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ABSTRACT:

BACKGROUND

To assess and analyze the clinical presentation of Juvenile nasopharyngeal angiofibroma and analyze the complications associated with JNA resection without pre surgical embolization using LeFort 1.

METHOD

This retrospective study conducted at the Department of Neurosurgery,CHK(Civil Hospital Karachi, DUHS(Dow University of Health Sciences), Karachi, Pakistan involves the review of medical records of patients with histologically confirmed Juvenile nasopharyngeal angiofibroma who were treated in between 2014-2016

RESULTS

25 patients were identified, with an average age of 14.5 years (9-20years). Majority of the patients presented with epistaxis. CT scan was the most commonly used radiological investigation for staging. According to RADKOWSKI staging 16 patients (64%) presented with stage IIIb. 6 patient (24%) presented with stage IIIa and 3 patients (12%) presented with stage IIc. Tumor recurrence was seen in 2 patients (8%) while malocclusion was seen in 1 patient (4%).

CONCLUSION

Our study show strong evidence in favor of the lefort 1 osteotomy approach for JNA removal extending into pterygopalatine, infratemporal fossae, paranasal sinuses and base skull regions.

KEYWORDS

Juvenile nasopharyngeal angiofibroma, Surgical intervention

INTRODUCTION

Juvenile nasopharyngeal angiofibroma (JNA) which was first described by Hippocrates, is a benign, infiltrating vascular neoplasm that in most of the cases occurs only in young boy ranging from the age of 9 to 13 years usually. Hippocrates described this tumor as a polyp of nasal origin that "wept blood". JNA makes up almost 5% of all tumors of head and neck origin. "Different terminologies used for this tumor are bleeding fibromas of the adolescence, juvenile basal

fibroma, myxofibroma and fibroids of the nasopharynx".(1)

On histopathological evaluation JNAs are found to be capsulated and non-invasive entities and comprise of many interlocked vascular channels, these channels are in turn lined by a single celled endothelium and interestingly lack any muscular part, which predisposes them to bleed (2). Clinically JNAs are attached to their base, contain many lobules, have a rubbery feel to it and appears to be redish-

pinkish to tan-gray in colour.

Origin of JNAs is from posterolateral aspect of nasal cavity involving the upper border of sphenopalatine foramen, at the posteroinferior aspect middle nasal concha (Figure 1), growing initially within the submucosa. The final configuration of the tumor is achieved by the shaping up and the branching of the pterygoid plate (origin), horizontal wing of the vomer, and the palatine bone. Tumor may grow forward, invading the naso-maxillary region, extending in the sphenoid bone, pterygopalatine and infratemporal fossa invading through the pterygomaxillary fissure. Invasion of the orbit, cranial cavity and cavernous sinus might also occur through the infratemporal fossa. (2, 3)

Vascularity of the tumor usually comes from maxillary branch of external carotid artery. At times, branches from the opposite external carotid artery and from the internal carotid artery from the same side may also contribute towards supply of blood. (2) Blood supply from bilateral carotids has been reported in different studies to be 26.3 % (4) to 46.4 % (5).

The systems used to stage this tumor according to its extension into the anatomic structures, include the Fisch (7) system, which was further modified by Andrews (8); Radowski system (Table 1) (6), Chandler (9), Sessions (10) and Bremer (11). Latest staging systems include Onerci (12), INCan (13) and UPMC (14). System used in this case series is Radowski System of staging JNAs given by Radowski et al in 1996 as it is most popular system among the clinicians. (15)

Diagnostic modalities used for the diagnosis and treatment planning of the tumor include Computed tomography (CT) with contrast and magnetic resonance imaging (MRI), along with a thorough clinical examination.

Routine presentation of the patients with JNA include; symptoms of persistent nasal obstruction and discharge along with severe, recurrent, & spontaneous epistaxis. (16). Expansion of tumor leads into development of, facial deformities, exophthalmos, loss of eyesight, weakness in the cranial nerve regions and recurrent headaches.

Preoperative angiographic embolization of the channels is widely used as a method to reduce the amount of intraoperative bleed and facilitate tumor

access to the mid facial skeleton. L-shaped titanium plates were adapted and fixated on the site of osteotomy and then removed to mark the site of plate removal. (14, 17) Although considerable advances have occurred in modern super selective microcatheterization and embolization techniques, embolizations of these tumors are related with the risk of serious complications such as hemiplegia and loss of eyesight. Lloyd et al. reported that "embolization might be a cause that further contributes to towards recurrence as embolization causes the shrinkage of the tumor making the total removal more difficult particularly when we see deep invasion into the sphenoid, hence patient with this pattern on the CT are at a higher risk of recurrence and in such cases embolization is contraindicated." (18). Also angiographic embolization is not available at all centers and at such hospitals surgical resection of JNA is required without this facility

Literatures published on this up till now have failed to reach a consensus in regard to surgical approach which must be preferred. Approach for access in the tumor depends on site, size, and expertise of the operating surgical team. The usual approaches that are commonly employed by the surgical team includes transpalatine, medial maxillotomy (commonly done using degloving or lateral rhinotomy), LeFort 1, infratemporal, and more recently nasal endoscopic surgery has been used" (19, 20)

In this article we have presented our experience with 25 patients who underwent JNA resection without presurgical embolization, using lefort 1 approach.

MATERIALS AND METHODS

25 cases of young male patients of ages 9-20 years age range diagnosed with JNA were surgically treated between January 2014 to October 2016 at the Department of Neurosurgery, CHK (Civil Hospital Karachi, DUHS (Dow University of Health Sciences), Karachi, Pakistan. Variables observed in the study were presenting signs and symptoms of the patients, Radowski tumor stage, average intraoperative blood loss, rate of recurrence and postoperative occlusal disturbances.

The most commonly employed investigations of choice include Magnetic Resonance Imaging (MRI) and Computed Tomography (CT). Intraoperative external carotid artery ligation for each patient was performed and one year follow up for any complications done.

SURGICAL PROCEDURE

"After induction of general anesthesia, Local anesthesia was administered on the line of incision and a maxillary vestibular incision was made extending from one maxillary tuberosity to the other, 3-5 mm superior to mucogingival junction. Subperiosteal dissection was performed to gain

fixation later so as to maintain pre-surgery occlusion. Lefort I osteotomies were performed from the lateral nasal wall starting from piriform rim across the anterior maxillary wall and extended to the posterolateral aspect of maxilla to the pterygomaxillary fissure using osteotomes. The nasal septum and anterior nasal spine were detached from each other and maxillary crest using septalosteotome directed caudally and posteriorly. The osteotomy of the medial wall of the maxilla was done through extending posteriorly from the piriform fossa through the inferior meatus to the palatine canal vessels. The dysjunction of the pterygoid was performed with the help of sharp osteotomes. The maxilla was then displaced caudally. And adequate access of the surgical field was achieved. Tumor mass along with its extensions was resected. Fixation of the osteotomized maxillary segment was achieved by using the previously adapted L-shaped Titanium plates (4 hole spaced), one at each zygomaticomaxillary buttress and one C shape plate at each piriform buttress area with screws (2.0 mm width, 6 and 8 mm long screws). The post-operative period was uneventful and follow-up was done to evaluate patient's ophthalmic symptoms, nasal disfigurement and recurrence."(1)

RESULTS

Twenty five male patients, age ranging from 9-20 year, diagnosed with juvenile angiofibroma were treated in the time period of 2 years.

Out of 25 patients 8 presented with epistaxis, 5 with nasal obstruction only and 5 with both symptoms. Out of 25 cases 5 presented with facial deformity and only 2 had ophthalmologic symptoms. All the cases were clinically diagnosed after correlation of clinical symptoms with CT or MRI. On staging the JNA according to Radowski system 3 cases were of stage IIC, 6 cases of IIIa, 16 cases of stage IIIb.

Complication recorded intraoperatively was blood loss. Mean blood loss for all patients was 1000 ml. Postoperatively one patient reported with malocclusion and 2 patients had recurrences. The recurrence was at the orbital region in one case and at the base of the pterygoid process in the other. Recurrences occurred 6 and 9 months post operatively.

Immediate post-operative complications such as diplopia, CSF leak and post-operative malocclusion were rectified between 2-4 weeks postoperatively. No major complications were observed.

DISCUSSION

Surgical resection is the acknowledged treatment of choice for JNA. Previously used methods for treatment of JNA were hormonal therapy, chemotherapy and radiation, but are now used only as adjunctive therapy.(21) Different surgical approaches have been used for creating access to these tumors such as

through palate, antrum, maxilla, or lateral rhinotomy, & infratemporal, with each method having its own pros and cons. (22,23)

Numerous elements play an important role when choosing the surgical approach to JNA such as reasonable access to the tumor site, bleeding control, to pre-empt post-operative facial asymmetry and preventing interference with growth of the face. Transfacial, lateral rhinotomy and other extraoral approaches have the advantage of wide access but facial scar, asymmetry, repeated nasal crusting, parasthesia and lacrimal apparatus injury are the unforgivable consequences that might occur postoperatively

The Lefort I osteotomy through maxillary vestibular approach is better than other approaches in that it provides good exposure to the nose, pterygopalatine fossa, infratemporal area and skull base in addition to prevention of visible scarring of the face. (24).

The LeFort I osteotomy for approaching diseases in the cranial base was first described by Cheever in it was used to for resection of a tumor of nasopharyngeal origin.(25) In contrast to the other techniques, down fracture of the maxilla provides an unhindered exposure for tumor removal. Also this approach provide ease of access for the repair of a tumor associated dural tear or mucoperiosteal defect and a better way of controlling haemorrhage.(26)

LeFort I approach provides outstanding cosmetic results as there is no facial scarring associated with this technique. Pre-osteotomy plate adaptation and screw hole marking helps in minimizing post-operative occlusal discrepancies, even though this technique was adopted in all our cases, 2 of our patients came with mild post-operative occlusal discrepancies. Another advantage associated with this approach is that there is no anticipated disruption of facial growth, as the growth centres are not disturbed by the osteotomy. However, damage to the tooth buds of permanent dentition in younger patients is anticipated.(1)

Lefort 1 osteotomy for JNA removal is applicable in both paediatric and adult patients with minimal life threatening complications both pre and post operatively.(27). Avascular necrosis of maxilla is anticipated in 1% of cases (28) which may occur due to one of the multiple possible factors. Intra-operative problems that might cause avascular necrosis of maxilla include rupture of the descending palatine artery (DPA)(29), perforation of the palatal mucosa that impairs blood supply to the maxillary segment(30), post-operative complication that might result into maxillary necrosis is vascular thrombosis.

.(1) To prevent such complications care should be taken while making the incision and periosteal stripping.

Other reported rare complications with this procedure

are summarized in the table 3.

No recurrences occurring in the reported cases in this study could be attributed to the use of the technique.

JNA has an extensive vasculature, and in most of the cases it receives feeding vessels from external carotid system via the internal maxillary artery or the ascending pharyngeal artery, in certain rare cases as the proliferation of the tumor occurs, a collateral blood supply from the other blood vessels and structures take place this includes branches from the internal carotid artery or from the carotid system from the opposite side.(38)

Although the cases were limited in number, our findings suggest that intraoperative temporary clamping of the external carotid artery is a safe and effective procedure to facilitate surgery, as this helps reduce intraoperative bleeding. This is helpful particularly in a hospital that does not have interventional radiology facilities. The only drawback of using this surgical approach is that a longer operating time is required to explore for the branches of the external carotid artery (38).Our study demonstrates that this approach gives excellent access, exposure and cosmetic results.

CONCLUSION

Our study show strong evidence in favor of the lefort 1 osteotomy approach for JNA removal extending into pterygopalatine, infratemporal fossae, paranasal sinuses and base skull regions.Considering the advantages and rareness complications of this technique makes it an excellent choice for the treatment of JNAs without pre-surgical embolization.

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Table 1 : RADKOWSKI JNA CLASSIFICATION SYSTEM

STAGE	FEATURES
IA	Restricted to Nasopharynx
IB	Extension atleast into one paransal sinus
IIA	Nominal extension through sphenopalatine foramen, involves small part of medial pterygomaxillary fossa
IIB	Complete occlusion of pterygomaxillary fossa with Hollman -Miller sign, lateral or anterior displacement of maxillary artery branches, may have superior extension with orbital bone erosion
IIC	Extension through pterygomaxillary fossa into cheek, temporal fossa, or posterior to pterygoids
IIIA	Skull base erosion with minimal intracranial extension
IIIB	Skull base erosion with extensive intracranial extension, +/- cavernous sinus.

Table 1: Total No. of patients included in the study was 25 with age range of 9-20 years. Majority cases were of IIC, IIIa and IIIb stages as surgery was performed in collaboration with Neurosurgery department CHK

TABLE.2: PRESENTING SYMPTOMS	No. of PATIENTS
Epistaxis	8
Nasal Obstruction	5
Epistaxis + Nasal Obstruction	5
Facial Deformity due to Tumor Expasion	5
Ophthalmologic Symptoms	2
RADKOWSKI STAGE	
IA	0
IB	0
IIA	0
IIB	0
IIC	3
IIIa	6
IIIb	16
POSTOPERATIVE COMPLICATIONS	
Tumor Recurrence	2
Malocclusion	1

TABLE.3: Complications Reported regarding the use of lefort 1 osteotomy approach in JNA removal

Subcutaneous Emphysema	Stringer et al, 1979 (31)
unilateral abducens nerve palsy	Watts et al, 1984 (33)
decreased sensation in the upper lip	Ueki et al.,2008 (34)
aseptic necrosis of the maxilla	Lanigan et al., 1990 (28)
Fatal arteriovenous fistula	LaetitiaGoffinet et al., 2010 (35)
loss of eyesight	Cruz and dos Santos, 2006 (32)
Diplopia	Lin et al., 2008 (37)

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Author's contribution:

Shaheen Ahmed; concept, data collection, data analysis, manuscript writing, manuscript review
Muhammad Imran; data collection, data analysis, manuscript writing, manuscript review
Atiq Ahmed Khan; data analysis, manuscript writing, manuscript review
Anwar Ali; data analysis, manuscript writing, manuscript review
Syed Ijlal Ahmed; concept, data analysis, manuscript writing, manuscript review
Asim Rehmani; data analysis, manuscript writing, manuscript review
Muhammad Faaq Ali; manuscript writing, manuscript review