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A vascular leiomyoma of the ethmoid. Report of case

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INTRODUCTION

Leiomyomas of the sinonasal tract are rare neoplasms. Only occasional reports have appeared in the medical literature since they were first described. (Maesaka et al., 1966). They may occur wherever smooth muscle is present.

The most common localisation is in the uterus, alimentary tract, skin and subcutaneous tissue (Stout and Lattes, 1967). Localisation in the upper respiratory and alimentary tract are extremely rare e.i., nasal cavity (McGaffrey et al., 1978; Lyovetzky et al., 1985); tonsils (Greenberg et al., 1987); larynx (Karma et al., 1978); trachea (Paludetti et al., 1984).

We present the first case of an ethmoidal vascular leiomyoma with secundary severe pansinusitis. Radiographic and pathologic findings will be presented.

CASE REPORT

A 33 year old caucasian man was urgently admitted to the hospital because of a right periorbital swelling, a blocked nasal cavity and mucopurulent nasal discharge. He suffered from severe right frontal headaches.

The medical history revealed a left orbital phlegmone and a frontal brain abscess secondary to a pansinusitis (1978). Therefore he was treated by a transnasal antrostomy, and resection of the diseased parts of the frontal lobe. Postoperatively he suffered from complete aphasia which slowly recovered. A frontal lobe syndrome was present revealing changes in character.

Repeated examination revealed a soft granulating pale mass in the right nasal fossa, replacing the middle turbinate, diplopia to the right, normal eye sight and neurological functions.

Radiological studies showed a bilateral pansinusitis.

We decided to perform a bilateral transnasal antrostomy and external drainage of the frontal sinus. The latter was completely filled with a highly viscous fluid. The bulk of the tumour was removed for further examination. Postoperatively patient recovered fairly well. Further investigations by computer tomography of the paranasal sinus in a coronal direction were done (Figures 1-3).

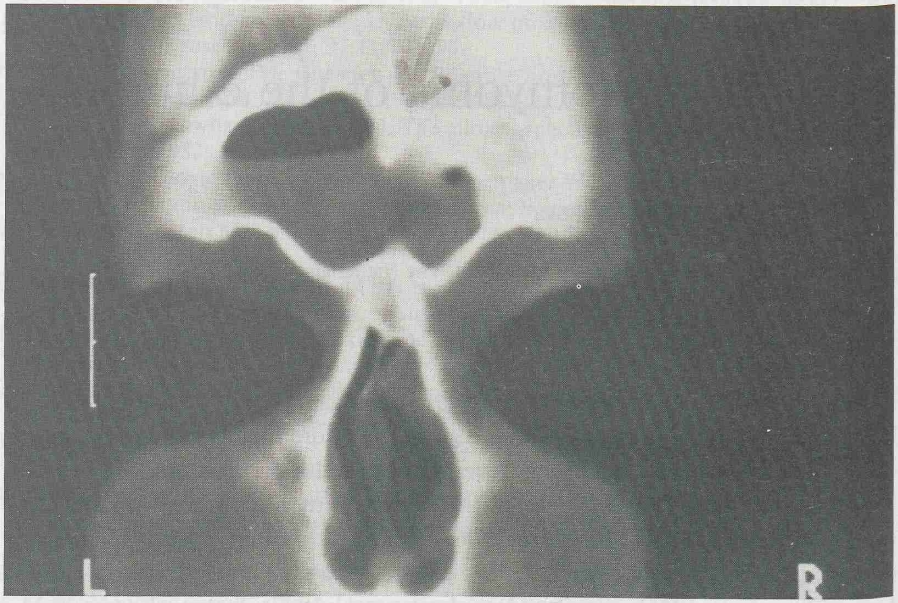

Figure 1. The left sinus frontal showing a fluid level on the coronal CT-scan. Thus pointing out a complete blockage of the nasofrontal duct. At the right side a chronic sinusitis can be suspected.

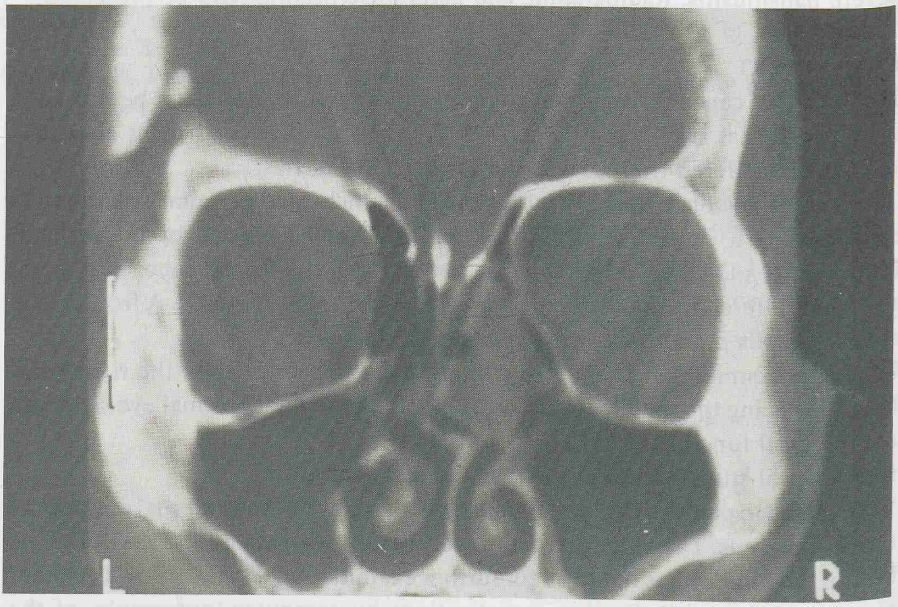

Figure 2. The tumour in the anterior ethmoid sinus can be seen. The open naso-antral window at the left side is patent.

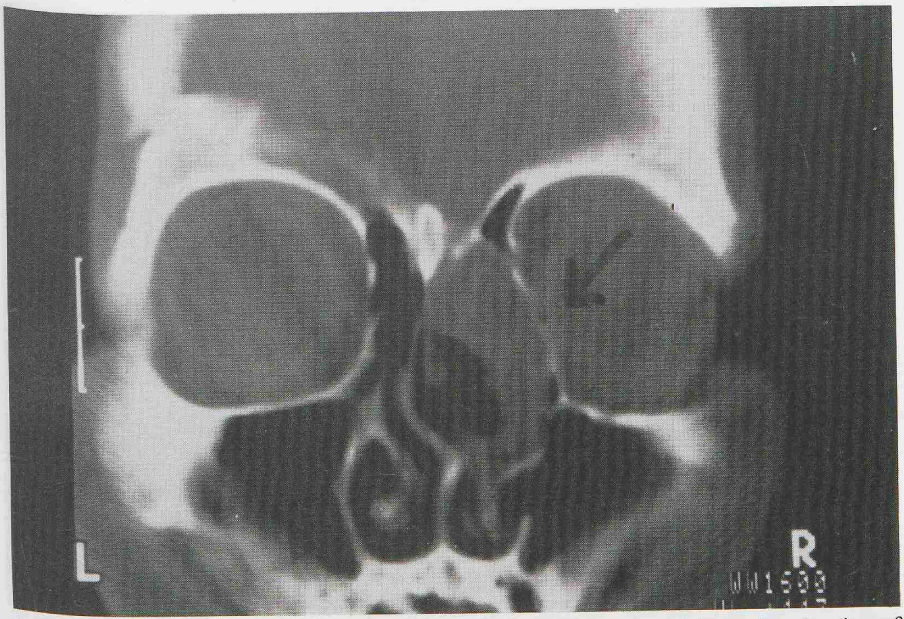

Figure 3. The tumour expands into the posterior ethmoid sinus, without clear erosion of the medial orbital wall. The right naso-antral window is also patent.

After several days a relapse of the frontal swelling was noted and the clinical picture worsened. A lateral rhinotomy was done. Tumour originating from the right ethmoid sinus was removed. The medial orbital wall was destructed by a longstanding process of infection and pressure! Postoperative recovery was uneventful.

Three monthly nasal endoscopy over a period of almost two years showed no sign of recurrence and the patient is free of symptoms (Figures 4 and 5).

PATHOLOGICAL FINDINGS

Grossly the tumour measured 4 × 2 cm in diameter. On section it was a solid and weak, papillomatous mass, greyisch coloured. Microscopically it appeared to be composed of rather uniform, spindle shape cells with eosinophilic cytoplasm and oval nuclei. There was no nuclear atypia and mitotic figures were not seen. There was an increase number of thin walled vascular structures (Figure 6). Immunohistochemical detection of the intermediate filaments showed positive staining for desmin en vimentin.

These findings sustained the smooth muscle origine of the tumour, which was diagnosed as a vascular leiomyoma.

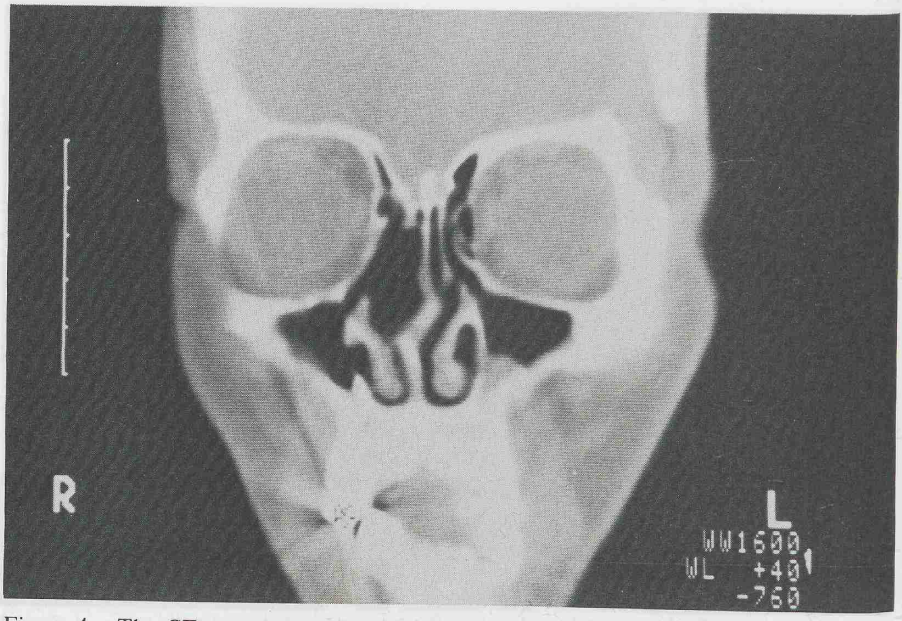

Figure 4. The CT-scan evaluation after one year. No tumour was evident.

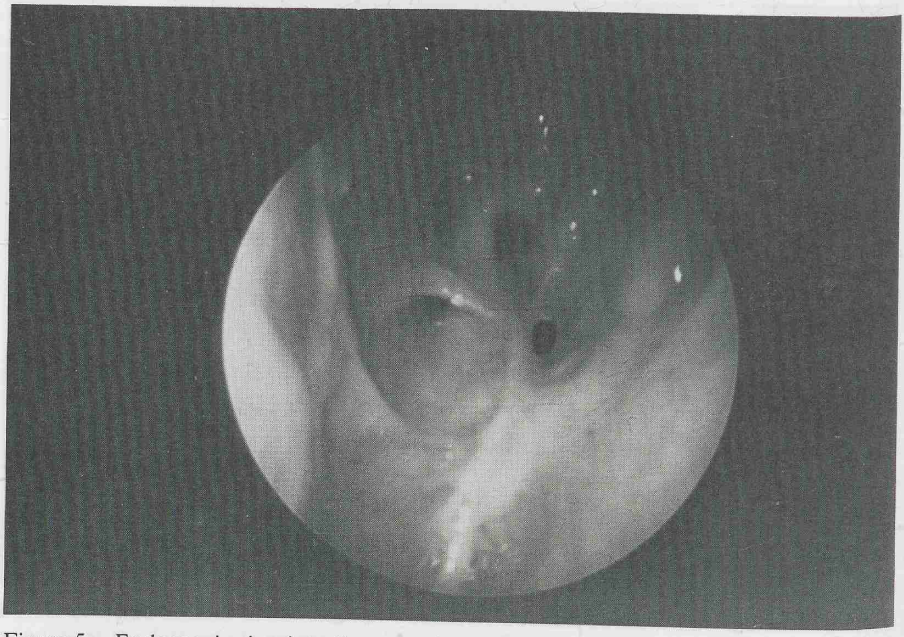

Figure 5. Endoscopic view into the operation cavity. Normal mucosal lining can be seen.

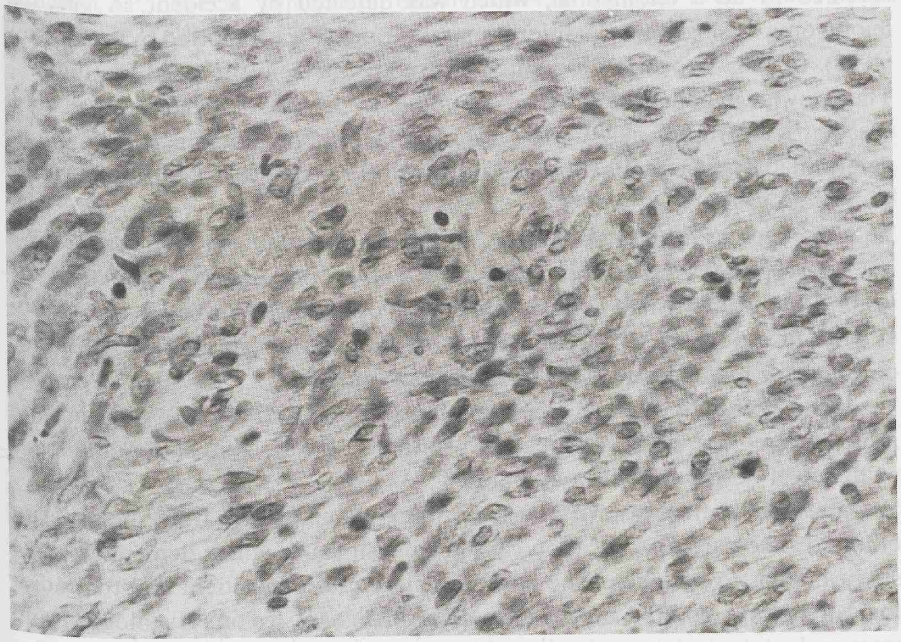

Figure 6. Vascular leiomyoma showing bundles of spindle-shaped cells. H&E X253.

DISCUSSION

The occurence of a leiomyoma other then in the uterus, the alimentary tract and skin is comprehensible rare, because of the relative lack of smooth muscle elsewhere. The chief source of smooth muscle in the nasal cavity is the tunica media of the blood vessels.

The first case was reported by Maesaka in 1966 (Table 1). The lesion involved the nasal vestibule. In 1973 Wolfwitz reported one case. In 1974 and 1975 Fu and Perzin presented a clinicopathological study of the non-epithelial tumours of the nasal cavity, paranasal sinus and nasopharynx. Two out of 156 benign tumours

Table 1. Leiomyoma of the nose.

Authors	Sex/Age	Location	Symptoms	Type
Maesaka et al. (1966)	F , 49	vestibule	pain	vascular
Fu and Perzin (1975)	F , 60	nasal polyp	none	non vascular
	M, 46	nasal polyp	none	non vascular
McCaffrey et al. (1978)	F , 76	inferior turbinate	bleeding	vascular
Lyovetzky et al. (1985)	F , 73	vestibule	pain	non vascular
Zijlker et al. (1987)	M, 33	ethmoid	pain	vascular

revealed to be a leiomyoma, which was detected by accident. A vascular leiomyoma arising from the inferior nasal turbinate was reported by McCaffrey (1978). In 1985 Lyovetzky presented a case arising from the nasal vestibule.
The knowledge about the etiology is limited. In the past is suggested that some vascular leiomyomas may not be true neoplasms, but vascular malformations determined by the combination of trauma and elevated levels of estrogen (Duhig and Ayer 1959).
The leiomyomas of the nasal cavity are clinically benign and recurrences have not been reported after complete resection. It is very important to distinguish this lesion from its malignant counterpart: leiomyosarcoma. This may be very difficult (Stout and Lattes, 1967). It is thought that the mitotic rate is the most reliable criterion for malignancy of smooth muscle tumour.
The clinical picture of a malignant leiomyosarcoma is different; symptoms are more severe like nasal obstruction, epistaxis, facial pain and radiological evidence of destruction of bony borders.
We present the first case of a vascular leiomyoma arising from the anterior ethmoid. The particular location caused a pansinusitis, which demanded acute surgical steps to be taken. The clinically picture of our patient and the radiological findings pointed out to a possible malignant lesion. Histological and immunohistological investigations however showed a benign tumour. After adequate local excision this tumour does not recur, unlike the leiomyosarcoma, which has a poor prognosis in spite of wide surgical excision in combination with radio- and chemotherapy.

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