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OSA Revealing Adenomatoid Respiratory Epithelial Hamartoma: Discussion of a Clinical Case and Review of the Literature

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Abstract | **Case Report**

Respiratory epithelial adenomatoid hamartoma (HERA) is a benign pseudo-glandular proliferation of the respiratory epithelium, often associated with chronic nasosinus polyposis, it localizes preferentially in the olfactory clefts. Its clinical presentation is non-specific or even atypical. Imaging, based on naso-sinusal CT focusing on the olfactory slits, plays an important role both diagnostically and therapeutically. Diagnosis of certainty is based on histology. Differential diagnosis is mainly represented by inflammatory polyps, inverted papillomas and malignant tumors of the nasal cavity. The treatment of choice for HERA is exclusively surgical, consisting of complete surgical removal of the tumor and its base of implantation, ensuring in the majority of cases an excellent prognosis with no recurrence. Malignant degeneration has not been reported. The aim of our article is to analyze the clinical, therapeutic and evolutionary characteristics of nasal adenomatoid respiratory epithelial hamartomas, based on a case report of a hamartoma discovered in a 74-year-old female patient during exploration for OSA, and a review of the literature.

Keywords: Respiratory epithelial adenomatoid hamartoma, rare tumors, nasal polyp, computed tomography, endoscopic surgery, recurrence.

INTRODUCTION

The respiratory epithelial adenomatoid hamartoma (HERA) of the nasal cavity and sinuses is a relatively rare anatomopathological entity [1,2], which may present in isolation or in association with other inflammatory pathologies. Its etiopathogeny is still poorly elucidated, but appears to be linked to intense and/or prolonged local inflammation [1,3].

The clinical picture is unspecific, dominated by chronic nasal symptoms, mainly unilateral nasal obstruction, often resistant to local medical treatments. Imaging is non-specific, generally showing enlargement of the olfactory cleft [1,2]. Complete removal of the tumour and its pedicle by endoscopic endonasal surgery remains the treatment of choice to avoid recurrence, which remains rare, malignant degeneration is not reported [2,3].

PATIENTS AND METHODS

A 74-year-old female presented to an ENT consultation with unilateral right nasal obstruction for two years, associated with hyosmia and facial heaviness. Endoscopic examination revealed a large, shiny, brownish polypoid mass completely filling the right nasal cavity, originating in the olfactory slits, nasosinusal computed tomography showed a voluminous right nasal lesion with a tissue-like appearance and no signs of osteolysis (Fig. 1 and 2), with enlargement of the right olfactory slits. Biopsy revealed a benign inflammatory polyp, and our patient underwent total endoscopic surgical excision.5 x 1 cm, corresponding to an adenomatoid respiratory epithelial hamartoma (Fig 3 and 4), clinical control at one year showed no signs of recurrence.

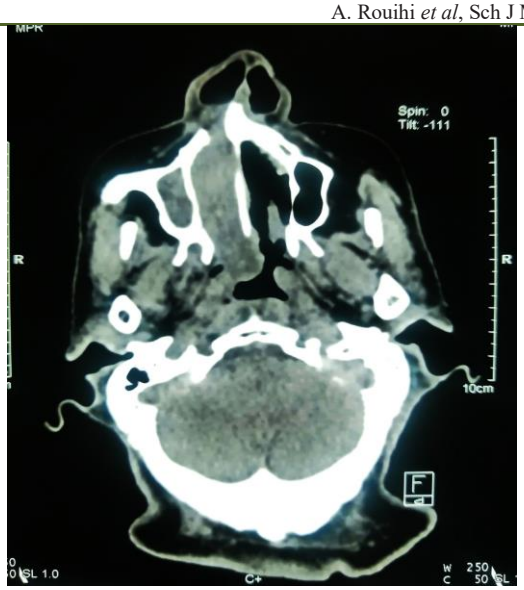

Fig. 1: Preoperative axial section naso-sinus CT scan showing a right nasal polypoidal lesion arising from the olfactory slits

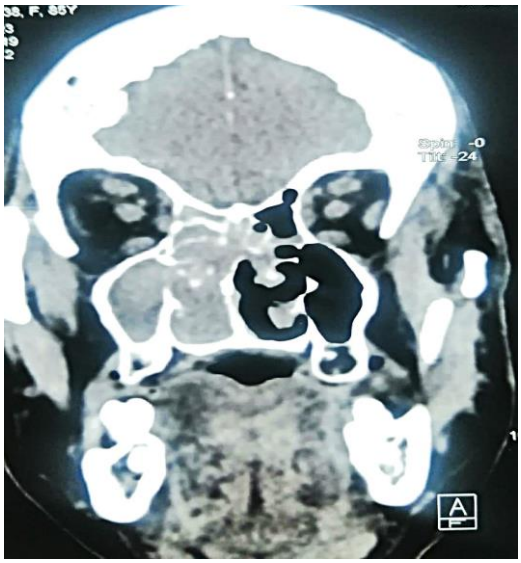

Fig. 2: Preoperative coronal sinus CT scan showing a right nasal polypoidal lesion with implant base originating from the olfactory clefts

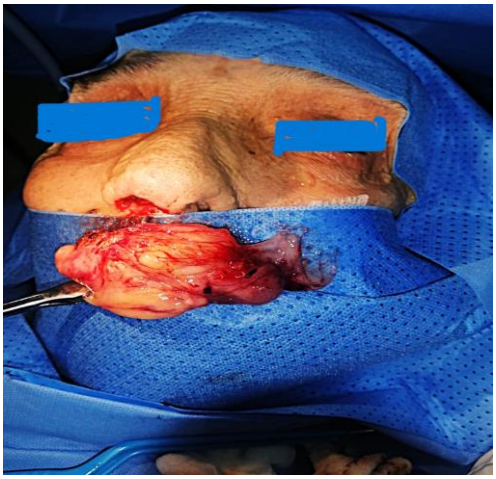

Fig. 3: Excised hamartoma. Macroscopic appearance of polypoidal shiny tissue with firm/granular texture

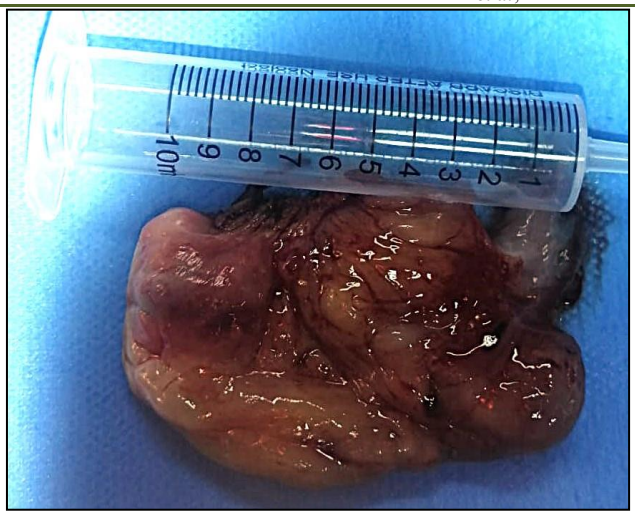

Fig 4: Image showing the cerebriform appearance of the adenomatoid respiratory epithelial hamartoma after surgical excision

DISCUSSION

Respiratory adenomatoid epithelial hamartoma (HERA, REAH) is a rare benign glandular proliferation of the sinus-nasal cavities: respiratory adenomatoid epithelial hamartoma) is a rare benign glandular proliferation of the sinus-nasal cavities, defined in 1995 by Wenig and Heffner [1-2], it generally affects people over 50 years of age with a clear male predilection , although this tumour may be more common, and may be under-diagnosed due to its clinical presentation, which mimics that of a PNS, and to a lack of knowledge of the anatomopathological diagnosis [1-2], it can present in two forms: in association with another nasosinus pathology (associated HERA), most often nasosinus polyposis, while isolated forms represent between 25% of cases depending on the series ,among isolated forms, olfactory localization is the most frequent (75% of cases) [2,3,4]. Its physio-pathogenic mechanism remains unknown, and two hypotheses have been put forward: congenital or chronic inflammatory. Its starting point is described in various ways in the literature, mainly in the olfactory cleft, the lateral wall of the nasal cavity, the maxillary sinus and the inferior turbinate [1-2].

The clinical picture is dominated by chronic nasal obstruction resistant to local medical treatment, rhinorrhea, epistaxis and sometimes hyposmia, cacosmia and facial pain or heaviness [3,4,5]. Careful endoscopic examination of the nasal cavities reveals polypoid masses developed in the nasal cavities that may be flush with the nostril sill, yellow or pink in color, with a shiny surface similar to inflammatory polyps with a cerebriform or metaplastic appearance, sometimes indurated or elastic, most often bilateral, with implantation sites mostly found at the olfactory cleft. Bone erosion and cranial invasion are uncommon [4,5]. Clinically, the main differential diagnoses are inflammatory polyps, schneiderial inverted papilloma and low-grade adenocarcinoma [4-6].

There is no pathognomonic sign of HERA on imaging, but certain scanographic criteria can give a strong presumption: opacities within the nasal cavities, frequently with enlargement of the olfactory clefts, at least partial filling of the ethmoidal cells and inconsistent (retentional) opacities within the other facial sinuses, without erosion bone or intracranial extension [3,4,6] (Fig. 1 ,2). In the case of HERA-FO, lateralization of the middle turbinates may be observed, as well as a discoid-shaped mass syndrome in the olfactory slits on sagittal sections. MRI complements CT, supporting the preoperative differential diagnosis by showing a homogeneous mass with contrast enhancement in T1 after gadolinium injection, and with an isointense or hyperintense signal in T2 [4,6].

Die-cut biopsy is not recommended, as a given mass may contain different zones, suggest several possible diagnoses and leading to therapeutic errors. Anatomopathologically, a hamartoma consists of invaginations of the surface epithelium, a proliferation and accumulation of glands and ducts without atypia, metaplasia or seromucous glands, and covered with respiratory epithelium, have been found. [2,4,5] (Fig. 1)

Antrochoanal polyps are benign lesions originating in the maxillary sinus, resulting in unilateral obstruction. Inflammatory polyps usually present as multiple masses [6,7]. Inverted papillomas (IPs) require radical excision, and can induce bone erosion, spread to mucosal surfaces and invade neighbouring structures [5,7]. They are often located in the lateral nasal wall and present as invaginations of a thickened, stratified squamous epithelium [4,5,7]. Finally, nasosinusal adenocarcinomas are usually located in the middle turbinate or ethmoidal sinuses. They may present with intracranial extension, dysplasia and a high mitosis rate [5].

There is no specific medical treatment reported in the literature. Patients with HERA and nasosinusal

polyposis generally benefit from long-term local corticosteroid therapy for the specific treatment of their polyposis, which can sometimes alleviate nasal symptomatology, but appears less effective than in common polyposis, particularly with regard to nasal obstruction and olfactory disorders [4,6,8].

When HERA is associated with nasosinusal polyposis, surgical treatment is based on endoscopic surgery with polypectomy of the nasal cavities, followed by bilateral total ethmoidectomy; when HERA is isolated and/or diagnosed preoperatively, surgical treatments range from simple complete excision (polypectomy) to subperiosteal dissection, sometimes with milling of the bone opposite the implant foot, so as not to undertreat a possible malignant lesion while awaiting anatomopathological confirmation [5,7,8]. A major challenge in the management of isolated HERA is differential diagnosis with other, more aggressive lesions (in particular, inverted schneiderian papillomas or low-grade adenocarcinomas), in order to provide appropriate treatment and avoid unnecessarily aggressive surgery for benign lesions.

There is no known malignancy potential for HERAs, and recurrences are extremely rare. They generally occur when the proposed surgery was a simple polypectomy, without excision of the implantation sites, and misinterpretation of these lesions can lead to excessive or, on the contrary, insufficient excision.

CONCLUSION

Adenomatoid respiratory epithelial hamartoma of the nasal cavities and sinuses is a benign glandular proliferation of the sinuso-nasal cavities of the elderly, relatively rare most often of the olfactory cleft, its symptomatology is unspecific, there are no pathognomonic signs of HERA on imaging, and the treatment of choice is complete surgical excision with fine, careful dissection of the olfactory cleft and cribriform lamina. The risk of malignant degeneration and recurrence is extremely rare.

Declaration of interest: The authors declare that they have no conflicts of interest in relation to this article.

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