ORIGINAL ARTICLE

Surgical Outcome of Recurrent Angiofibroma

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ABSTRACT

Objective: To present our experience with 14 patients of recurrent angiofibroma treated surgically.
Design: A descriptive study
Setting: Department of ENT, Head & Neck Surgery Postgraduate Medical Institute, Lady Reading Hospital Peshawar.
Patients and methods: This study included 14 patients with recurrent angiofibroma. All patients were admitted. A detailed history followed by thorough clinical examination of head and neck region was done. The extent of growth of angiofibroma was studied radiologically by contrast enhanced CT scanning or MRI. All patients were treated surgically and were followed up postoperatively for a period of 2 years.
Results: All patients were male and the average age at diagnosis was 15 years. Nasal obstruction and recurrent epistaxis were the presenting complaints in all patients. Other signs and symptoms included nasal/nasopharyngeal mass, decreased hearing, headache, cheek swelling and proptosis. The tumor and its extensions were well delineated by contrast enhanced CT and/ or MRI. All patients underwent surgical resection of the tumour, in 6 patients by lateral rhinotomy, in other 6 by transmaxillary approach via Weber- Fergusson incision and in 2 via midfacial degloving approach.
Conclusion: In the vast majority of patients recurrent angiofibroma are completely resectable with little morbidity and should be treated surgically after recognition of the extent of tumour by preoperative imaging. Complete surgical resection of the tumour is recommended in order to reduce the high rate of recurrence.
Keywords: Angiofibroma, Recurrence, Residual disease, Surgical approach

INTRODUCTION

Angiofibroma is a rare, benign and highly vascular tumour that occurs primarily in adolescent males. It makes up about 0.05% of all head and neck neoplasms.[1] Angiofibroma originates from the area surrounding sphenopalatine foramen in the posterolateral wall of the nasal cavity. It may extend to the nasopharynx, paranasal sinuses and pterygopalatine fossa. Advanced tumours may involve infratemporal fossa, Orbit, cheek and may even invade the skull base to extend intracranially.[2] Histopathologically, angiofibroma is an unencapsulated tumour that is made up of proliferating, irregular vascular channels within a fibrous stroma. Tumour blood vessels lack smooth muscle and elastic fibres and this feature accounts for the property of the tumour to bleed heavily on manipulation.[3] The blood supply of angiofibroma comes primarily from branches of the external carotid system, although feeders from the internal carotid artery could contribute to its vascularity. To date the tissue of origin has remained unknown[4].

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The classical early symptoms include recurrent epistaxis and nasal obstruction; this in association with a nasal and /or nasopharyngeal mass strongly suggests an angiofibroma. As the tumour expands, hearing loss, facial deformities, proptosis, headache and neurological deficits may occur[1,5]. Clinical examination and nasal endoscopy usually shows a mass behind the middle turbinate filling the choana. Biopsy is absolutely contraindicated because of a considerable risk of massive haemorrhage. Imaging techniques after contrast enhancement (CT and MRI) are essential to confirm the clinical diagnosis and to assess the extension of the lesion.[1,2] Different staging systems based on tumor extension have been proposed. The most practical staging system is that proposed by Fisch. Surgery is the treatment of choice for angiofibroma and the surgical approach is chosen according to the disease stage. The most commonly used surgical approaches include endoscopic, transpalatal, lateral rhinotomy, transmaxillary via Weber-Fergusson incision or midfacial degloving approach. These approaches provide adequate exposure for removal of the tumour and its extensions and if needed can be combined with other approaches. Preoperative angiographic embolization is used to reduce intra operative bleeding. External carotid artery ligation or temporary clamping can be

used as an alternative to angiographic embolization where the later facility is not available.[6, 7] Radiotherapy is reserved as adjuvant treatment in un-resectable tumours, in failure of complete tumour removal or for extensive intracranial extension.[8]
Recurrence is a distinctive feature of angiofibroma which reflects incomplete initial resection. Age of the patient and stage of the tumour at presentation are important factors in predicting the recurrence. The younger the age of the patients and later the stage of the tumour, the higher are the chances of recurrence.[9.10] It is widely believed that recurrence of angiofibroma is principally determined by tumour growth rate coupled with incomplete surgical excision. When initial surgery happens to coincide with early maximum growth rate, recurrence is almost inevitable, if there is anything less than total removal at initial surgery.[2.] Some authorities recommend post operative MR Imaging after removal of the nasal pack and until 72 hours for early identification of any suspicious residual disease.[11] Clinical and radiological follow up helps in early detection of recurrence. This study shares our experience of 14 cases of recurrent angiofibroma treated surgically in our institution.

PATIENTS AND METHODS

We treated 14 patients with recurrent angiofibroma from June 2005 through June 2010. All patients had undergone initial resection of the tumour 1-4 years earlier. No patients had received radiotherapy at any stage. After a detailed history and thorough clinical examination all patients were investigated by contrast enhanced CT Scanning and/or MRI to know the extent of the recurrent tumour. Pre operative embolization was not used in these patients because of local non availability all patients were treated by surgical excision using open surgical approaches. Clinical diagnosis was confirmed postoperatively through histopathological examination done on surgical specimens. Regular clinical and radiological follow up of patients was done for a period of 2 years.

RESULTS

All patients were male and the average age at presentation was 15 years (range 13-20 years) (Table 1). The interval between initial surgical intervention and recurrence was 1-4 years. Recurrent epistaxis and nasal obstruction were the presenting complaints in all patients (100%). Other symptoms included headache (57.14%) and unilateral decreased hearing (42.85%). Important signs on clinical examination were mass in the nose and/ or nasopharynx (100%) cheek swelling (42.85%) and proptosis (28.57%) (Table 2). Routine investigations were within normal range. The extent of the tumour was clearly demonstrated by contrast enhanced CT Scan in all patients (100%). MRI was performed in 8 cases (57.14%) with suspected skull base erosion by tumour. The frequency of involvement of various anatomical sites is shown in the table 3. According to Fisch staging system, 8 patients (57.14%) belonged to stage II and the remaining 6 patient (42.85%) had stage III disease.

Table 1: Age distribution

Age (Years)	No.	%
13-15	9	64.28
15-20	5	35.71
Above 20	-	-

Table 2: Clinical features (n=14)

Clinical feature	No.	%
Recurrent epistaxis	14	100
Nasal obstruction	14	100
Mass in nose/ Nasopharynx	14	100
Headache	8	57.14
Decreased hearing	6	42.85
Cheek swelling	6	42.85
Proptosis	4	28.57

Table 3: Frequency of involvement of various anatomical sites

Site	No.	%
Nose and Nasopharynx	14	100
Pterygopalatine fossa	9	64.28
Sphenoethmoidal region	7	50.0
Maxillary sinus	7	50.0
Infratem poral fossa	6	42.85
Cheek swelling	6	42.85
Orbital proptosis	4	28.57
Intracranial spread (minimal extra dural)	3	21.42

Table 4: Stage of the tumour

Stage	No.	%
I	-	-
II	8	57.14
III	6	42.85
IV	-	-

Table 5: Approaches adopted in this study (n=14)

Approach	No.	%
Lateral rhinotomy	6	42.85
Transmaxillary using WeberFergusson incision	6	42.85
Midfacial degloving	2	14.28

All patients underwent surgical resection of the tumour without pre operative angiographic embolization. Intra operative unilateral ligation of the external carotid artery was used in 8 patients (57.14%) while in 6 patients (42.85%) temporary clamping of the carotid artery was used to reduce

intra operative blood loss. Open surgical approaches were used for tumour resection. Lateral rhinotomy was done in 6 patients (42.85%) to remove the tumour, transmaxillary approach via Weber-Fergusson incision was used in other 6 patients (42.85%) and midfacial degloving approach was adopted in 2 patients (14.28%). All patients required blood transfusion. One to two pints of blood were given intra operatively. No significant post operative complications occurred and no recurrence was detected in any case during 2 years follow up (Tables 4-5).

DISCUSSION

Angiofibroma is notorious for recurrence after treatment. Recurrence is reported in upto 25% of patients regardless of the method of treatment. Since angiofibroma are benign tumours recurrence reflects incomplete initial resection and is more appropriately classified as persistent diseases.[2,3] All patients in this study were male and the average age at presentation was 15 years which compares favourably with other studies[1,5,12]. Surgical excision of angiofibroma at a younger age is associated with higher recurrence rate[3,9] and this was observed in 5 patients (35.71%) who had undergone surgical removal at younger age (10-12 years). These patients had undergone primary surgical resection of moderately advanced angiofibroma 1-4 years earlier to the presentation. In the majority of patients symptoms had recurred within two years of the first operation which is at par with other studies.[2,3,13] Nine patients had previously undergone transpalatal excision of the tumour; four other patients had undergone surgical treatment by lateral rhinotomy approach and in one patient midfacial degloving approach, had been used for removal of the tumour. This shows that the recurrence rate is higher with transpalatal approach and with advanced stage disease[14-16].

Recurrent epistaxis, nasal obstruction and the presence of mass in the nose and/or nasopharynx were the most common (100%) presenting features as seen in most of the studies[3,5,17]. In addition to this usual presentation features of advanced disease like headache, cheek swelling and proptosis were frequently observed. Zeba et al[14] found that 72% of patients in their study had features of advanced diseases (stage III). Tirmizy et al[12] also found relatively advanced cases in their study. Since the advent of CT in 1964, it has been realized that tumours are generally more extensive than can be assessed clinically. The extent of growth of angiofibroma was studied radiologically by contrast enhanced CT scanning (100%) and MRI (57.14%). Imaging studies clearly delineated the tumour and its extensions and helped in staging the disease. The tumour occupied the nose and nasopharynx in 14 cases (100%) each, while the spheno ethmoidal region and maxillary sinus were involved in 7 patients (50%) each. The tumour extended laterally into the pterygopalatine fossa in 9 cases (64.28%), in 6 (42.85%) of whom it had spread farther into the infratemporal fossa and cheek. In 4 cases (28.57%) minimal extension into the orbit through inferior orbital fissure was observed. This has resulted in mild unilateral proptosis. Intracranial involvement was seen in 3 cases (21.42%) in the form of small extradural extension. In these three cases the tumour had gained entry into the middle cranial fossa by eroding the greater wing of the spheroid. Thus in 8 patients (57.14%) the tumour extent was equivalent to Fisch stage II and in 6 patients (42.85%) it corresponded to Fisch stage III.

The treatment of choice in vast majority of patients is surgical resection. Surgical techniques for angiofibroma include open surgical approach and endoscopic surgery. Due to lack of experience with endoscopic approach we did not use it in our study. All 14 patients underwent surgical resection of the recurrent tumour by open surgical approaches. The role of pre operative angiographic embolization in the surgical management of angiofibroma is controversial [3]. It is widely accepted that the use of embolization reduces the occurrence of intra operative bleeding and facilitates tumour removal. Moulin et al do not indicate embolization as a routine pre operative procedure and recommends that it should only be offered to patients with larger tumours.[18] Mccomb et al[19] pointed out that embolization increased the chances of relapse.

Because of local unavailability, high cost, and association with increased recurrence rate pre-operative angiographic embolization was not done. Ligation of ipsilateral external carotid artery or its temporary clamping was adopted in this study which significantly reduced intra operative blood loss. This is in agreement with other studies in the literature[7,18,20]. Ahmad et al[7] suggested that intra-operative temporary clamping of the external carotid artery is a safe and effective means of facilitating surgery and reducing intra-operative bleeding. Among the open surgical approaches lateral rhinotomy, midfacial degloving and trans maxillary approach via Weber Fergusson incision were adopted in this study. Lateral rhinotomy was used in 6 patients (42.85%) with stage II disease. This approach has the advantage of access to the nose, paranasal sinuses, nasopharynx and Pterygopalatine fossa. However it leaves a facial scar and removal of facial bones in prepubertal boys could lead to facial asymmetry. Intranasal crusting, facial numbness and

damage to nasolacrimal duct are other disadvantages of this approach[15]. Same approach was used by Zeba et al[14] in Karachi and Tosun et al[16] in Turkey for the same stage disease with good results.

Midfacial degloving approach is appropriate for tumours in the same locations as those accessible via lateral rhinotomy approach. The advantage is that there is no facial scar but extensive removal of facial bones could affect facial development. Intranasal crusting and vestibular Stenosis are other complications of this approach[14,15]. We used this approach in 2 patients (14.28%). Cansiz et al used midfacial degloving approach in majority of patients with advanced disease and reported good results.[10] Lateral extensions of the tumour to infratemporal fossa in 6 patients (42.85%) necessitated a transmaxillary approach using Weber- Fergusson's incision, the horizontal limb of which was extended as far over the zygoma as required. The anterior, lateral, medial and posterior walls of the antrum were removed, leaving the orbital floor and upper alveolar arch as two intact shelves. Removal of medial antral wall including the vertical plate of the palatine bone along with removal of other walls of the antrum convert the nasal cavity, antrum, nasopharynx, pterygopalatine fossa and infratemporal fossa into one large continuous space. Complete tumour removal was accomplished under direct vision. Three patients with minimal intracranial extradural extension of the tumour were successfully managed by this approach with complete tumour removal from below without formal craniotomy. Thus in recurrent cases with lateral spread to infratemporal fossa a trans maxillary approach using a Weber-Fergusson incision allows complete removal of tumor under direct vision. Small intracranial extension can be safely tackled from below using this approach. Postoperative complications are similar to lateral rhinotomy and midfacial degloving approach. D.A Tandon et al used this approach in majority of patients with recurrent angiofibroma with good results[21]. There was no mortality. Cosmetic results after lateral rhinotomy and Weber- Fergusson incision were acceptable to all patients. The only significant problem post operatively was nasal crusting due to large resultant cavity. The average follow up of these patients was 2 years. No recurrence was detected in any case during this period.

CONCLUSION

Recurrence is a distinctive feature of angiofibroma which reflects incomplete initial resection. Recognition of the extent of the tumour by pre operative CT bone study reduces the risk of recurrence. Aggressive re- excision should always be considered for resectable recurrent disease. The entire tumour should be removed in a wider surgical field under direct vision. Tran's maxillary approach using a Weber- Fergusson incision or midfacial degloving allows complete removal of tumour under direct vision. Limited disease can be approached by lateral rhinotomy.

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