CASE REPORT

Leiomyoma of the nasal cavity—Report of two cases and review of the literature

Ch. Tsobanidou *

Department of Pathology, Hippokratio General Hospital, Al.Svolou 10, 54622 Thessaloniki, Greece

Received 3 October 2005; accepted 3 October 2005

Summary Leiomyoma of the nasal cavity is an extremely rare tumor and a search of the literature revealed only 24 prior reports. We present two cases of leiomyomas arising from the wall of the nasal cavity with the symptom of nasal obstruction, which they were treated by complete surgical excision.

© 2006 Elsevier Ltd. All rights reserved.

KEYWORDS Leiomyoma; Nasal cavity

Introduction

Leiomyomas are benign tumours of myogenic origin that may occur wherever smooth muscle is present. They are rarely found in the nose and paranasal sinuses where they constitute about 1% of all benign tumors.1

In this paper we present the clinical and histological features of two patients with leiomyomas of the nasal cavity and review of the literature.

Report of cases

Case 1

A man aged 86 years presented to the hospital with a two-years history of right-sided nasal obstruction. There were no other symptoms relating to the nose. On examination he was found to have a mass in the right nasal fossa which appeared polypoidal and seemed to be arising from the region of the inferior turbinate. A computerized tomography (CT) scan of the nose and sinuses showed the lesion to be localized to the nose with no direct invasion of the adjacent structures. The patient was otherwise well.

The mass was removed and macroscopically the surgical specimen consisted of two pieces of smooth, pale, grey, solid tissues with a polypoid surface which measuring 2.2 × 1.5 × 1 cm and 2 × 1 × 1 cm, respectively.

Microscopically the lesion was well circumscribed and was situated just under the mucosa. Numerous thick-walled vascular spaces with compressed or stellate partially pattern lumina were obvious. The inner layers of smooth muscle of the vessel wall were arranged in an orderly, circumferential fashion, while the outer layers merged with leis orderly peripheral muscle fibres. The tumor was composed of spindle cells with ovoid blunt-ended nuclei and pale pink cytoplasm. Mitoses were not seen. Immunohistochemically the spindle cells showed strong positivity for SMA, moderate staining for Desmin and they were negative for NSE, S-100 and EMA.

© 2006 Elsevier Ltd. All rights reserved.

* Tel.: +302310898248; fax: +302310845514.
E-mail address: tsompanidu@yahoo.gr.
The diagnosis was that of an angioleiomyoma.
On follow up about two-years after surgery, no recurrence of the tumor was seen.

Case 2
A 63-year-old woman visited our hospital with the complaint of a several month history of facial pain and nasal obstruction. The patient’s health was otherwise normal. Clinical examination demonstrated a bluish-coloured mass in the region of the right middle meatus. A CT scan revealed a mass in the right nasal cavity that appeared to arise in the region of the superior turbinate with no direct invasion of adjacent structures. The mass was surgically removed.

Macroscopically the mass (measuring $1.5 \times 1 \times 1 \text{ cm}$) consisted of homogeneous pale brown smooth tissue with polypoid surface.

Histologic examination showed that the mucosa over the tumor was thin owing to compression by the tumor. In the subcutis a round solid tumor was seen well circumscribed by a thin fibrous capsule. The lesion was composed of spindle cells with ovoid nuclei and pale cytoplasm. Many vessels some of which had a thin single layer of endothelial cells were seen (Fig. 1). Immunohistochemistry the spindle cells showed strong positivity for SMA and Desmin, moderate staining for vimentin and were negative for S-100 and keratin.

From these findings a diagnosis of leiomyoma with many vessels was made.

Twenty one months after surgical resection the patient is doing well, without evidence of recurrence.

Discussion
Leiomyomas are benign tumors of myogenic origin that occur common in the uterus, frequently in the walls of the alimentary tract and rarely in the skin. They are very rarely found in the head and neck area. According to Enzinger and Weiss 95% of 7,748 leiomyomas occurred in the female genital system, 3% in the skin, 1.5% in the gastrointestinal tract and the remainder in various other sides.²

Vascular leiomyoma is an uncommon type of leiomyomas that is predominantly found in the lungs. The onset of these tumors on the face is rare and this location occurs in only 5–10% of all vascular leiomyomas.³

The first report of an intranasal leiomyoma, that of Maesaka et al. concerning an angioleiomyoma, appeared in 1966.⁴ Since then, 26 cases have been reported, our presents patients included, indicating the extreme rarity of this condition. The rarity is partly attributable to the fact that smooth muscle is sparsely present in the nasal cavity apart from the wall of blood vessels.

In a review of 562 cases of vascular leiomyoma by Hachisuga et al. over a 17-year period, only 48 were seen in the head and neck area and only five of these in the nasal cavity.⁵

Other names for vascular leiomyoma include angiomyoma and angioleiomyoma. However vascular leiomyoma is a more accepted term because it is the traditional name that accurately describes the lesion and is more commonly used in the literature.⁶

There is some controversy as to the nature of vascular leiomyomas. Most authors of related papers assumed that vascular leiomyoma arise from smooth muscle elements in the walls of blood vessels or from aberrant undifferentiated mesenchyme.

Magnier and Hill showed the presence of groups of mature fat cells in 2 of 15 tumour and favored the same genesis as that of the hamartomatous tumor in the kidney and renal capsule angiomyolipoma.

Duhig and Ayer (1959) suggested that some vascular leiomyomas may not be true neoplasms but are vascular malformation. They suggested a progression of the lesion from haemangioma to angioma with much non-striated muscle to vascular leiomyoma to leiomyoma with many vessels and finally to solid leiomyomas.

The reason of vascular leiomyoma is still uncertain, although trauma, steroid therapy and hormonal imbalance have been implicated in the past.⁷

The tumors occur most frequently in the right side of the nasal cavity and in middle-aged women, with a male to female ratio of approximately 2–3.

The age of the patients have ranged from 49 to 76 years (mean age 54 years).

The most common presenting symptoms of nasal cavity vascular leiomyomas are facial pain or head ache, obstruction and epistaxis.

The size of the tumor at the time of the operation ranges from 0.2 to 4.3 cm in diameter, with most of them less than 2 cm.

The small size of these tumors and the lack of symptoms may account for the infrequency with which these tumours are identified in the nose.

The consistency of the tumors was usually described as elastic, hard or firm, but sometimes as soft mass. The cut surface was gray–white or brown in many cases.

The histopathologic typing of soft tissue tumors by World Health Organization classified leiomyoma into three groups: leiomyoma, angiomiyoma (vascular leiomyoma), and epithelioid leiomyoma ( bizarre leiomyoma and leiomyoblastoma).

Leiomyomas of the nasal cavity and paranasal sinuses are commonly the vascular type.

Figure 1 Blood vessels with concentric arrangement of tumors cells (H–E $\times 400$).
Microscopically, the tumors composed of smooth muscle cells with an intricate vascular component. A concentric arrangement of smooth muscle cells was evident around a medium sized vein, together with the usual fascicular arrangement. No mitotic figures were seen and pleomorphism was absent.

Degenerative changes such as fibrosis, calcifications and giant cell reactions also can be present. If serial sections are reviewed, hypercellular zones of smooth muscle cells and thick-walled, large and small vessels may be demonstrated.

Morimoto classified these tumours into three histologic subtypes in 1973: solid or capillary, cavernous and venous. Tumors of the solid type composed of smooth muscle bundles which surround the vascular channels and intervene these vessels, are closely compacted and intersect with one another. Vascular channels in this type of tumor are large in number but usually small in size and slit-like. Tumors of the cavernous type are composed of dilated vascular channels with smaller amounts of smooth muscle, and the muscular walls of these vessels are difficult to distinguish from intervascular smooth muscle bundles. Tumors of the venous type have vascular channels of a venous type with thick muscular walls, and smooth muscle bundles in the tumor are not so compact, therefore the vascular walls can be easily distinguished from intervascular smooth muscle bundles. According to this classification the total of 562 cases of angioleiomyoma could be separated into: 374 cases (66%) of this solid type, 61 (11%) of the cavernous type and 127 (23%) of the venous type. The most common type found in the head and neck area is the venous type.

Several stains have been used to identify vascular leiomyomas including desmin, vimentin, actin, myosin and Masson’s trichrome.

In our patients, the pathologic findings of the tumors exhibited features of vascular leiomyoma and leiomyoma with many vessels.

Malignant variants and recurrences of this neoplasm have been reported, however, they are rare. The most of the tumors were classified as benign, none showing mitoses or the presence of atypia to suggest possible malignant behavior.

It appears that the absence of mitosis is the most useful histologic indicator of benign lesion. Size, infiltration, bone erosion in significant sites and mitotic index are valuable diagnostic aids for these tumors.

The histopathologic differential diagnosis for these lesions include hemangioma, nasal angiofibroma, fibromyxoma, leiomyoblastoma, hemangiopericytoma, angiosarcoma, angiomylipoma and vascular leiomyosarcoma.

The most satisfactory treatment for these lesions is complete excision if they become symptomatic or if there is doubt concerning the nature of the lesion. The true prevalence of these lesions is probably considerably greater than is indicated by the rarity of reports.

References