Sinonasal leiomyoma: Report of 2 cases

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Abstract
Leiomyomas are benign smooth-muscle tumors that are common in the alimentary tract, uterus, skin, and subcutaneous tissue. They are very uncommon in the upper respiratory tract and rare in the nasal cavity and paranasal sinuses. To the best of our knowledge, only 23 such cases have heretofore been published in the literature. We report 2 new cases of sinonasal leiomyoma that originated at different sites in the nasal cavity. We also discuss the various investigative and therapeutic modalities available.

Introduction
Leiomyomas are benign smooth-muscle tumors. They are common in the alimentary tract, uterus, skin, and subcutaneous tissue, but they are very uncommon in the upper respiratory tract. Leiomyoma of the nasal cavity and paranasal sinuses is even more rare. We report 2 new cases of sinonasal leiomyoma that were treated at Lok Nayak Hospital in New Delhi.

Case reports
Patient 1. A 45-year-old woman presented with an 18-month history of recurrent nasal bleeding and nasal obstruction. The bleeding was profuse and had required anterior nasal packing on two previous occasions. She had no history of maxillofacial trauma or nasal surgery. She was normotensive but a known diabetic; her diabetes was adequately controlled with oral hypoglycemics. She had no history of sneezing, watery nasal discharge, or protrusion of a mass from the nose.

Anterior rhinoscopy showed that the patient had a normal midline nasal septum. It also revealed the presence of a fleshy pinkish mass that had filled the left nasal cavity. The mass appeared to arise from the lateral wall of the nose, and it bled upon probing. The right nasal cavity appeared to be normal. No cervical lymphadenopathy was present, and no other abnormality was detected on local and general systemic examination.

The results of routine hematologic and biochemical blood investigations were within normal limits. Contrast-enhanced computed tomography (CT) demonstrated an intensely enhancing soft-tissue mass in the left nasal cavity. The ipsilateral turbinates could not be identified separately. CT also showed that the mass had widened and obstructed the osteomeatal complex; fluid levels were seen in the left frontal and maxillary sinuses. These findings suggested that the mass was a vascular lesion.

The left nasal cavity was exposed via a lateral rhinotomy approach. The mass was found to originate in the lateral nasal wall, and it was adherent to the nasal septum. It was excised in totality. No significant intraoperative bleeding or postoperative complications were noted. Histopathologic examination of the excised mass identified it as a leiomyoma.

Patient 2. A 65-year-old woman sought evaluation for a 4-month history of recurrent right nasal bleeding and nasal obstruction. She had no history of surgery or trauma to the area.

Anterior rhinoscopy revealed that a fleshy pinkish mass was occupying the right nasal cavity. No other abnormalities were found in the left nasal cavity, nasopharynx, ears, throat, and neck. Contrast-enhanced CT showed that the contrast-enhancing mass had filled the anterior portion of the right nasal cavity (figure 1). The tumor appeared to have originated in the inferior turbinate, and it had come into contact with the nasal septum. No bone destruction was evident.

The patient underwent an excision biopsy via an endoscopic endonasal route under general anesthesia. Direct endoscopic visualization of the mass showed that it had originated in the anterior end of the inferior turbinate on its medial aspect. The tumor was completely excised along with a margin of the nasal mucous membrane. Bleeding was minimal. An antibiotic pack was placed in the nasal cavity for 24 hours. Histopathologic examination of the excised mass revealed that it was a leiomyoma (figure 2).

Discussion
Leiomyomas can occur wherever smooth muscle is present. In a study of 7,748 cases of leiomyomas, Enzinger and Weiss reported that 95% of them originated in the

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female genitalia, 3% in the skin, and the remainder at other sites. Fewer than 1% of all leiomyomas in the human body occur in the head and neck. Maesaka et al are credited with publishing the first report of an intranasal leiomyoma in 1966.

To our knowledge, only 23 cases of leiomyoma involv-
ing the nasal cavity and the paranasal sinuses have been reported previously in the literature since 1966. Two of these leiomyomas involved the paranasal sinuses, but because the lesions were so large, it was difficult to speculate as to where they had originated. Of the remaining cases, 9 arose from the inferior turbinate, 5 from the nasal vestibule, 3 from the nasal septum, and 2 from the middle turbinate; the location of the remaining 4 was not specified. To these we add our 2 cases involving different sites in the nasal cavity.

The rarity of smooth-muscle tumors such as leiomyomas in the nasal cavity is probably attributable to the paucity of smooth-muscle fibers in this location. Smooth muscles are present as piloerector muscle in either the anterior part of the vestibule or in the walls of the blood vessels. The literature supports this contention because lesions have been found in the vestibule, the septum, and both the inferior and middle turbinates. The incidence of leiomyomas arising from the turbinates is higher than that at other locations because the turbinates have an abundance of vascular contractile tissue that contains smooth-muscle elements.

Microscopically, leiomyomas are classified as one of two types: vascular and nonvascular. On histology, the vascular type, which is less common, exhibits double-walled vessels, indicating that the mass originated in the smooth muscles of the veins. It has been suggested that a nonvascular nasal leiomyoma might represent a late evolutionary stage in the progression of the lesion from a vascular hemangioma, to an angioma with much nonstriated muscle, to a leiomyoma with many vessels, and finally to a solid leiomyoma.

Three hypotheses have been proposed to explain the origin of smooth-muscle tumors in the nasal cavity:

- They originate in aberrant undifferentiated mesenchyme.
- They originate in smooth-muscle elements in the walls of the blood vessels.
- Nasal vestibule tumors originate in the erector pili of the nostril hair follicles.

Most authors support the idea that leiomyomas that develop in the nasal cavity are of vascular origin. Our review of the literature revealed that sinonasal leiomyomas have occurred primarily in women in a ratio of 5:1. In most of the reported cases, patients were middle-aged or elderly, and most of the lesions had arisen in the right...
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nasal cavity. The reason for these particular characteristics is still uncertain. These tumors grow slowly. Patients with a sinonasal leiomyoma usually complain of epistaxis, pain, and nasal obstruction. Symptoms tend to be related to the obstruction of normal mucociliary flow by the mass effect, which results in pain, headache, and/or acute sinusitis.
A change in airflow may result in nasal crusting, nasal mucosal desiccation, and epistaxis. Local pressure may result in bony erosion and local tumor extension.

Leiomyomas exhibit no characteristic radiologic features. However, in rare cases of extensive sinonasal tumors, CT or magnetic resonance imaging is helpful in determining the extent of tumor invasion and in planning treatment. Because the clinical and radiographic manifestations of an intranasal mass are nonspecific, a biopsy is required for diagnosis.

Gross examination of cut sections typically reveals that the tumor is made up of solid, tan, soft tissue. Histologic study finds interlacing bands of uniform, elongated, spindle-shaped cells, whose nuclei usually have blunt ends (cigar-shaped nuclei). Cells are arranged in fascicles and whorls. Nuclear atypia is absent, and mitosis is rare. With extraterine smooth-muscle tumors, the less-well-established diagnostic criteria for malignancy are high cellularity, the presence of pleomorphism and bizarre giant cells, a high mitotic count, the size of the tumor, and the presence of necrosis. In our 2 patients, a high differentiation of tumor cells, a minimal amount of pleomorphism, and a low degree of mitotic activity indicated that these tumors were benign.

In view of the limited amount of experience in the treatment of sinonasal leiomyomas, surgical excision was the treatment of choice in all reported cases. Recurrence is extremely rare if the excision is complete. In most cases, complete removal is easily accomplished because the pedicle or base of the tumor can be removed along with a surrounding rim of normal mucosa. The size and location of a tumor in the nasal cavity dictate the type of surgical approach. In cases where the mass involves the paranasal sinuses or skull base, a more extensive procedure, including craniofacial resection, may be required. In recent years, endoscopic sinus surgery has proved to be a very useful approach to treating benign tumors.

Although no malignant degeneration has been reported in any of the published cases of nasal leiomyoma, Fu and Perzin observed some findings in one of their leiomyosarcomas that raised the possibility that the leiomyosarcoma had arisen in a preexisting benign neoplasm. Therefore, complete surgical excision should remain the treatment of choice.

References