glands composed of ciliated respiratory epithelium and admixed mucocytes. A direct continuity with surface epithelium could be observed as well as small seromucinous glands. The stroma among the glands was oedematous with ectasic blood vessels and mild inflammatory infiltrate, both acute and chronic.

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Occasionally a prominent thickened hyalinization of the stroma was observed surrounding the glands (Fig. 1). Histological pattern was suggestive for bilateral naso-sinusal glandular hamartoma associated to inflammatory naso-sinusal polyposis.

Follow-up
Over a twelve-months follow-up period no evidence of recurrence was found.

Discussion

Hamartomas are non-neoplastic or inborn malformations of the developing tissues, consisting in excessive proliferation of one or more of the tissue types [1]. Respiratory epithelial hamarthomas (REHA) are rare entity, deriving from naso-sinusal epithelium, generally affecting the nose and the nasopharynx. Paranasal involvement is quite unusual [2], as reported by the most consistent case-series of REAH, published by Wening and Heffner in 1995 [2], describing 25 out of 31 cases of REHA located within the nasal cavity or nasopharynx, without involvement of the paranasal sinuses. REAH generally develops from the posterior nasal septum (about 70% of reported cases) [2], or from the anterior nasopharynx, and are sometimes associated with nasal polyposis [3,4]. Considering paranasal sites, only sporadic cases involving maxillary or ethmoid sinuses have been published [5], and, to our knowledge, only one case of bilateral localization of REHA in ethmoid and maxillary sinuses has been reported [5].

Our case distinguishes for the bilateral location of REAH, the bilateral involvement of nasal cavities, maxillary and ethmoid sinuses, and for the unusual site of development of the masses, being on the lateral nasal walls, instead of the posterior nasal cavity. Moreover, despite REAH may occur in adult patients with ages ranging between 20 and 81 years old, the most affected age is the middle one, with very few cases over 70 years [1-5]. Our patient was therefore one of the oldest with the diagnosis of REAH. As previously reported coexistence of REAH and naso-sinusal inflammatory polyposis is quite frequent [3-4], although it is still unknown if REAH develops easily on a chronic inflammatory setting or, otherwise REAH itself induces a chronic inflammatory condition favoring development of nasal polyps. In our patient the clinical and radiological pattern, beside preliminary histologic examination, were suggestive for an inverted squamous cell papilloma, a locally invasive neoplastic lesion, inducing bone erosion. This diagnosis was not confirmed at the definitive histopathological examination, which assessed the presence of a glandular hamartoma of the nasal cavity, bilaterally extended to both ethmoid and maxillary sinuses, associated to a massive naso-sinusal inflammatory polyposis. This diagnostic pitfall must be taken into account whenever managing nasal masses, as inverted papilloma may present a surface composed of respiratory-type epithelium similar to REAH, while some histologic findings such as proliferating thickened epithelium with mucocytes, intraepithelial mucous cysts and inflammatory cells within the epithelium are typical of inverted papilloma and may be helpful in distinguishing it. This distinction is clinically relevant because inverted papillomas are true neoplasms and require a complete surgical excision since their locally destructive attitude [2]. REAH can be distinguished for its adenomatoid glandular structures with a single layer epithelium. In addition to inflammatory polyps and inverted papilloma, low grade naso-sinusal adenocarcinoma must be considered in the differential diagnosis with REAH, in order to avoid any invasive or defacing treatment. In low grade adenocarcinomas the cell type composing the glandular proliferation, high grade cytologic features and mitotic activity address the right diagnosis.

Conclusion

Differential diagnosis of unilateral nasal mass may be a troublesome question, and also unusual conditions such as glandular hamartoma, consisting in non-neoplastic or inborn malformation, must be considered, in order to address the most pertinent treatment.

References

Illustrations

Illustration 1

Respiratory epithelial adenomatoid hamartoma
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