CASE REPORT

Pleomorphic adenoma of the nose*

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SUMMARY

Pleomorphic adenoma is the most frequent tumor of the major salivary glands. It is rarely located in the lacrymal glands and is unusual in the nasal cavity. A rare case of a pleomorphic adenoma of the nose is presented along with a review of the literature. The use of nasal endoscopy in removing tumors of the nose and paranasal sinuses is discussed.

Key words: mixed tumor, nose pleomorphic adenoma, tumors of minor salivary glands

INTRODUCTION

Pleomorphic adenoma is the most common tumor of glandular tissue in the head and neck area. About 8% of all pleomorphic adenomas are located outside the major salivary glands mainly in the oral cavity (Rauch et al., 1970) corresponding to areas with ectopic salivary tissue. Although the nasal cavity has numerous serous and mucous glands, pleomorphic adenoma at this site is a great rarity. The largest series of intranasal mixed tumors reported today in the English literature was by Compagno and Wong (1977) of the Armed Forces Institute of Pathology, Washington DC with 40 documented cases.

Here a case of a pleomorphic adenoma of the nasal septum is reported along with a review of the literature.

CASE REPORT

A 27-year-old woman was referred to our department complaining for a progressive left sided nasal obstruction and intermittent epistaxis for the past 10 months. She had no other E.N.T. symptoms and her past medical history was uneventful. On clinical examination a swelling at the left side of the nose could be seen, while nasal endoscopy revealed a polypoid swelling grayish-pink in color, with a broad base and a smooth surface situated on the left side of the septum 1 cm posterior to the nasal valve and measuring approximately 1.8 cm in diameter. The X-ray of the nose and paranasal sinuses did not reveal any other pathology in the paranasal sinuses. A high-resolution computer tomography scan of the nose and paranasal sinuses revealed a mass confined to the anterior left nasal cavity. The septal mass did not appear to invade the septal cartilage (Figure 1). A biopsy was taken and the histologic result was a pleomorphic adenoma.

Treatment consisted of surgical excision of the mass with the use of nasal endoscope 0° and 30° . The tumor was resected en



Figure 1. High resolution computer tomography scan of the nose and paranasal sinusses, revealing a mass confined to the anterior left nasal cavity.

bloc (including the underlying mucoperichondrium) (Figure 2). The surgical margins of the specimen were free of tumor. The final histopathologic result was pleomorphic adenoma.

The postoperative course was uneventful and the patient was discharged on the second postoperative day.

She was followed for 12 months after the surgery with nasal endoscopic examination at monthly intervals at first and then every 3 months with no recurrence of the disease.



Figure 2. Sugical specimen of the tumor.

DISCUSSION

It seems that the first reported case of pleomorphic adenoma of the nasal cavity was by Denker and Kahler (1929). Batsakis (1980) states that within the upper respiratory tract the most favored site of origin is the nasal cavity, followed by the maxillary sinus and the nasopharynx.

Although the vast majority of minor mucous and serous glands are located on the lateral nasal wall, pleomorphic adenomas in the nasal cavity are mostly originated from the nasal septum. Compagno and Wong in 1977 reviewed 40 intranasal pleomorphic adenomas among which the most common site of origin was the bony or cartilaginous septum (25 out of 40). Suzuki et al., (1990) in a review of 41 cases in Japan reported that only 4 originated in the lateral nasal wall. A list of 9 cases reported in the english literature is presented in Table 1: (Ataman M et al., 1994; Fucci MJ et al., 1994; Prager D et al., 1991; Wallace R et al., 1990; Freeman S et al., 1990; Haberman RS et al., 1988; Wenig BL et al., 1985; Bergstrom B et al., 1981).

Many authors have speculated as to the aetiology of these tumors. Stevenson (1932) suggested that mixed tumors in the nasal septum originate from the remnants of the vomeronasal (or Jacobsons) organ. However mixed tumors are also seen in the lateral nasal wall where no such areas exist and in sections of the tumor no elements have yet been found which could be attributed to the vomeronasal organ (Kamal, 1984).

Matthew et al., (1944) believed that the origin of these tumors was from displaced embryonic ectodermal epithelial cells which are carried via the nasal pits into the septal region. Stevenson (1932) pointed out that growths of embryologic origin are rare in the nasal septum and that to invoke an embryonic explanation for mixed tumors is unjustified. Evans and Cruickshank in 1970 contradicted the previous two theories and claimed that these tumors are entirely epithelial tumors and that they arise in fully developed salivary gland tissue.

Compagno and Wong (1977) in their review found that although the most common presentation is from 30 to 60 years old, mixed tumors of the nose can be found in any age and that females slightly outnumbered males.

Clinically patients usually present with unilateral nasal obstruction. Intermittent epistaxis, a swelling or a mass of the nose, are less frequent complaints, while epiphora, mucopurulent rhinorrhea and headache are rarely reported. The patient usually seeks medical advise within a year of the onset of the symptoms (Batsakis, 1980).

On nasal examination the lesion is frequently described as polypoid, broad-based swellings, grayish-white in color and bleeding easily in response to touch.

The soft tissue mass grows slowly and because it has sufficient space to expand it may cause destruction of the surrounding bone late in its course. Routine X-rays and CT-scans are very important in planning the treatment of lesions of the nose and paranasal sinuses. A high resolution CT-scan reveals the anatomic borders of the mass and its expansive patterns. The pattern of bony alteration whether or not the changes are expansive or destructive provides a reliable clue as to the benign or malignant character of the lesion. The magnetic resonance imaging properties of a mixed tumor are variable. They often have a well circumscribed margin with a low to intermediate signal to T1-weighted images and a high signal to T2-weighted images (Rao et al., 1992).

Histologically, the pleomorphic adenoma of the nasal septum differs from that commonly seen in the major salivary glands by being highly cellular (epithelial), with little or no stromal component. Difficulty may arise in initial biopsies determining the

Table 1. Cases of pleomorhic nasal adenoma. (*: n.m.; not mentioned).

| Author | sex | age | site | symptom | treatment | follow up |
|--------------------|-----|-----|-------|--------------------------------------|----------------------|------------|
| Ataman M (1994) | F | 47 | left | obstruction | lateral rhinotomy | 10 months |
| Fucci M (1991) | F | 80 | left | obstuction epistaxis | surgical removal | n.m.* |
| Prager D (1991) | F | 55 | left | obstruction epistaxis | lateral rhinotomy | n.m. |
| Wallace R (1990) | М | 39 | left | obstruction epistaxis swelling | surgical removal | n.m. |
| Freeman S (1990) | F | 32 | left | obstruction | external rhinoplasty | 1 year |
| Haberman RS (1989) | М | 33 | left | obstruction swelling | lateral rhinotomy | 20 months |
| Wenig BM (1985) | М | 21 | left | obstruction | lateral rhinotomy | 18 months |
| Bergstrom B (1981) | М | 33 | right | epistaxis | lateral rhinotomy | 4 years |
| # | F | 22 | left | swelling occlusion | lateral rhinotomy | 18 months. |

exact nature of the lesion as its cellular pattern stimulates more aggressive neoplasms (Haberman and Stanley, 1989). The increased cellularity of pleomorphic adenomas of this site should be familiar to the pathologist.

When dealing with a unilateral nasal mass one must consider nasal benign polyp as the most likely entity. However the various histologic cellular components of the nose can develop into a wide variety of tumors. The differential diagnoses of benign lesions includes polyps, papillomas (inverting or not), angiofibromas, osteomas, hemangiomas, neurinomas and benign minor salivary glands tumors. In malignant lesions squamous cell carcinoma is the most common intranasal malignancy. In children with unilateral nasal obstruction congenital lesions such as dermoid cysts, external nasal gliomas and encephaloceles must be considered. In adolescents the highly vascular juvenile angiofibroma must be excluded.

Pleomorphic adenomas have a low recurrence rate compared with that of intraoral mixed tumors 25% (Bergstorm et al., 1981). Compagno and Wong (1977) report a recurrence rate of less than 10% in 34 patients followed for more than 3 years. Krolls and Boyers (1972) suggest that tumors with a high amount of myxoid stroma, such as parotid mixed tumors, are more likely to recur because the myxoid stroma can easily be spilled into the surgical field providing a focus of recurrence. Extreme care should be taken not to disrupt the tumor in an effort to prevent local and distant seeding of neoplastic cells, that may result in subsequent local recurrence or metastasis.

Rare cases of metastasizing pleomorphic adenoma of the nasal septum have been reported. Freeman et al., (1990) reported a metastasis in an ipsilateral submandibular lymph node, suggesting lymphatic spread. Wenig et al., (1992) in their review analyzed the clinicopathologic and flow cytometric findings of 11 metastasizing mixed tumors (one case originated from the nasal septum). The retrospective analysis of histologic parameters (mitotic rate, cellular pleomorphism, infiltrative growth, vascular or lymphatic invasion) and flow cytometric analysis failed to identify criteria to predict the development of metastasis in these tumors.

Treatment of these tumors is either by local excision or radical procedures. Earlier reports by Mathew et al., (1944) and Stevenson (1932) suggest that mixed tumors in this area should be treated as malignant tumors. Batsakis (1974), stated that for practical reasons all minor salivary gland tumors of the sinonasal tract, regardless of their histological composition, behave in an aggressive manner. Compagno and Wong (1977) claim that removal of these lesions by local but adequate excision is the treatment of choice and the radical procedures to prevent recurrence are unwarranted. Many authors with cases reported in the literature agree with that point of view.(Ataman et al., 1994; Fucci et al., 1994; Freeman et al., 1990; Wallaca, 1990).

The use of nasal endoscopy offered us the possibility to remove the tumor en bloc without an external rhinotomy approach. The follow up of the patient with nasal endoscopy revealed normal findings on all examinations. We believe that the generally benign tumor of the nasal pleomorphic adenoma and its low rate of recurrence supports the approach of conservative surgical management. Excision of small tumors can be safely performed with the use of nasal endoscopy.

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