CASE REPORT

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Leiomyoma of the nasal septum*

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SUMMARY

Leiomyoma is a benign myogenic tumor that may develop wherever smooth muscle is present. It occurs commonly in the uterus, skin, and gastrointestinal tract and is rare within the nasal cavity. Only three of twenty-four reported cases of sinonasal leiomyoma have been found to originate from the nasal septum. Treatment of choice for these neoplasms is surgical excision. We present two cases of nasal septal leiomyoma. Unique features discussed include recurrence of one neoplasm and the technique used to endoscopically repair a cerebrospinal fluid leak resulting from resection of the neoplasm.

Key words: leiomyoma, nasal neoplasms, endoscopic resection, nasal septum, cerebrospinal fluid leak

INTRODUCTION

Leiomyomata are benign myogenic tumors that originate in smooth muscle. They occur most commonly in the uterus (95%), skin (3%), and the gastrointestinal tract (1.5%) (Murono et al., 1998). They rarely occur within the nasal cavity and paranasal sinuses. Maesaka et al. (1966) reported the first case of angioleiomyoma of the nasal vestibule in 1966. Since then, twenty-three cases of sinonasal leiomyoma have been reported in the literature (Murono et al., 1998). Of these, only three were found to originate from the nasal septum (Ardekain et al, 1996; Barr et al., 1990; Llorente et al., 1996). The remaining reported sinonasal leiomyomata are distributed in the turbinates (9), vestibule (4), maxillary sinus (3), nasal cavity not otherwise specified (3), anterior nasal floor (1), and choana (1) (Ardekain et al., 1996; Barr et al., 1990; Llorente et al., 1996). The rarity of this tumor in the sinonasal cavity is thought to be related to the regional paucity of smooth muscle. In the nasal cavity, smooth muscle is located in the walls of blood vessels and as piloerector muscle in the anterior part of the vestibule (Murono et al., 1998). The higher frequency of intranasal leiomyoma originating on the turbinates is attributed to the abundant local vascular contractile tissue containing smooth muscle (Barr et al., 1990). The most common presenting complaint is a painless mass. Grossly the submucosal lesion is solitary, well-demarcated, sessile, tan-white to blue-gray, and homogenous on cut section. Histology will classically demonstrate interlacing fascicles of cells composed of blunt-ended nuclei with abundant eosinophilic cytoplasm and without significant pleomorphism or mitotic activity (Wenig, 1993). We present two cases of leiomyoma occurring in the nasal septum, discuss the necessity of complete excision, and present a technique for endoscopic repair of cerebrospinal fluid fistulae.

CASE REPORTS

Case 1

A 50-year old woman undergoing magnetic resonance imaging for persistent facial nerve weakness following an episode of Guillan Barré disease was found to have a right nasal mass with sphenoid obstruction (Figure 1). On further questioning, she reported occasional nonspecific head pain and right nasal obstruction. Nasopharyngoscopy revealed a fleshy submucosal mass extending from the posterior superior septum to the medial aspect of the middle turbinate. The lesion was biopsied in the office using endoscopic equipment. Several days later, the patient sneezed out the remaining tumor and was found to be grossly free of disease on endoscopic examination at that time. Histopathologic analysis of the specimen demonstrated leiomyoma. On routine follow-up eight months later, she complained of recurrent head pain and nasal obstruction, and examination revealed a recurrent right posterior-superior septal mass extending to the middle turbinate. Formal endoscopic resection was then performed, and a portion of the cribiform plate was resected in continuity with the mass to ensure complete removal with

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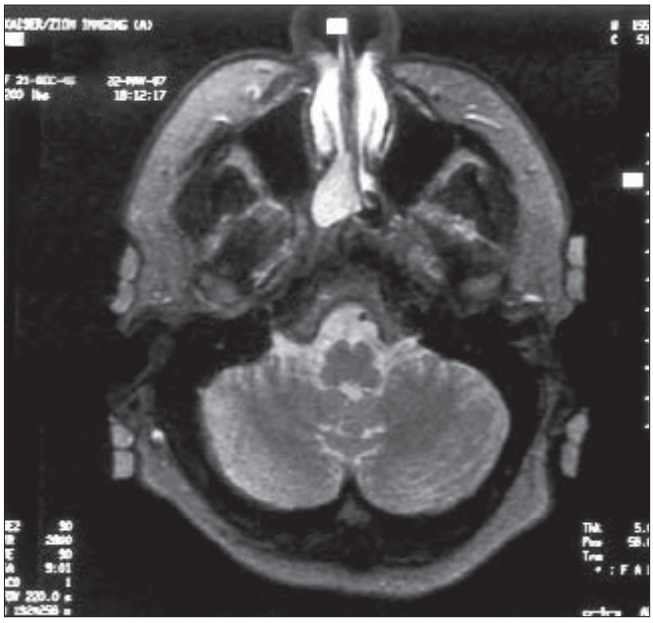

Figure 1. MRI of case 1 showing obstruction of the sphenoid sinus by a right posterior nasal mass which is bright on this T2-weighted image.

a margin of normal tissue. During excision, a cerebrospinal fluid leak was identified and patched with mucosa from the middle turbinate. Histology of this recurrent mass again demonstrated leiomyoma (Figure 2). The patient had no complications from the repair of the cerebrospinal fluid leak and is currently free of disease at three years of follow-up.

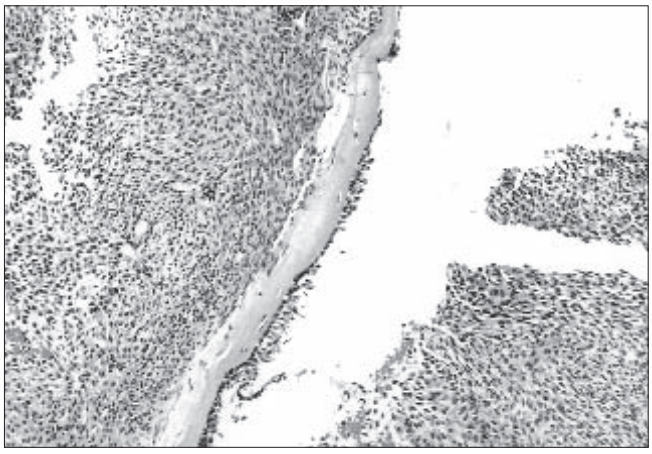

Figure 2. Hematoxylin-Eosin stain of the vascular leiomyoma from case 1 (40x), demonstrating spindle cells arranged in tight fascicles with few mitotic figures and little nuclear atypia.

Case 2

A 70-year old woman was referred for evaluation of an asymptomatic right nasal mass. She had no history of trauma, nasal surgery, or paranasal sinus surgery. She denied alcohol and tobacco use, epistaxis, pain, crusting, or airway obstruction. She had no chronic medical conditions and presented in her state of usual good health. Physical examination demonstrated a 0.5 cm fleshy submucosal mass arising from the right anterior septum. The remainder of her head and neck examination was unremarkable. She underwent an endoscopic excisional biopsy of the mass with histology consistent with leiomyoma. She had an uneventful postoperative course and has been free of disease for six months.

PATHOLOGIC FINDINGS

Histopathologic examination in both cases revealed highly cellular samples of small ovoid and spindle cells arranged in tight fascicles with no identifiable mitoses and scant, ill-defined cytoplasm. The neoplasms stained positive for smooth muscle actin and negative for S100, factor 8, CD34, and keratin. The final diagnosis in both cases was vascular leiomyoma. The lack of nuclear atypia with evenly distributed chromatin and paucity of mitotic figures are important factors in ruling out leiomyosarcoma, which is a malignant lesion warranting far more aggressive treatment than leiomyoma.

DISCUSSION

The World Health Organization classified leiomyomata into three groups: leiomyoma, vascular leiomyoma, and epithelioid leiomyoma (Maesaka et al., 1966). Leiomyomata of the nasal cavity and paranasal sinuses are commonly of the vascular type (Maesaka et al., 1966) as in the cases presented here. Leiomyomata grow slowly and may persist for many years before becoming symptomatic. Patients usually complain of nasal obstruction but may present with pain or epistaxis. In all the reported cases, surgical excision was the treatment of choice without any reported cases of recurrence after complete excision (Llorente et al., 1996). Our second case recurred eight months after the patient spontaneously extruded a portion of the lesion, most certainly as a result of growth of residual tumor tissue. This event demonstrates the potential for recurrence following incomplete removal and the necessity of complete excision to ensure definitive therapy.

The recurrent leiomyoma we have described involved the fovea ethmoidalis and lateral aspect of the cribiform plate, requiring their excision to ensure complete removal of the mass. A small cerebrospinal fluid leak resulted and was corrected at the time of excision. Cerebrospinal fluid fistulae most commonly occur as a result of trauma, either as an accident or during surgical intervention. The treatment of CSF leak of the sinonasal cavity is greatly enhanced utilizing endoscopic sinus surgery techniques. The dura is adherent to the cribiform plate and fovea ethmoidalis, and therefore, if the bone is to be penetrated, or as in this case, partially excised, a CSF leak should be anticipated. Intraoperative leaks should be repaired immediately. Graft material may include fascia lata, temporalis fascia, septal or turbinate mucosa, muscle, or fat. The key to endoscopic repair is exposure; if the septum or middle turbinate is obstructing the area of interest, it should be removed to facilitate the repair. This technique has the added benefit of providing tissue that can be used as a graft in the repair. Tucking the graft between the bone and the dura is ideal but not necessary, and is difficult near the cribi-

form plate and fovea ethmoidalis because of dural adhesions. Dural elevation is best not performed to prevent further tearing. If a large defect is evident, the graft can be reinforced with nasal septal cartilage or bone. Postoperatively, all patients should be on bed rest for several days with the head of the bed elevated.

Three cases of nasal septal leiomyoma have been reported in the literature; we have reported two further such cases. This review demonstrates the utility of endoscopic equipment in the evaluation, diagnosis, biopsy, definitive treatment, and postoperative follow-up of all patients with sinonasal tumors. These rare tumors should be in the differential diagnosis of all patients presenting with a nasal mass and should be completely excised to provide definitive treatment.

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