CASE REPORT

Rhinology, 42, 45-47, 2004

Inferior turbinate angiofibroma: an atypical preservation*

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SUMMARY

Angiofibromas are nasopharyngeal tumours mostly seen in adolescent males. They usually present with nasal obstruction and epistaxis. They account for less than 0.5% of all head and neck tumours. Extranasopharyngeal angiofibromas although rare, have been reported (Ali et al., 1982). The most common extranasopharyngeal site is the maxillary sinus. We report a case of extranasopharyngeal angiofibroma arising from the inferior turbinate which had an unusual clinical presentation.

Key words: angiofibroma, inferior turbinate, extranasopharyngeal.

CASE REPORT

A 30-year-old male of Middle Eastern origin, presented to the Casualty Department with severe bilateral epistaxis, ten days after a partial inferior turbinectomy. He had originally presented to the ENT clinic with a 6 months history of right sided nasal obstruction. The right inferior turbinate was found to be hypertrophied and subsequently underwent a right partial inferior turbinectomy. He had no other medical problems, nor did he give a history of previous epistaxis.

The patient presented with an episode of severe epistaxis and needed resuscitation in casualty with colloids initially and then with 500 mls of blood. He was subsequently transferred to the ward and had intranasal Bismuth iodine paraffin packs placed, and commenced on antibiotics. Within the next 6 hours he had two further episodes of epistaxis losing about 500 mls each time. He then underwent examination under anaesthesia. In theatre, a fleshy mass was found at the posterior end of the inferior turbinate, which was excised. This was followed by profuse bleeding which was controlled with anterior and posterior intranasal packing. Frank bleeding reoccurred on removing the packs and an angiogram was carried out with maxillary artery embolisation. The packs were left *in situ.*

On histology, the excised mass showed a metaplastic squamous surface epithelium and irregular thin walled blood vessels lying in a fibrous stroma. This suggested the diagnosis of nasopharyngeal angiofibroma (Figure 1). The nasal packs were removed uneventfully on the fourth postoperative day and an endoscopic examination of the nose was performed.

A CT Scan performed two months after the surgery showed

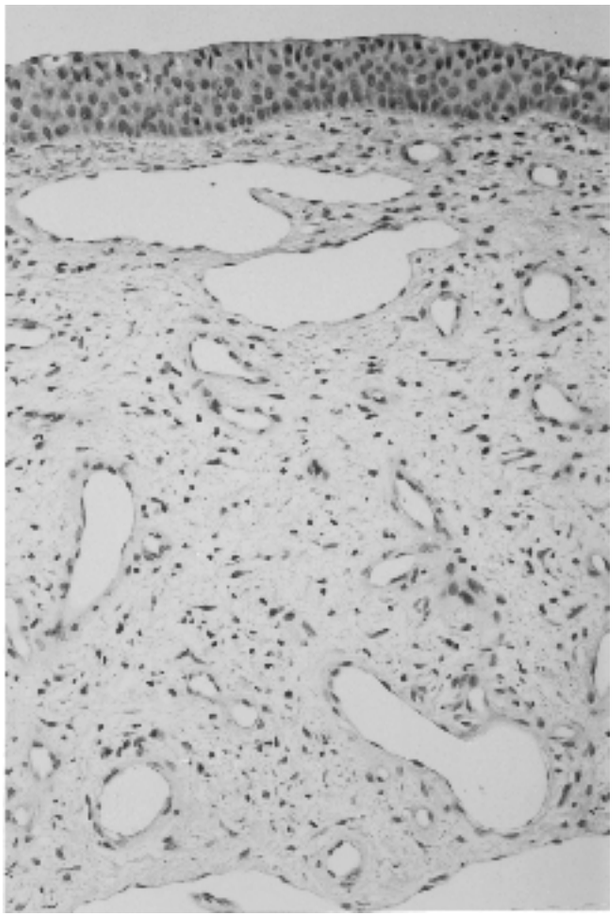

Figure 1. Angiofibroma: numerous irregular thin walled blood vessels lying in a fibrous stroma. Metaplastic squamous epithelium is present on the surface (H&E; x 200 magnification).

*Received for publication: July 17, 2003; accepted: November 17, 2003

no residual tumour. When reviewed in clinic a year later, the patient was asymptomatic and there is no endoscopic evidence of recurrence.

DISCUSSION

Angiofibroma is a benign but locally aggressive vascular tumour, occurring mainly in adolescent males, age range 7-19 yrs and rarely seen beyond 25 yrs (Antonelli et al., 1987). It accounts for 0.5% of all head and neck tumours. The origin of the tumour is usually from the pterygopalatine fossa in the recess behind the sphenopalatine ganglion at the exit aperture of the pterygoid canal (Lloyd et al., 1999). The tumour then spreads into the nasal cavity, nasopharynx and adjacent areas.

Primary extranasopharyngeal tumours are rare, but have been reported. Ali and Jones (1982) reported the first series of 36 cases of extranasopharyngeal angiofibromas, which was updated by Sarpa and Novelly (1989). A recent compilation of 55 cases of extranasopharyngeal angiofibromas was reported by Huang (2000). The mean age at presentation in these extranasopharyngeal angiofibromas is 22 yrs, in comparison to mean age of presentation in nasopharyngeal angiofibromas which is at 17 yrs. Fourteen out of 55 patients with extranasopharyngeal angiofibromas were women (25.5%). Thus, unlike classical juvenile angiofibromas, extranasopharyngeal lesions are seen in older age groups and more often in females.

The commonest site of extranasopharyngeal angiofibroma is the maxillary sinus (17 cases of 55) followed by the ethmoid sinus (5 cases out of 55). Eight cases have been reported arising from the nasal cavity, two each arising from the nasal vault, nasal septum, middle turbinate and inferior turbinate. The first case was reported in a 78 yr old woman with a history of intermittent epistaxis and nasal obstruction and a friable right sided mass on examination of nasal cavity (Alvi et al., 1996). The second case of an inferior turbinate angiofibroma was reported in a 9 yr old boy, who presented with epistaxis and a left nasal polyp, and examination revealed a grey red lesion in the left nasal cavity (Gaffney et al., 1997). In both these patients CT scan revealed a mass confined to the anterior aspect of the nasal cavity. Unlike these two cases, in our case the patient didn't present with any epistaxis prior to the first surgery. The mass was found at the posterior end of the partially excised inferior turbinate.

The usual presentations of angiofibromas are with painless unilateral nasal obstruction, epistaxis, facial deformity expressed as fullness of cheek and proptosis. It progresses gradually and can extend into infratemporal, anterior and middle cranial fossa (Deschler et al., 1992). It presents as nasal mass (80%), orbital mass (15%) and proptosis (10-15%) (Lloyd et al., 1999). Extranasopharyngeal angiofibromas present with variable symptoms depending on their location, but due to the restricted space intranasal tumours present earlier. Various modalities have been used for treatment including surgery, hormonal therapy, radiation, and systemic chemotherapy.

Surgery remains the primary course of treatment (Bremer et al., 1986; Paris et al., 2001). Preoperative investigations include imaging and endoscopy. CT scan and MRI are used to confirm the location and extent of the tumour. CT scan provides vital information about bony invasion especially of the sphenoid, which is the main predictor of recurrence in angiofibroma and therefore helps in proper management of these cases (Lloyd et al., 2000). Angiography along with embolisation is performed preoperatively, to minimise the blood loss. The surgical approaches used vary according to the location and extent of the tumour. A combination of preoperative embolisation and intranasal endoscopic approach combined with laser has been reported to achieve good results in tumours limited to nasal cavity, nasopharynx and the paranasal sinuses (Scholtz et al., 2001).

Recurrence rates vary from 10% to 61% depending upon the early detection of the tumours and its extent. A long term follow up of these patients is recommended.

The importance of this case was to stress on the unusual presentation. Extranasopharyngeal angiofibromas are seen in a much older age group and also in both sexes. They do present with similar symptoms, but when they present in a postoperative scenario like in our case they can be a challenge for the management and treatment of the patient. In an ideal situation, the patient undergoes scanning and angiography prior to surgery for exact localization and in some cases embolisation to reduce operative blood loss, but since our patient presented with a secondary hemorrhage after an elective surgery, he was taken for examination under anaesthesia when the tumour was found and excised. Embolisation was carried out to control the postop bleeding. A high index of suspicion for extranasopharyngeal angiofibromas should be kept in the differential diagnosis of a patient with refractory epistaxis.

ACKNOWLEDGEMENTS

We would like to thank Dr. A.L. Sheehan, Consultant Histopathologist, Doncaster Royal Infirmary, for her help with the histology illustration and in the preparation of the paper.

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ANNOUNCEMENT