CASE REPORT

Locally Advanced Staged Juvenile Nasopharyngeal Angiofibroma Treated with Endoscopic Endonasal Approach and Angio-embolization at a Tertiary Institution in Jamaica

P Brown[1], A Mitchell-Daley[1], G Channer[1], P Johnson[2]

ABSTRACT

Locally advanced staged juvenile nasopharyngeal angiofibroma (JNA) traditionally treated with external approaches has been associated with significant morbidities such as blood loss with subsequent blood transfusions, scars, impaired speech, poor swallowing, recurrence and prolonged hospital stays. This case report describes our initial experience in a patient with a locally advanced JNA treated with endonasal endoscopic approach and angio-embolization at Kingston Public Hospital, Jamaica, with subsequent benefits.

Keywords: Angiofibroma, embolization, endoscopic, nasopharyngeal, sinonasal tumour

Angiofibroma nasofaríngeo juvenil en etapa localmente avanzada tratado con un enfoque endonasal endoscópico y angioembolización en una institución terciaria en Jamaica

P Brown[1], A Mitchell-Daley[1], G Channer[1], P Johnson[2]

RESUMEN

El angiofibroma nasofaríngeo juvenil (ANJ) en etapa localmente avanzada, tratado tradicionalmente con enfoques externos, se ha asociado a morbilidades significativas, tales como pérdida de sangre con transfusiones subsecuentes de sangre, cicatrices, trastornos del habla, pobre deglución, recurrencia y estancias hospitalarias prolongadas. Este reporte de caso describe nuestra experiencia inicial con un paciente con ANJ localmente avanzada tratado con un enfoque endonasal endoscópico y angioembolización en el Hospital Público de Kingston, Jamaica, con beneficios subsiguientes.

Palabras clave: Angiofibroma, embolización, endoscópica, nasofaríngea, tumor sinonasal

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INTRODUCTION

Juvenile nasopharyngeal angiofibroma (JNA) is a locally aggressive benign vascular neoplasm, composed of vasogenic and myofibroblastic elements, accounting for 0.05–0.5% of all the head and neck neoplasms (1). It typically affects adolescent males (2).

From: [1]Ear, Nose and Throat Division, Department of Surgery, Kingston Public Hospital, Kingston, Jamaica, West Indies and [2]Department of Radiology, University Hospital of the West Indies, Kingston, Jamaica, West Indies.

Correspondence: Dr P Brown, Ear, Nose and Throat Division, Department of Surgery, Kingston Public Hospital, North Street, Kingston, Jamaica, West Indies. Email: drphillipbrown.ent@gmail.com

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Despite being benign, JNA may cause extensive tissue destruction and bone remodelling *via* extensive submucosal spread (3, 4). It originates usually in the region of the sphenopalatine foramen and may extend anteriorly into the nasal cavity, laterally into the pterygopalatine fossa and superiorly into the intracranial cavity (5). Classical presentations of JNA include unilateral recurrent epistaxis, nasal obstruction and a nasopharyngeal mass (6). More advanced lesions may present with features of intracranial extension (10–20%) as well as orbital involvement with diplopia, facial hypoaesthesia, trismus, and facial deformity. Additionally, due to the vascularity of these lesions, patients may have life-threatening epistaxis and associated massive intraoperative haemorrhage (5).

Staging using different systems is undertaken with the utilization of computed tomography, magnetic resonance imaging (MRI) and angiography (7–9). Computed tomography best demonstrates the bony changes, while soft tissue extent is better evaluated on MRI. Angiography is useful for demonstrating vascular supply as well as intratumoural vascular anatomy, such as arterio-venous shunting, which are important in guiding endovascular pre-surgical embolization (10).

The primary treatment for JNA is surgical excision, by endoscopic, endoscopic-assisted or open surgical approaches (5). The approach is determined by a combination of patient, tumour and institutional factors. The Interdisciplinary Team included team members from Ear, Nose and Throat (ENT) surgery, anaesthesia, interventional radiology and support staff. There is a paradigm shift in the surgical approach to JNA where more endoscopic approaches are being undertaken even for lesions traditionally treated by external approaches. A number of factors (such as improved surgical experience, advances in surgical instrumentation and interventional radiology) are responsible for the current trend. Furthermore, it has been demonstrated that endoscopic approaches have comparable surgical outcomes with less morbidity compared to open approaches, even in advanced staged cases (5, 11, 12). Our index case highlighted the tenets of this evolving modern approach to JNA.

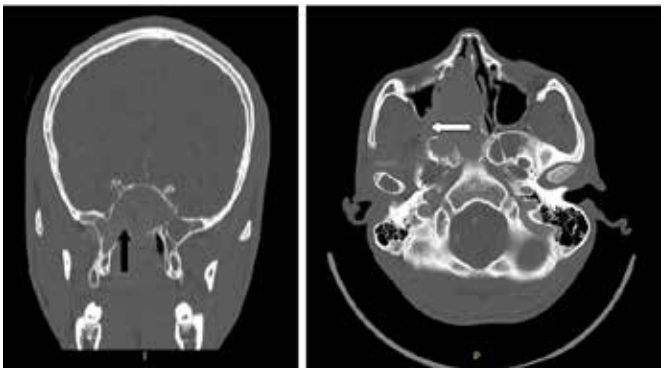
CASE REPORT

A 19-year-old male presented to the outpatient department of Kingston Public Hospital, Jamaica, with a three-month history of recurrent right-sided epistaxis, bilateral nasal obstruction, hyposmia and rhinolalia clausa. The significant clinical finding was a friable right-sided intranasal fleshy mass that prolapsed into the nasopharynx bilaterally. The rest of his ENT examination was normal.

Computed tomography revealed a heterogeneous enhancing mass involving the right nasal cavity, pterygopalatine fossa, nasopharynx with associated bony erosion of the right pterygoid process and right sphenoid sinus floor and anterior wall (Figs. 1, 2).

An interdisciplinary approach was undertaken with the patient having pre-surgical embolization done 48 hours prior to endoscopic surgery. At embolization, a strong tumour blush was demonstrated with feeding branches from the internal maxillary artery (IMAX). The dominant vascular supply to the tumour was predominantly from the sphenopalatine artery and a few tiny branches of the distal IMAX and was embolized utilizing 300 μ polyvinyl alcohol particles. Post-embolization angiography demonstrated satisfactory angiographic devascularization of the tumour (Fig. 3).

The operation was undertaken with the patient placed in the reverse Trendelenburg position and utilizing total intravenous anaesthesia. The surgical technique



Figs. 1, 2: Tumour involving the right sphenoid sinus floor and pterygoid process (black arrow) and involving the right pterygopalatine fossa *via* a widened sphenopalatine foramen (white arrow).

Fig. 3: The feeding tumour vessels of the right internal maxillary branches with tumour blush pre- (black arrow) and post-angio-embolization (red arrow).

employed involved microdebrider-assisted removal of the mass and its bony attachments (Fig. 4). A small nasopharyngeal remnant was removed transorally. The total duration of surgery was 6 hours 30 minutes with blood loss of 700 ml.

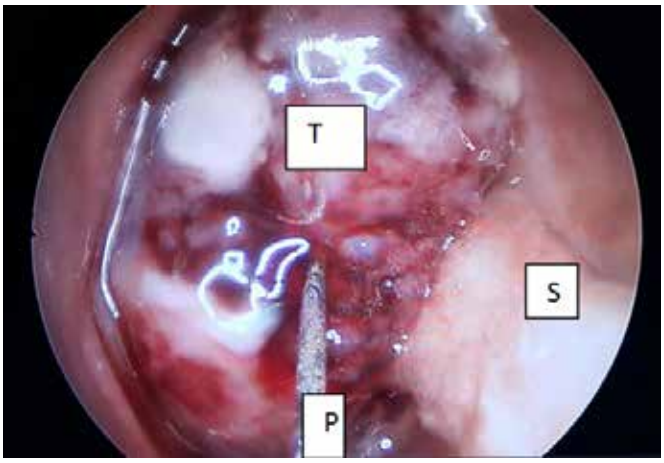


Fig. 4: The intraoperative endonasal view (S: nasal septum; T: tumour; P: probe in tumour).

The patient was discharged on postoperative day 1 with no sequelae. His last follow-up at four months post-surgery was unremarkable.

DISCUSSION

This case highlights the principles in the management of JNA and reflects the evolving surgical approach and paradigm shift in the wider application of endonasal endoscopic approaches.

Firstly, the interdisciplinary team approach is important in the development of an optimal treatment plan in these cases. It allows for collective input from the relevant specialties in identifying important management issues and facilitates a coordinated approach in executing management.

Secondly, early detection of the disease facilitates the application of the less invasive endoscopic approaches. Clinicians should therefore be aware of the possibility of JNA in young male patients with the classical clinical features (6), which will facilitate early investigation and referral. However, more advanced lesions traditionally treated with external approaches are now being treated endoscopically. This is predicated on improved surgical experience, modern radiological imaging and availability of angio-embolization. Pre-operative endovascular embolization has been reported to reduce blood loss at surgery by up to half compared to surgery alone (13). In a systematic review (5), the average blood loss from the purely endoscopic approach was 544.0 ml (range: 20–2000 ml) compared to 1579.5 ml (range: 350–10 000 ml) for the open approach. Endoscopic-assisted cases had an average blood loss of 490.0 ml (range: 100–950 ml). This proved advantageous in our index case, as an endoscopic approach instead of an open approach was facilitated due to the recent improvements and availability of interventional radiology locally.

Intraoperatively, excellent anaesthetic technique is prudent in achieving outcomes such as reduction in intra-operative blood loss, improved surgical field visualization and good analgesia without significant side-effects (14).

The improving surgical experience and training, advances and availability of surgical instrumentation along with the advantages of the endoscopic techniques have been the main driving force of this surgical approach locally. These advantages include excellent visualization of the surgical field through magnified and multiangle exposure to prevent tumour remnants, low morbidity without any external scar or osteotomy, less blood loss and short hospitalization times (15). These were all achieved in our patient. For example, the improved visualization of surgical field allowed us to remove the bony tumour remnants along the pterygoid process and sphenoid body, potential sources of recurrence.

Follow-up of these patients is prudent as recurrence rates vary from 4.7% to 22.6%, regardless of techniques (endoscopic *versus* open) (5, 16). It entails regular clinico-radiological postoperative patient assessments. There was no evidence of recurrence at four months, and he had an improved quality of life.

CONCLUSION

There is a paradigm shift in the management of JNA in which endoscopic endonasal approaches along with angio-embolization is slowly evolving to be the standard of care. This is predicated on the appropriate patient selection, a multidisciplinary team approach, advances in surgical expertise, instrumentation and improvement in interventional radiology locally with the additional advantages of improved surgical outcomes and decreased morbidity. The team at the Kingston Public Hospital was able to achieve all these objectives in our index case with a locally advanced JNA.

AUTHORS' NOTE

P Brown participated in the concept, organization, writing and editing of the manuscript. A Mitchell-Daley was involved in the concept, editing and revision of the

manuscript. G Channer and P Johnson edited and revised the manuscript.

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