CASE REPORT

Postembolization Infarction in Juvenile Nasopharyngeal Angiofibroma

Syed Khalid Ahmed Ashrafi, Zahid Suhail and Yousuf Khambaty

ABSTRACT

We report the occurrence of cerebro-vascular accident, following selective embolization of internal maxillary artery (IMA) in a young male patient aged 18 years admitted at ENT Department, Abbasi Shaheed Hospital (ASH) with diagnosis of recurrent juvenile nasophayrngeal angiofibroma (JNA). Angiography and selective trans-femoral embolization was done as an adjunctive measure before the definite surgical removal of this benign tumourous condition. The child developed weakness of the right half of body with slurred speech and drowsiness. CT scan of brain revealed the diagnosis of post-embolization infarction. The patient recovered with no obvious signs of clinical residual weakness. Surgical excision after ligation of feeding vessel has been planned.

Key words: *Selective embolization. Juvenile nasopharyngeal angiofibroma (JNA). Complications. Cerebrovascular accident. Internal maxillary artery.*

INTRODUCTION

Recognized since ancient times by Hippocrates, juvenile angiofibroma is an uncommon benign and extremely vascular tumour that arises in the tissues within the sphenopalatine foramen. It accounts for less than 0.5% of all head and neck tumours.[1] The most common presenting symptoms are a triad of nasal obstruction, nasopharyngeal mass and recurrent nasal bleeding. It can also present with facial deformity, proptosis, headache and deafness. The diagnosis is based on CT and MR appearances sometimes confirmed by angiography. Several staging systems have been proposed but that of Fisch is the most robust and practical one.[2] The treatment option is surgical resection (open vs. closed) with adjunctive measures like pre-operative embolization and/or chemotherapy. External beam radiation is another option used alone or in combination but usually is considered a suboptimal primary treatment modality.[3]

The concept of therapeutic embolization of lesions of head and neck can be traced as early as 1930. The primary indication for pre-operative embolization in head and neck is to decrease the intraoperative blood loss. However, it can lead to some other uncomman complications as described in this case report.

CASE REPORT

A young boy presented in the ENT Outpatient Department at Abbasi Shaheed Hospital, Karachi, with complaints of difficulty in nasal breathing for last 4-5 months and intermittent nasal bleeding for 3-4 months. Relevant ENT examination was done. The child had a pinkish globular mass in the right nostril that was sensitive and bled on touch. On posterior rhinoscopy a mass was seen in the right posterior choana. His past medical history was unremarkable but he had a history of nasal surgery for excision of angiofibroma at some other tertiary care centre almost a year back. Based on the history and examination a diagnosis of recurrent JNA was made. The child was admitted with plan of excision of JNA with all the required baseline investigations including CT scan of nasal cavity and paranasal sinuses. Pre-operative angiography and embolization was advised. The angiography revealed that it was supplied by right internal maxillary artery (IMA) with small contribution from left IMA. Selective trans-femoral catheter embolization using gel foam particles was done. The tumour blush completely disappeared (Figure 1).

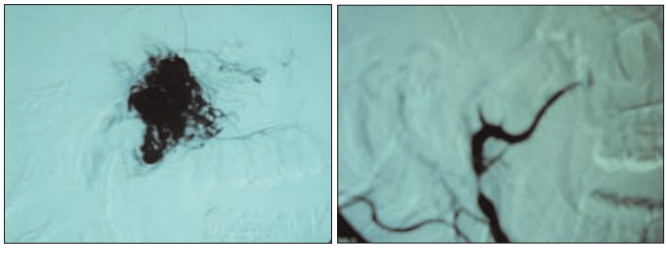

Figure 1: Pre- and postembolization tumour blush.

The child was drowsy after the procedure and subsequently started to develop right sided weakness of the body and had slurred speech. Neurological opinion was sought and a CT scan of brain was advised. It revealed no abnormality. The child was shifted to the Neurology

Department of ENT, Karachi Medical and Dental College and Abbassi Shaheed Hospital, Karachi.

***Correspondence:** Prof. Dr. Syed Khalid Ahmed Ashrafi, A-211, Block-III, Gulistan-e-Jauhar, Karachi.*
E-mail: kashrafi2010@hotmail.com

Received June 15, 2010; accepted July 02, 2010.

Department for optimum care and management. After a lapse of 24 hours a repeat scan was done that showed findings consistent with infarction and so a diagnosis of embolization-related intracranial infarction was made. The child remained under the care of neurophysician for 2 weeks. The child recovered and had no clinical residual weakness. Now he has been planned for surgery with arterial ligation as an adjunct to surgery.

DISCUSSION

Embolization (or embolotherapy) is defined as, "therapeutic introduction of various substances into the circulation to occlude vessels, either to arrest or prevent haemorrhaging; to devitalize a structure, tumour, or organ by occluding its blood supply; or to reduce blood flow to an arteriovenous malformation".[4] The goals of embolization are an adjunctive goal (ex. pre-operative use), a curative goal (for traumatic bleeding or epistaxis) and a palliative goal (in relieving symptoms when surgery isn't an option). It can either be temporary or permanent. It is done by variety of agents which are broadly classified as coils (of tungsten, steel), balloons (of latex, silicone), particulate type (gel foams) and liquid type (alcohols, glues).[5] Each has its own merits and demerits.

The goal of pre-operative embolization in the surgical management of this tumour is controversial. While some surgeons regards it as essential, others have less strict views or frankly disagree. Literature search shows many papers pointing out the benefits of pre-operative embolization in controlling intra-operative bleeding reducing it to as much as 50%,[6-8] allowing full excision with lower relapse rates.

Complications associated with embolization depends upon multiple factors as patient's diagnosis (type and duration of disease, vascular anatomy), type of embolizing agents used and the expertise available.[9] The complications are postembolization syndrome (PES), cerebrovascular accidents, central retinal artery occlusion, infection and abscess formation, failure of embolization of feeder vessel(s), vessel related trauma and even death can occur. Selective maxillary artery embolization is a safe invasive procedure. Rates of complication can be as high as 27%,[10] most of which are temporary inspite of few (2%) severe ones.[11]

The aim of this case report was to provide an awareness that complications can occur during the said procedure. Any such complications should be anticipated and looked for, so as to facilitate early treatment, prompt specialist's referral and prevent any further damage. More so, patient and family members also need to be properly counselled for the risk and benefits of the procedure.

REFERENCES

1. Gleeson M. Juvenile angiofibroma. In: Gleeson M, editor. Scott Brown's otorhinolaryngology, head and neck surgery. 7th ed. London: *Edward Arnold Publishers*; 2008.p. 2437-44.
2. Andrew JC, Fisch U, Valavanis A, Aeppli U, Makak MS. The surgical management of extensive nasopharyngeal angiofibromas with infratemporal approach. *Laryngoscope* 1989; **99**: 429-37.
3. Witarak BJ, Koupmann CF, Turisi AT. Radiation therapy as an alternative to surgery in the management of intracranial juvenile nasopharyngeal angiofibroma. *Int J Pediatr Otorbinolaryngol* 1993; **28**:51-61.
4. Stedman T, editor. Stedman's medical dictionary. 27th ed. Philadelphia: *Lippincott William & Wilkins*; 2000.
5. Byrne JV. Interventional techniques. In: Gleeson M, Scott Brown's otorhinolaryngology, Head and neck surgery. 7th ed. London: *Edward Arnold Publishers*; 2008.p.731-41.
6. Wormald PJ. Endoscopic medial maxillectomy for maxillary and pterygopalatine fossa tumours. In: Wromald PJ, editor. Endoscopic sinus surgery: anatomy, three dimensional reconstruction and surgical technique. New York: *Thieme Publishers;* 2005.p. 125-34.
7. Nicolai P, Berlucchi M, Tomenzoli D, Cappieollo J, Trimarchi M, Maroldi R, *et al.* Endoscopic surgery for juvenile nasopharyngeal angiofibroma: when and how. *Laryngoscope* 2003; **113**:775-82.
8. Katsiotis P, Tzostzis G, Kraminis C. Transcatheter arterial embolization in nasopharyngeal angiofibroma. *Acta Radiol Diagn* 1979; **20**:443-8.
9. Hemingway AP, Allison DJ. Complications of embolization: analysis of 410 procedures. *Radiology* 1988; **166**:669-72.
10. Garcia-Cervigon E, Bien S, Rüfenacht D, Thurel C, Reizine D, Tran Ba Huy P, *et al.* Pre-operative embolization of nasopharyngeal angiofibromas: report of 58 cases. *Neuroradiology* 1988; **30**:556-60.
11. Onerci M, Gumus K, Gil B, Eldem B. A rare complication of embolization in juvenile nasopharyneal angiofibroma. *Int J Pediatr Otorbinolaryngol* 2005; **69**:423-8. Epub 2005 Jan 5.