

# Angioleiomyoma of the Nasal Septum: A Case Report

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## —Abstract—

Angioleiomyoma of the sinonasal tract is a rare benign tumor. We report a case of angioleiomyoma of the nasal septum in a 51-year-old woman who complained of frequent epistaxis for 3 months. Surgical excision was performed. The excised specimen was 0.7 × 0.5 × 0.4 cm in size, well circumscribed, grayish white, rubbery, and soft. Histological examination showed thick-walled blood vessels and smooth muscle cell proliferation. No nuclear atypia or mitoses were present.

**Key Words:** Angioleiomyoma, Vascular leiomyoma, Nasal cavity

## Introduction

Angioleiomyoma (vascular leiomyoma) is an uncommon, benign tumor. It usually occurs on the extremities, especially the lower extremities.<sup>1)</sup> Occurrence in the nasal cavity is extremely rare.<sup>2)</sup> There is a peak of occurrence in the 6th decade. The turbinates are affected most frequently.<sup>2)</sup> Angioleiomyomas are characterized by mature, well-differentiated smooth muscle cells mixed with thick-walled vessels.<sup>3)</sup> Angioleiomyomas of the nasal cavity

have rarely been described in the English literature.<sup>4-7)</sup> To our knowledge, seven cases of angioleiomyoma of the nasal cavity have been reported in the Korean literature.<sup>8-12)</sup> We report a case of angioleiomyoma that occurred in the nasal septum of a 51-year-old woman and offer a review of the relevant medical literature.

## Case Report

A 51-year-old woman presented with frequent

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Table 1. Summary of angioleiomyoma of the nasal cavity in Korean literature

Reference	Age/Sex	Size (cm)	Site	Symptoms
Lee <i>et al.</i> <sup>8)</sup>	45/F	1.0	Left inferior turbinate	Epistaxis
Lee <i>et al.</i> <sup>9)</sup>	40/M	1.5×1.0×1.0	Left nasal dorsum	Nasal mass
Kim <i>et al.</i> <sup>10)</sup>	60/F	2.0×1.2×0.9	Right inferior turbinate	Rhinorrhea, sneezing
Park <i>et al.</i> <sup>11)</sup>	42/F	1.4	Left nasal vestibule	Epistaxis
Park <i>et al.</i> <sup>11)</sup>	67/M	2.0×1.5	Left lateral nasal wall	Nasal obstruction
Kim <i>et al.</i> <sup>12)</sup>	70/F	2.5×1.5	Left inferior turbinate	Epistaxis, headache
Present case	51/F	0.7×0.5×0.4	Left nasal septum	Epistaxis

epistaxis for 3 months. She had suffered from snoring, occasional headaches, and nasal dryness. She had no prior surgical history. On physical examination, a 0.7 cm, papillomatous mass was present at the left anterior nasal septum. Laboratory findings were within normal limits. On plain radiologic examination, no specific lesion was found. The clinical impression was a nasal papilloma. The mass was excised. It measured 0.7 × 0.5 × 0.4 cm in size. On sectioning, the cut surface was well-circumscribed, grayish white, rubbery, and soft(Fig. 1). No capsulation

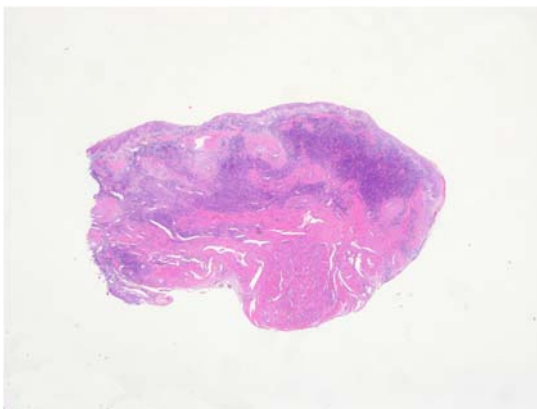


Fig. 1. Whole mount section. The tumor is well circumscribed (Hematoxylin-eosin stain, ×10).

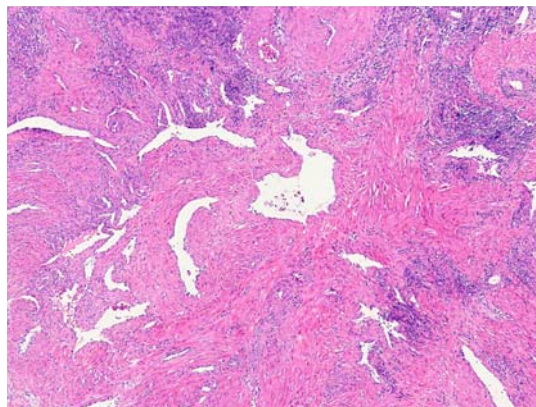


Fig. 2. Microscopic finding. The tumor is composed of thick-walled vessels and smooth muscle cells proliferation. Lymphocytes and plasma cells are infiltrated (Hematoxylin-eosin stain, ×40).

was noted. On histological examination, the tumor was characterized by thick-walled blood vessels and smooth muscle cell proliferation(Fig. 2). Lymphocytes and plasma cells infiltrated the subepithelial connective tissue. The smooth muscles of the thick-walled blood vessels blended with the surrounding smooth muscle tissue of the tumor(Fig. 3). The smooth muscle cells were spindle-shaped and had cigar-shaped, blunt-ended nuclei with eosinophilic cytoplasm. No

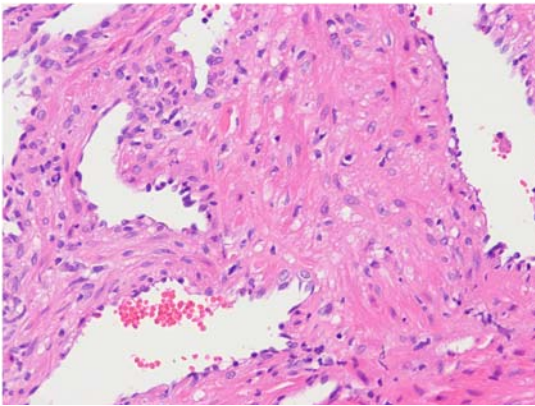


Fig. 3. Microscopic finding. The smooth muscle in the wall of the vessels blends with surrounding smooth muscle tissue of the tumor. The smooth muscle cells have cigar-shaped, blunt ended nuclei with eosinophilic cytoplasm (Hematoxylin-eosin stain,  $\times 200$ ).

mitoses or nuclear atypia were present, and no necrosis was noted. On immunohistochemical staining, the smooth muscle cells showed diffuse positivity for smooth muscle actin and were negative for CD34, estrogen receptor, and progesterone receptor (Fig. 4).

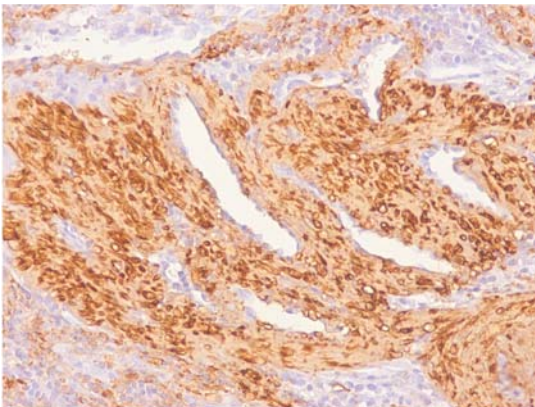


Fig. 4. Microscopic finding. The smooth muscle cells are positive for smooth muscle actin (Immunohistochemical stain,  $\times 100$ ).

The endothelial cells of the thick-walled blood vessels were positive for CD34. A histopathological diagnosis of angioleiomyoma was made. The patient has remained recurrence-free over a period of 6 months.

## Discussion

Angioleiomyomas account for approximately 4.4% of all benign soft tissue tumors.<sup>1)</sup> Most angioleiomyomas occur in the extremities, especially the lower extremities. Angioleiomyomas of the sinonasal tract are very rare. Although subcutaneous angioleiomyomas are commonly painful, tumors occurring in the nasal cavity are not known to be painful.<sup>10)</sup>

Grossly, angioleiomyomas are sharply demarcated, spherical, gray-white or brown nodules, and most of them are less than 2 cm in diameter.<sup>1)</sup> Histologically, angioleiomyomas are composed of thick-walled blood vessels and smooth muscle cell proliferations. Angioleiomyomas are classified into three subtypes according to the dominant histologic pattern; solid, venous, or cavernous. According to this classification, the angioleiomyomas reported by Hachisuga et al.<sup>13)</sup> were classified into 374 cases (66%) of the solid type, 127 cases (23%) of the venous type, and 61 cases (11%) of the cavernous type. The present case was venous type.

Most of the cases reported in the nasal cavity have derived from the inferior or middle turbinate. A summary of angioleiomyomas of

the nasal cavity reported in the Korean literature is shown in Table 1. Tumor locations included the inferior turbinate in three patients, nasal dorsum in one patient, nasal vestibule in one patient, lateral nasal wall in one patient, and nasal septum in one patient. Symptoms included epistaxis in four patients, nasal mass in one patient, rhinorrhea and sneezing in one patient, and nasal obstruction in one patient. Tumor sizes ranged from 0.7 cm to 2.5 cm (mean, 1.6 cm).

The origin of angioleiomyomas of the sinonasal area is uncertain. They may arise from smooth muscle elements in the nasal cavity or from embryonic tissue remnants.<sup>14)</sup> In the nasal cavity, smooth muscles are present as either the pilorector muscles of the vestibule or in the walls of blood vessels. The blood vessels in angioleiomyomas are difficult to classify because they are not altogether typical of veins or arteries.<sup>15)</sup> Their thick walls and small lumens are reminiscent of arteries, but they consistently lack internal and external elastic laminae.

Marioni et al.<sup>14)</sup> described a case of a progesterone receptor-positive, estrogen receptor-negative nasal angioleiomyoma and suggested that the growth of this tumor may be hormone-dependent. Progesterone receptors may influence angioleiomyoma development.<sup>13, 16)</sup> In the present case, the tumor showed negativity for both progesterone receptors and estrogen receptors. On cytogenetic

studies, angioleiomyomas have near-diploid karyotypes, but no consistent abnormalities have been detected.<sup>1)</sup>

The differential diagnosis of angioleiomyoma includes glomus tumor, angioliopoma, and leiomyosarcoma.<sup>3)</sup> In glomus tumors, the constituent cells are round rather than elongate, and the cytoplasm is not fibrillar. Fat may be found in angioleiomyomas,<sup>17)</sup> and this can cause them to be confused with angioliopomas. However, angioliopomas lack smooth muscle, and thrombi are commonly present in the vessels of angioliopomas. Angioleiomyomas can be distinguished from leiomyosarcomas by the characteristic vessels, small size, and lack of mitotic activity and nuclear atypia.

Angioleiomyomas are benign, but very rarely they can recur locally after simple excision. Simple local excision is curative. In the series of Hachisuga et al,<sup>13)</sup> only two patients had recurrence, although their follow-up data were incomplete. The patient in the current study showed no evidence of recurrence at the 6-month follow-up visit.

In summary, angioleiomyomas may be rarely encountered in the nasal cavity. Clinically, they present as small nodules. Surgical excision is important for diagnosis and treatment.

## 요 약

혈관평활근종은 주로 팔다리에 발생하는 양

성 종양으로 비강에 발생하는 경우는 매우 드물다. 저자들은 코중격에 발생한 혈관평활근종 1예를 경험하였기에 문헌 고찰과 함께 보고한다. 51세 여자가 잦은 코피를 주소로 내원하였다. 왼쪽 코중격에 경계가 좋은 종괴가 관찰되었다. 종괴에 대한 절제를 시행하였다. 절제된 종괴는 0.7 × 0.5 × 0.4 cm 크기였으며 회백색을 띠었다. 조직학적으로 종괴는 두꺼운 혈관벽을 가진 혈관과 민무늬근육세포의 증식으로 구성되어 있었다. 민무늬근육세포는 방추형으로 시가형태의 핵을 가졌으며 세포질은 호산성이었다.

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