Naso-Ethmoidal Angioleiomyoma: A Mini-Invasive Combined Endoscopic and Microscopic Surgical Approach

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Abstract

Background: Angioleiomyomas of the paranasal sinuses are extremely rare benign tumors, representing less than 1% of all leiomyomas in the human body.

Methods: A case report is presented describing a patient with a tender mass growing in the subcutaneous tissue with right nasal obstruction.

Results: Transnasal surgical resection was performed through a combined use of endoscopy and microscopy approach. Histological examination revealed the presence of vascular formations with thick muscular wall and abundant fibrous connective stroma, with a diagnosis of angioleiomyoma.

Conclusion: In cases of rare benign tumors of the naso-ethmoidal region, such as angioleiomyomas, a microscopic excision was performed with endoscopic confirmation of tumour resection from recesses not visualized microscopically. This mini-invasive avoids facial demolition and disfiguring surgery as well as external scars of the facial skin.

Keywords: Nasal; Ethmoid; Angioleiomyoma; Endoscopic; Microscopic

Introduction

Angioleiomyomas (AL) of the nasal cavity and paranasal sinuses are extremely rare benign tumors, representing less than 1% of all leiomyomas in the human body [1]. Common sites are the uterus and the gastrointestinal tract [2]. It is very rare into the nasal-paranasal cavities (only fifteen cases are reported in the literature) due to the paucity of smooth muscle tissue in this site [3].

Because of its rarity, the literature of AL in the nasal cavity and paranasal sinus is reviewed and the management is discussed.

Case Report

A 40-year-old man was admitted to the Radiology Department of our institution for the evaluation of a palpable tender mass protruding at the right medial canthus. The patient had observed a slow growth of this mass, associated with progressive reduction of smell, right nasal obstruction and ophthalmologic recurrent self-limited nasal bleeding.

Ultrasound examination revealed a lobulated solid tissue with a rich homogeneous vascularity at the color-doppler. Nasal fibro-endoscopic examination revealed an easily bleeding mass occupying the right nasal cavity completely. MRI confirmed a solid mass (38 x 28 x 28 mm) centered at the level of the right nasal cavity and anterior ethmoid cells, crossing the midline with lateral involvement of the medial orbital wall and dislocation of the right peri-bulbar fat with cranial extension. Disruption of the right paramedial cortical bone of the anterior cranial fossa due to bone remodeling was observed at the contextural CT examination with coronal reformatted images (Figure 1). After gadolinium-containing contrast agent (Omniscan) intravenous administration, the lesion showed intense and homogeneous enhancement with tiny hypointense spots in the context of the lesion. The pathologic study revealed fragments of nasal mucosa with aspects of hyperaemia and mild inflammation.

Transnasal surgical resection was planned and performed as follows by S.F. through a combined use of endoscopy and microscopy approach. Cottonoids soaked in oxymetazoline solution were positioned in the nasal cavity and left in place for ten minutes. One percent lidocaine was used to anesthetize the nasal cavity and paranasal sinuses.

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Figure 1: MR/CT diagnostic work-up. Solid mass showed hypointense signal on sagittal T1 weighted images (A) with net and homogeneous enhancement after intravenous administration of gadolinium contrast agent on coronal (B) and axial (C) planes. CT images (D and E) demonstrated bone remodelling of the cribriform lamina and the medial wall of the right orbit.

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leiomyoma or angiomyoma) and epithelioid leiomyoma (bizarre
organization
nasal polyps and from the nasal septum [6].

In the nasal cavity, the most frequent location is the inferior
smooth muscle in the wall of blood vessels, or from both [4]. When
angioleiomyomas: from aberrant undifferentiated mesenchyme, from
other reported sites include the nasal vestibule, within
the root of the middle turbinate and of the uncinate process.

A completely endoscopic approach was not possible because
tumour bleeding precluded identification of key endoscopic landmarks.
A primary microscopic resection was thus performed, assisted by the
use of 0, 30 and 45 degree telescopes to assure tumour resection from
recesses not visualized microscopically.

The specimen was resected “en bloc”, with care taken that the
dissection was carried out in the subperiosteal plane. The intraoperative
histological examination ruled out the malignant tissue. A CSF fistula
was created during division the adhesions between the mass and dura
mater. The fistula was repaired under endoscope vision using an overlay
technique with lower turbinate graft; fibrin glue was applied to stabilize
the site, which was then covered with surgical absorbable hemostatic
gelatine sponge. The nasal packing was removed on the second day.
The postoperative recovery period was uneventful. Blood transfusions
were unnecessary. The patient was discharged three days later.

Histological examination of the mass revealed the presence of
vascular formations with thick muscular wall and abundant fibrous
connective stroma, with a definite diagnosis of angioleiomyoma
(Figure 2).

One month after surgery MRI examination showed a solid tissue
located at the ethmoidal vault with post-gadolinium enhancement. At
the six and twelve months follow-up MRI examination, reduction of
size and of gadolinium enhancement suggested the presence of post-
surgical granulation tissue with no evidence of residual tissue (Figure
3).

Discussion

We report a case of endo-scopic removal of a large naso-ethmoidal
vascular leiomyoma with cortical bone disruption of the orbital and
anterior cranial fossa.

Vascular leiomyomas of the nasal cavity and paranasal sinuses are extremely rare, making up less then 1% of all human leiomyomas. This rarity is partially due to the paucity of smooth muscle in the
anterior cranial fossa.

According to the literature, the most common symptoms are: nasal
obstruction (56.25%), facial pain (25%) and headache (25%) [1,3,10-
13]. Progressive anosmia was the only clinical complaint in this case,
due to the deep unusual location of the mass in our patient.

In the nasal cavity, the most frequently reported location of the
vascular leiomyoma has been the inferior turbinate. According to Barr
et al. [3], the high incidence of leiomyomas in the inferior turbinate
may be attributed to the large local amount of contractile vascular
tissue in the smooth muscle.

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Treatment for these tumours is based on local resection, and there
are no reports of recurrence after total excision [1,2,8,9].

Surgical approach by naso-sinusal endoscopic surgery or by lateral
rhinotomy depends on tumour location and extension, as well as the need for a better bleeding control and surgeon experience [1,9,10]. A preoperative CT scan is essential for the evaluation of extension to the paranasal sinuses, to look for bony erosion and to plan the most appropriate surgical approach. Our patient underwent tumor “en bloc” removal by an endonasal combined microscopic endoscopic technique. We believe that endoscopic combined with microscopic approach allows to achieve minimal invasiveness. The surgeon benefits from the depth-of-field and the three dimensional view magnified by the microscope. The endoscope, meanwhile, grants a close up view of the lesion and allows to inspect from the midline with the use of wide-angle lenses.

Conclusion

In cases of rare benign tumors of the naso-ethmoidal region, such as angioleiomyoma, a mini-invasive combined surgical approach avoids suffering a demolitive and disfiguring surgery as well as external scars of the facial skin.

References