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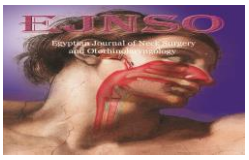

Exclusive Endoscopic Endonasal Transpterygoid Resection of Juvenile Nasopharyngeal Angiofibroma

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Abstract:

Objective: The objective of this study is to investigate the management outcome of the endoscopic endonasal transpterygoid approaches in patients with juvenile nasopharyngeal angiofibroma (JNA).

Patients & Methods: This is a prospective study carried out in the Otolaryngology Department, Cairo University Hospitals, Cairo, Egypt for 16 patients with JNA who underwent an endoscopic transnasal transpterygoid resection from January 2018 to October 2020. Postoperative evaluation including the clinical presentations, surgical findings and results, were included in the analysis. Follow up for all cases was done for 18–24 months.

Results: Sixteen patients were enrolled. All patients with JNA were staged according to Fisch classification, 56.3% of patients were in stage II and IIIa (extracrania), and 43.7% of patients were in stage III b and IV (intracranial). Wide MMA was sufficient for (50%) of cases. This corridor can be utilized in patients with stage IIIa in spite of extension of the lesion up to ITF. Our studies have confirmed the safety and efficacy of EETA approaches even in advanced cases. Single EETA (type A) was sufficient in only 2/16 cases of stage II (12.5%) while the combined EETA were done for the rest of the cases 14/16 (87.5%).

Conclusions: JNA may be successfully treated in most cases, even advanced ones, with expanded endonasal /endoscopy transpterygoid approaches after appropriate preoperative evaluation.

Key words: Angiofibroma - endoscopic endonasal transpterygoid approaches

Introduction

Juvenile nasopharyngeal angiofibroma (JNA) is a benign, vascular, locally aggressive tumour which originates in the sphenopalatine foramen. It originates at the pterygoid base and extends within the pterygopalatine fossa and adjacent compartments. The most common pathway in its extension is its lateral expansion into the pterygopalatine fossa (PPF), as it can have multidirectional extension anteriorly into the maxillary sinus, laterally to the infratemporal fossa through the pterygomaxillary fissure (PMF), posteriorly to the pterygoid fossa, and superiorly to the orbit, cranial fossa through the inferior orbital fissure. JNA can extend intracranially through the Vidian canal.[1-4]

There are different staging systems according to the tumour expansion to classify patients with JNA. Andrews-Fisch system is one of the most popular grading systems in use today:[5]

- Stage I is restricted to the nasal cavity/nasopharyngeal vault.
- Stage II comprises pterygopalatine fossa or any sinus extension.
- Stage IIIa includes infratemporal fossa or orbital extension,
- Stage IIIb includes intracranial, extradural extension into the parasellar region.
- Stage IVa includes intradural extension signifies more progressive disease.
- Stage IV b includes optic chiasm, pituitary fossa, or cavernous sinus invasion.

Surgical resection is the mainstay of treatment.[6] Radiotherapy or chemotherapy is recommended on some occasions, such as intracranial extension with cavernous sinus and carotid artery involvement.[7] Several surgical approaches have been recommended, currently, endoscopic approach is considered the standard approach for excision of JNA reaching the PPF with high rates of complete removal and low morbidity. But, many authors considered that any lesion with infratemporal fossa (ITF) or intracranial extension need external approach.[4,8]

With recent developments in the arena of endoscopic surgery which have allowed the surgeons to gain access to the foramen lacerum, petrous ICA, cavernous sinus, Meckel's cave, lateral nasopharynx and infratemporal fossa through the endoscopic endonasal transpterygoid approaches (EETAs).[9-13]

All EETAs need a corridor that passes through the maxillary sinus then by partial or complete removal of the pterygoid process (transpterygoid approach) to gain access to these areas.[9,11-14]

The endoscopic endonasal approach is less invasive than other conventional external approaches to access the anterior skull base with no retraction or manipulation of critical structures.[15-19]

Preoperatively the EETAs can be planned by vertical and horizontal lines that pass through the vidian canal and foramen rotundum (Fig 1). The petrous segment of ICA lies between the two horizontal lines while the PPF and the lateral recess of the sphenoid sinus (LRS) lie between the two vertical lines. The PPF lies beneath the horizontal line passing through the vidian canal while the LRS lies above this line. The sino-nasal tract (SNT) lies medial to the vertical line passing through the vidian canal while the middle cranial fossa (MCF) and the ITF lie lateral to the vertical line passing through the foramen rotundum. The middle cranial fossa lies above the horizontal line passing through the foramen rotundum, and the infratemporal fossa lies beneath the horizontal line passing through the vidian canal.[20]

The EETAs are classified into five major types according to the target area and the lesion extension (Fig 2):[20]

- **Type A** includes a partial resection (or thinning) of the lateral and/or medial pterygoid plates.
- **Type B** includes resection of the anteromedial part of the pterygoid base for lesions extending to the LRS above the vidian canal.
- **Type C** includes dissection of the vidian nerve to reach the petrous internal carotid artery with resection of the pterygoid base to access the petrous apex, cavernous sinus, or Meckel's cave.10,14,21,22
- **Type D** requires partial or complete resection of the pterygoid plates to reach to the ITF and upper parapharyngeal space.21
- **Type E** requires partial or complete removal of the pterygoid process, and the medial third of the eustachian tube (ET) to gain access to the lateral nasopharynx.

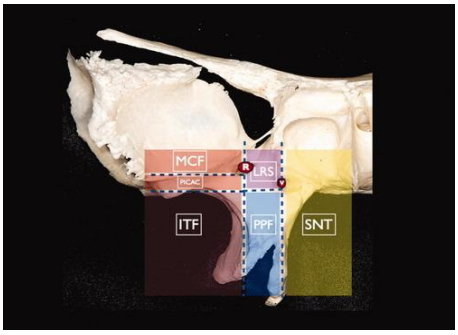

Fig 1: Vertical and horizontal lines passing through the foramen rotundum (R)and the vidian canal (V) dividing the EETAs field.[20]

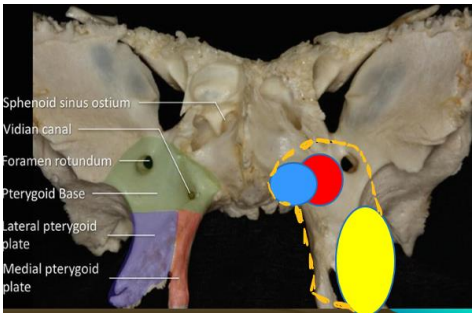

Fig 2: shows the five major types of EETAs. Type B (the blue circle), type C (the red circle), type D (the yellow area), type E (the dotted area)

Several reports about the recurrence rate of juvenile nasopharyngeal angiofibroma were done, that ranged from 0 to 57%.[23-27] Many authors considered the involvement of the foramen lacerum, cavernous sinus, and anterior cranial fossa, the large size tumor, the more progressive stage[28], intraoperative bleeding as predisposing factors for tumor recurrence.[29]

Aims and objectives:

The main objective of the study is to investigate the management outcome of the EETAs in juvenile nasopharyngeal angiofibroma (JNA) patients.

Material & methods:

The study was done as a prospective clinical trial which carried out for 16 patients with JNA who underwent an endoscopic EETA from January 2018 to October 2020 in the Otolaryngology Department, Cairo University Hospitals, Cairo, Egypt following institutional and ethical review board approval. Patients included in this study underwent purely EETA to pterygopalatine fossa, sphenoid sinus, ITF, petrous apex, cavernous sinus and Meckel's cave.

Preoperative assessment and staging workup was done preoperative to confirm the resectability of the lesion by an EETA. High resolution computerized tomography of the nose and paranasal sinuses (HRCT) with intravenous contrast and contrast enhanced Magnetic resonance imaging (MRI) for assessment of lesion site, size, extension and its relation to adjacent important structures such as optic nerve, ICA, cavernous sinus, type of sphenoid sinus pneumatization, position of vidian canal and foramen rotundum. Angiography and embolization were done 24 to 48

hours before surgery. These cases were classified according to Andrews-Fisch system.[5]

Pre-operative preparations:

- The three dimential CT images, angiography and MRI images have been incorporated into the surgical navigation system to identify the critical vessels.
- Type and cross matching for packed RBCs.
- Preoperative wide spectrum prophylactic antibiotics were administrated in the form of third-generation cephalosporin that can cross the blood brain barrier.

Treatment strategy and operative techniques:

Under general anesthesia with controlled hypotension, the surgical procedure was modified according to the lesion extension as follows:

1- Initial steps:

The endoscopic endonasal approach begins by making a nasal and an antral corridor before gaining access to the PPF. The procedure begins with medialization of middle turbinate to acess the osteo-meatal complex. Standard FESS is done with uncinectomy, ethemoidectomy and wide meatal antrostomy to widely expose the posterior wall of the maxillary sinus (PWMS). In selected cases, posterior septectomy was done to gain more lateral control with passing the surgical tools from the contralateral side.[30]

2-Sino-nasal antral corridor:

The surgical window to access the PPF was modified according to the site and lesion extension as following:

1- For lesions restricted superior to the horizontal line crossing the sphenopalatine foramen, a wide maxillary antrostomy was sufficient for exposing and resection the superior part of the PWMS medial to the infraorbital nerve (ION). This exposure provided access to the pterygoid base, foramen rotundum, vidian canal, LRSS, petrous apex and Meckel's cave.[20] (fig3)

2- when the lesion is inferior to this horizontal line, the surgical corridor was expanded by removal of the vertical plate of the palatine bone and the posterior part of the medial maxillary wall (a medial maxillectomy).[11] Prelacrimal recess approach provided a wide exposure from the posterolateral part of the maxillary sinus to the LRSS.[31,32] (Fig 4)

3- In selected cases, if more lateral acess to the infratemporal fossa was needed, a Denker's approach was performed by removing the pyriform aperture and the anterior wall of the maxillary sinus until the lateral maxillary wall can be seen.[33] (Fig5)

3.Approach to PPF:

After identification of the sphenopalatine foramen and the ION passing through the maxillary sinus roof, the posterior antral wall was removed as much as needed by Kerrison rongeurs, from medial to lateral direction.[34] (Fig 6)

The neural structures in the pterygopalatine fossa (the Vidian nerve, the maxillary nerve, and the palatine nerves) were dissected from the tumor. During the surgical dissection, these nerves were preserved to minimize the postoperative morbidity.[35]

While dissecting in the medial part of the pterygopalatine fossa, the sphenopalatine artery (SPA) can be found passes through the sphenopalatine foramen. The vidian nerve can be found at the junction of the medial pterygoid plate and the floor of the sphenoid sinus and slightly superior and lateral to it the maxillary nerve entering foramen rotundum.[36] (fig 7)

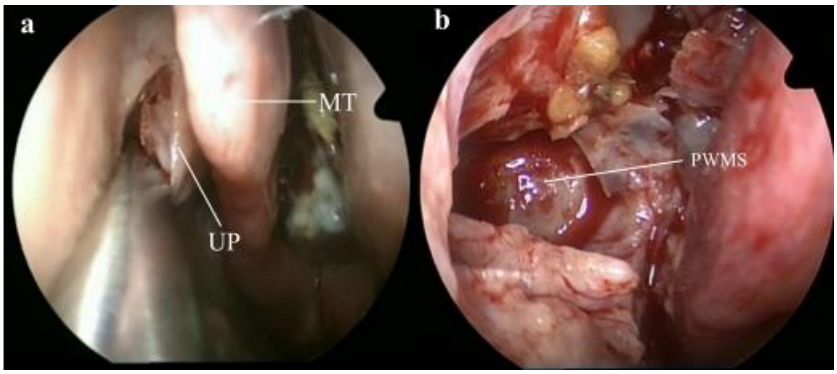

Fig 3: Endoscopic view showing wide maxillary antrostomy which extended from the nasolacrimal duct to the posterior wall of the maxillary sinus and from the orbital floor to inferior turbinate. Wma is sufficient for exposure of the upper half of the PWMS.

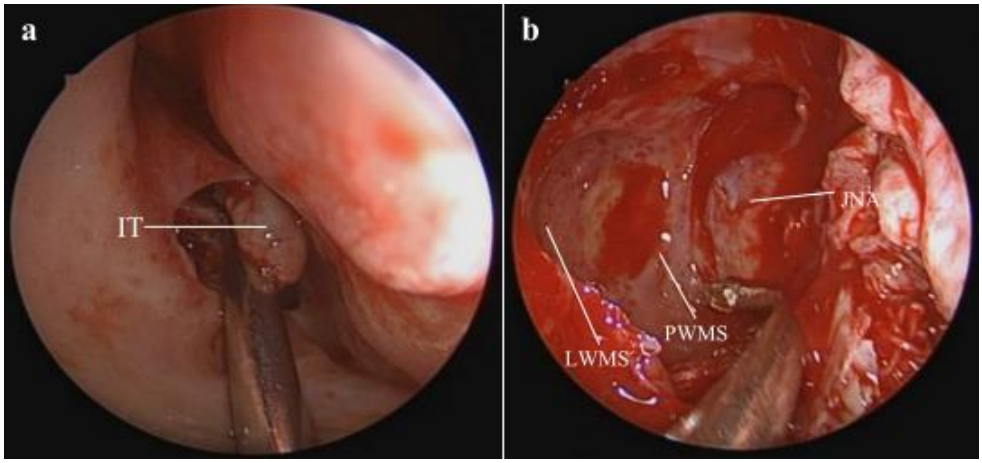

Fig4a: Endoscopic prelacrimal approach in a case with JNA: a) Incision is made anterior the inferior turbinate where it turns inferiorly down to the pyriform aperture. b) After medial maxillectomy, through a pre-lacrimal approach, until the posterior and lateral wall of the maxillary sinus are visualized.

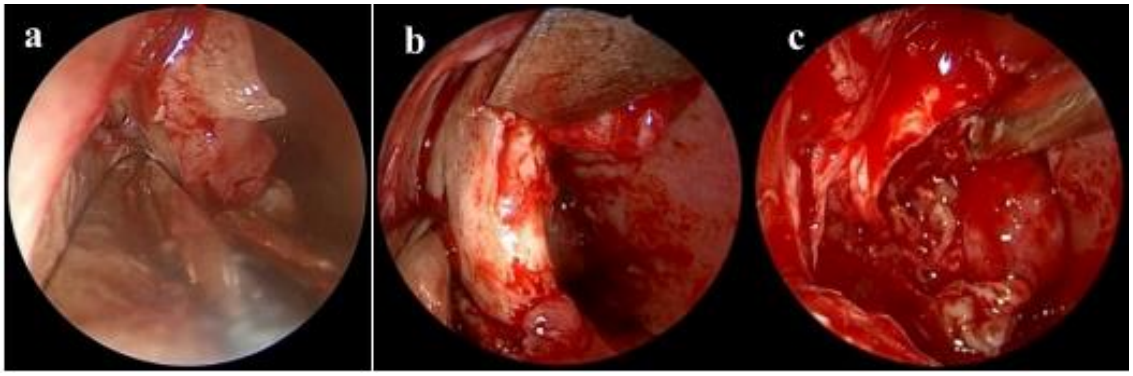

FIG5a, b, c: Endoscopic Denker's approach: In a case with JNA the the pyriform aperture and the anterior wall of the maxillary sinus until the lateral maxillary wall can be seen.

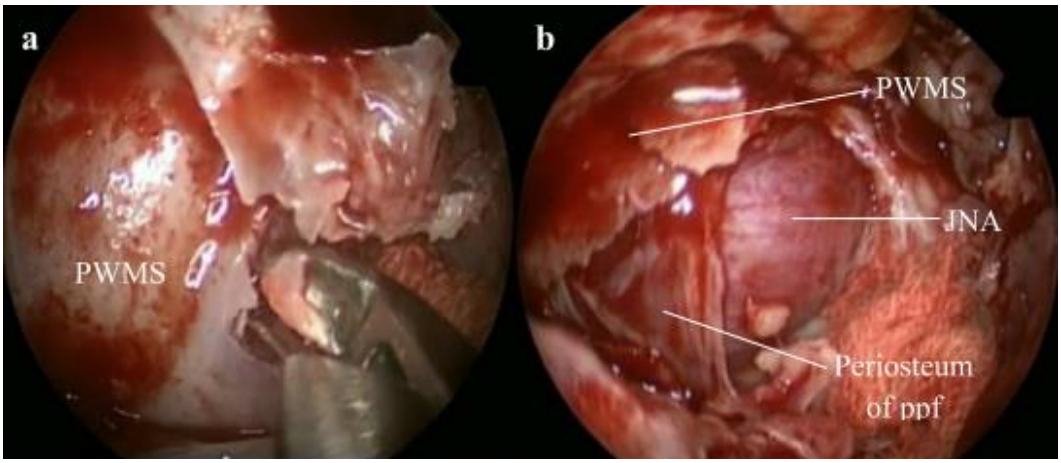

FIG 6: A) Endoscopic view of a case with JNA showing the removal of PWMS by Kerrison rongeur from medial to lateral direction. B) After removal of PWMS, the periosteum is seen covering the tumour.

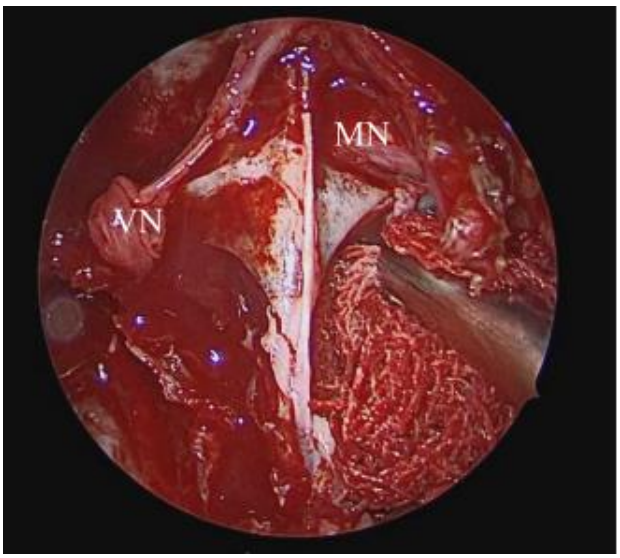

Fig 7: Endoscopic view showing maxillary nerve (MN) entering foramen rotundum and slightly inferior and medial to it the vidian nerve (VN) entering pterygoid canal.

C) Limiting bone (transpterygoid dissection):

EETA type A was done in cases with extention of the lesion only to the PPF by partial removal (or **thinning**) of the pterygoid process.[20]

EETA type B was done in cases with sphenoid sinus extension. The **pterygoid base** was drilled out and the pterygoid plates were left intact. Sphenoidotomy was expanded laterally by removing the anterior sphenoidal wall above the line crossing the vidian canal.

EETA type C was done in cases extends to the area of petrous ICA or cavernous sinus.[20] The risk in an EETA in these cases is mainly to the petrous and lacerum part of the ICA. [9,36-37]

Endoscopically, the vidian nerve can be seen within the PPF and seen entering its canal at the junction between the medial pterygoid plate and the floor of the sphenoid sinus and was used as a key landmark to the petrous part of the ICA (Fig.8).[10, 36,38]

Type C includes removal of **the pterygoid base** to acess the petrous apex, cavernous sinus, or Meckel's cave.[10,14,21,22] Additional bone was removed from the pterygoid base and

the superior part of the medial pterygoid plate to expose foramen lacerum, that can be identified by its cartilaginous part. The foramen lacerum was widely opened carefully to avoid injury of the lacerum portion of the ICA (Fig.9).[39]

EETA type D was done in these cases that extend **ITF.** The posterior antral wall was resected to the extent needed, from medial to lateral direction, by kerrison rongeur, to fully expose periosteum of the PPF and the ITF.[20] With more posterior dissection, the lateral pterygoid muscle insertion on **the lateral pterygoid plate** can be seen, the lateral pterygoid muscle can be dissected from its medial insertion if needed to enhance the tumour removal (Fig.10).[16]

The feeding branch from maxillary artery was divided. Then, the vidian neurovascular bundle was divided just distal to the vidian canal to expose the entire length of the pterygoid process. (Fig 11) EETA **Type D** involved variable resection of the pterygoid **plates** to reach the ITF. However, the resection of **the entire** pterygoid process (EETA Type E) may be indicated in extensive lesions reaching the ITF with or without involvement of the middle cranial fossa.[20]

To prevent the recurrence of JNA, drilling of the cancellous bone of the pterygoid root and basisphenoid was done in all cases, especially around the vidian foramen, to remove any residual lesion that may not be immediately evident.[40]

All the patients in this study were followed up for 18–24 months.

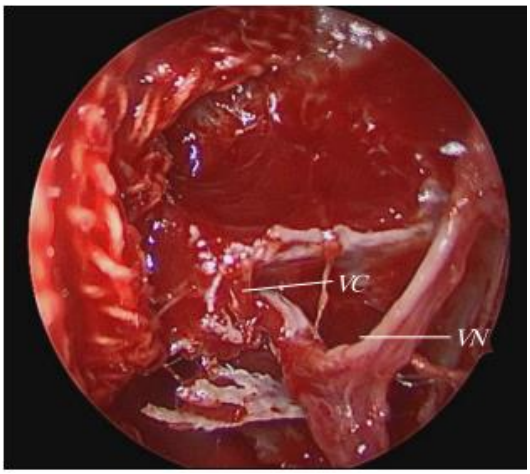

FIG 8: Endoscopic view shows the vidian nerve (VN) passing through its canal (VC) which was used as key landmark to the petrous part of the ICA

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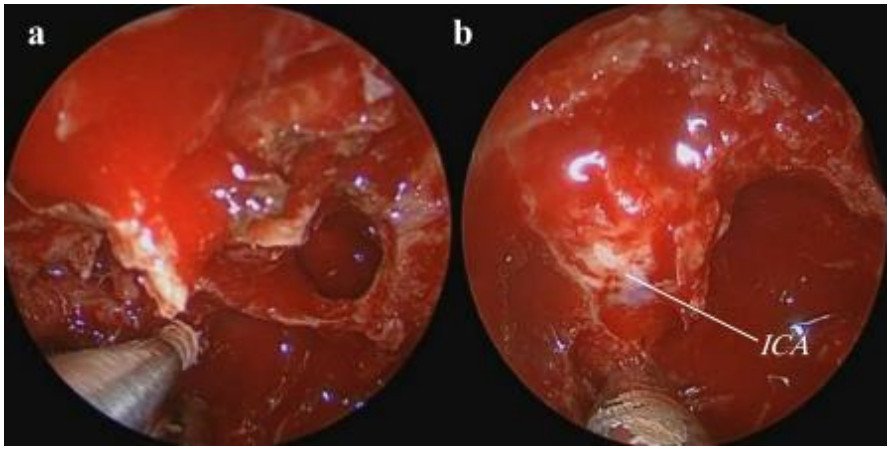

FIG 9: Endoscopic view with (a) partial resection of the pterygoid base by electric drill and dissection of vidian nerve until the **petrous** ICA can be seen (b).

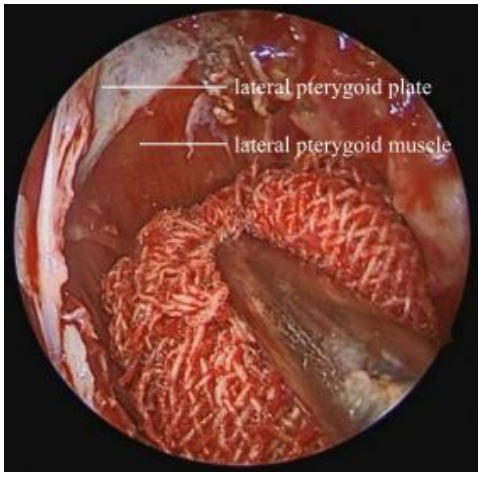

FIG 10: Endoscopic view showing attachment of lateral pterygoid muscle to the lateral pterygoid plate.

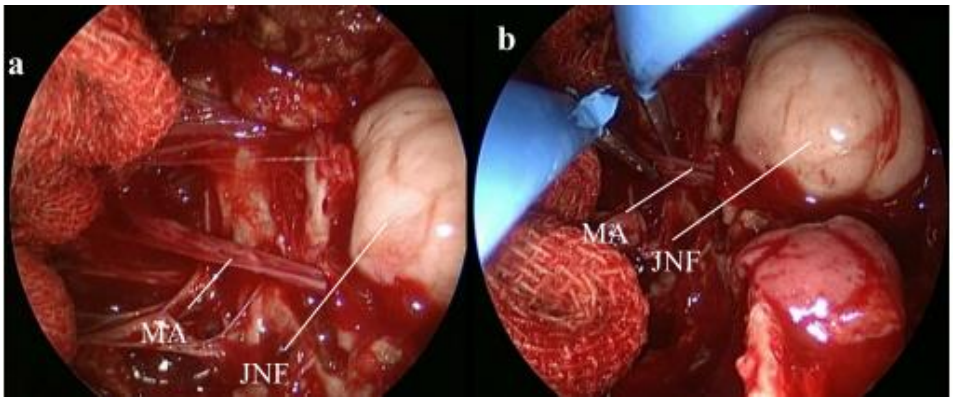

FIG 11: Endoscopic view of a case with JNA showing (a) feeding branch of maxillary Artery (MA) to the tumor which was identified and then (b) divided to allow removal of the tumor.

Results:

All the 16 JNA cases were exclusively males and mean age was 17 ± 4.76 years with range between 8 and 27 years. Majority 13 (81.3%) of them had right affected side, while 3 (18.8%) patients presented with left side tumor. Nasal obstruction was present in 16 (100 %) patients. Fifteen (93.8%) patients experienced recurrent bouts of spontaneous epistaxis.

Preoperative radiological assessment revealed that in additional to the nasal cavity, PPF and nasopharynx were involved areas in all these 16 patients with JNA (100%) while ITF, sphenoid sinus and petrous-ICA, cavernous sinus were involved areas in 11 (68.8%), 14 (87.5%) ,6 (37.5%) and 2 (12.5%) patients, respectively. (Table 1) (Fig 12, 13)

All the patients with JNA were staged according to Fisch classification. Five (31.25%) patients were in Fisch stage II, 4 (25%) patients were in stage IIIa, 5 (31.25%) patients were in stage IIIa whereas 2 (12.5%) patients were in stage IV b. (Fig 14, 15, 16)

The Used Corridor among JNA Patients:

Wide MMA was the most frequent utilized corridor in 8 patients with JNA (50%). Medial maxillectomy corridor with or without modified Denker's was utilized in 8 (50%) patients (Table 2).

In 5 patients with stage II, the wide MMA only was done and this corridor was frequently utilized in patients with stage III a (50%). Medial maxillectomy with or without modified Denker's corridor was needed in 6/8 patients stage III & IV (75%). (Table 3)

The surgical EETA among JNA patients:

Single EETA (type A) was sufficient in only 2/16 cases of stage II (12.5%). The combined EETA were done for the rest of the cases 14/16 (87.5%). The combined EETA type A, B were utilized in in another 3 cases (4/16) with stage II and the combined EETA type A, B, D were utilized in in all cases (4/16) with stage IIIA. The combined EETA type B, C, D were used in all advanced cases (7/16) with stage III b and IV b. (Table 4)

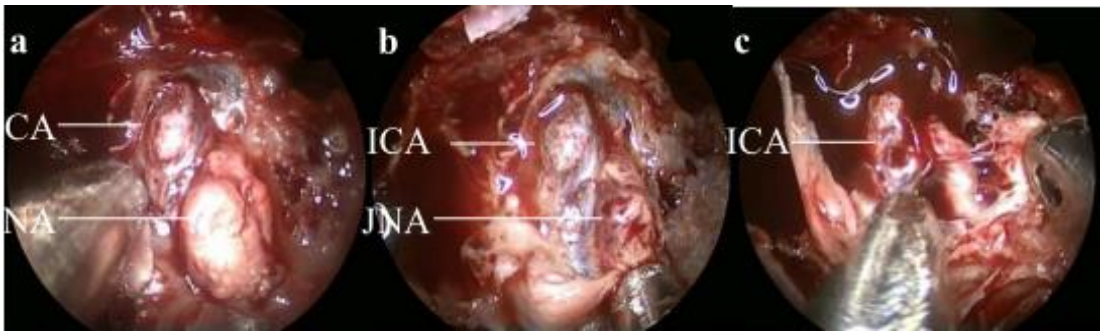

FIG 12: Endoscopic view showing a case with JNA (a,b) attached to the petrous carotid (c) ICA after removal of the tumor.

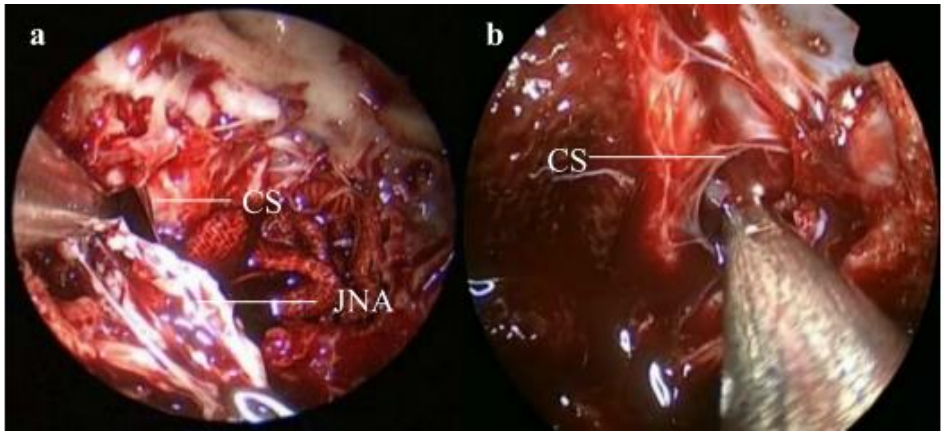

FIG 13: Endoscopic view of case with JNA showing: A) the removal of part of tumor invading the cavernous sinus (CS). B) After removal of the tumor.

Table 1: Involved compartments in CT & MRI among JNA patients =16

Involved compartments	NO	%
Nasal cavity	16	(100%)
PPF	16	(100%)
Nasopharynx	16	(100%)
Sphenoid sinus	14	(87.5%)
ITF	11	(68.8%)
Petrous-ICA	6	(37.5%)
Cavernous sinus	2	(12.5%)

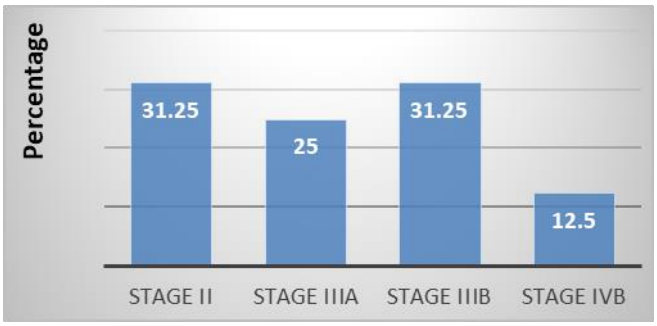

Fig 14*:* Tumor stages according to Fisch classification among enrolled patients

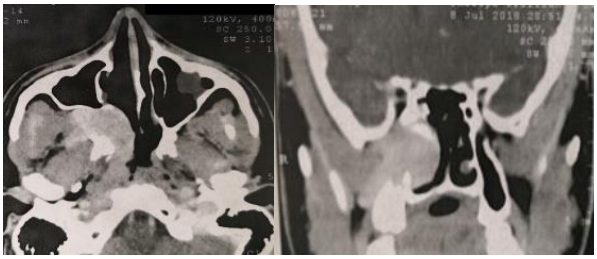

Fig15: CT image showing angiofibroma shows a mass in right side of the nasal cavity and a grossly enlarged sphenopalatine fissure to reach PPF and extended to ITF. The posterior wall of the right maxillary sinus is bowed anteriorly creating the classic Holman-Miller sign. The right pterygoid is partially destroyed involving pterygopalatine fossa, infra temporal fossa and nasal cavity. (Stage IIIA)

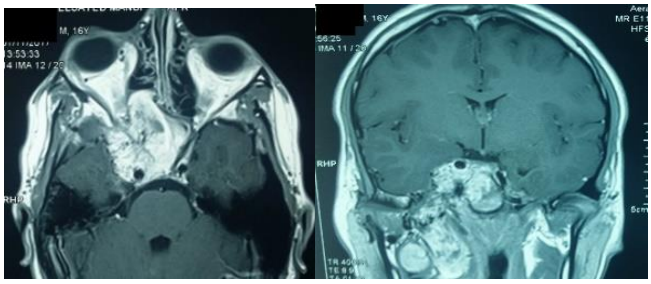

Fig16: MRI images of a case with JNA in right side of the nasal cavity, PPF, ITF, nasopharynx and bilateral side of sphenoid sinus with intracranial extension encasing the right ICA and invading right cavernous sinus. (Stage IV B)

Table 2: The Surgical corridor among patients with JNA:

Antral corridor	N= 16
Wide MMA	8 (50%)
Medial maxillectomy	6 (37.5%)
Medial maxillectomy& modified Denker's	2 (12.5%)

Table 3: JNA stage (based on Fisch classification) and the type of used corridor:

	Wide MMA	Medial maxillectomy+/- modified Denker's	Total
Stage II	5/5 (100%)	0/5	5
Stage IIIa	2/4 (50%)	2/4 (25%)	4
Stage III b	0/5	5/5 (20%)	5
Stage IV b	1/2 (50%)	1/2	2
Total	8	8	16

Table 4: JNA stage and the type of used surgical EETP approach:

EETA STAGE	Type A=2	Type AB=3	Type A, B, D=4	Type B, C, D=7
II=5	2	3	0	0
III a=4	0	0	4	0
III b=5	0	0	0	5
IV b=2	0	0	0	2

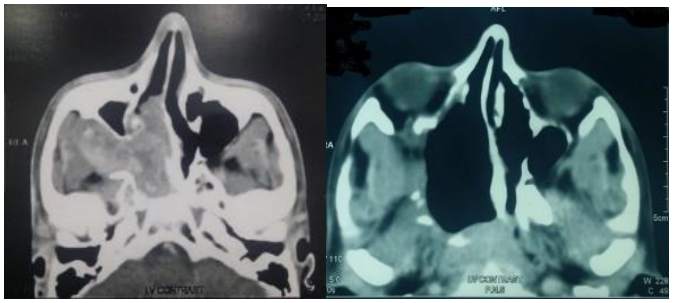

FIG 17: CT imaging of (a) a case with JNA of **stage IIIa** involving PPF, ITF, nasal cavity. (b) Post-operative imaging with no recurrence.

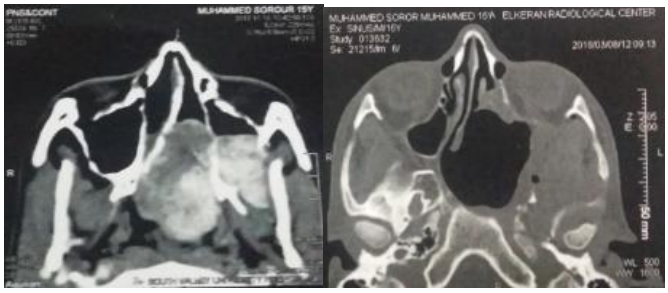

FIG 18: CT imaging of (a) a case with JNA of **stage IIIa** involving PPF, ITF, nasal cavity. (b) Post-operative imaging with no recurrence.

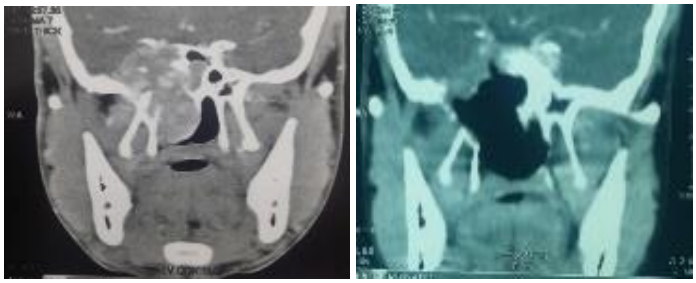

FIG 19: CT image showing (a) JNA with cavernous sinus involvement (**stage IV b**).
(b) Post-operative imaging with no recurrence

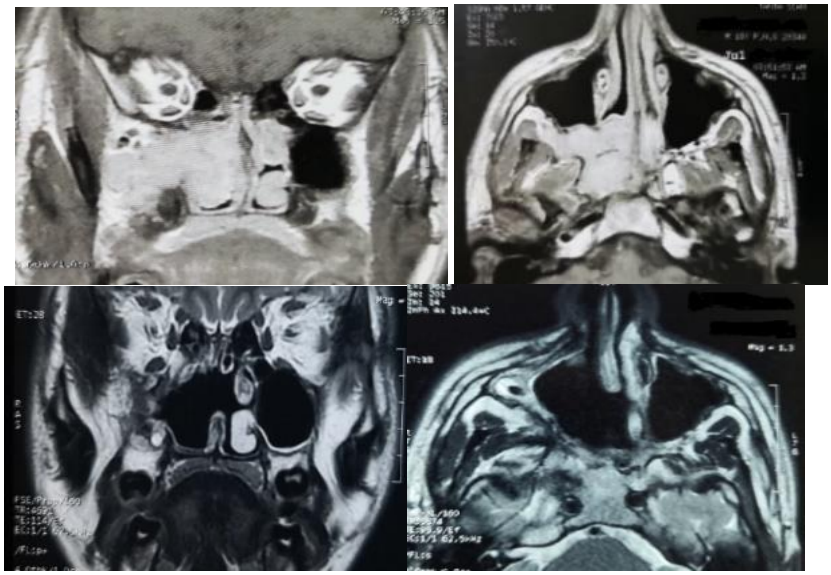

FIG 20: MRI of case with JNA: pre-operative imaging shows the tumor involving right side of nasal cavity, PPF, ITF, sphenoid sinus closing to ICA (**stage IV b**). Post-operative imaging shows no recurrence with EETA with posterior septectomy to aid in removal of the lesion.

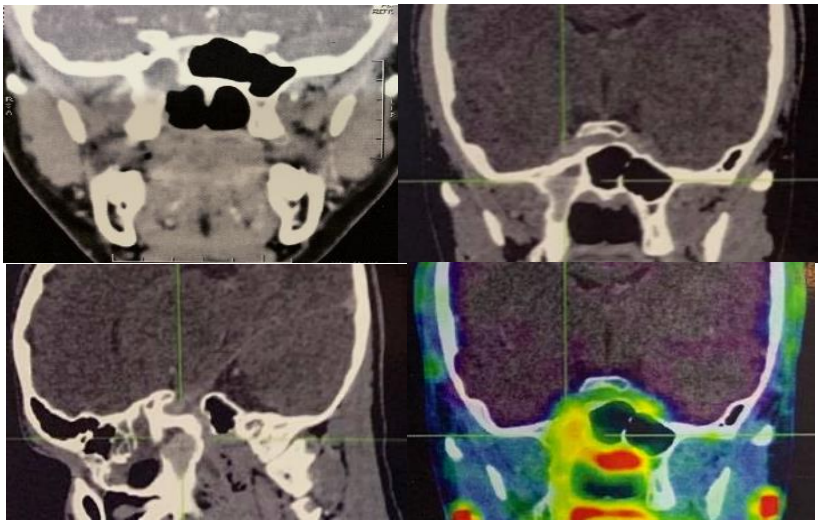

FIG 21:Post-operative PET/CT image of a case with JNA showing recurrence of the tumor in the right pterygoid base.

Discussion :

Nicolai et al. 2010 described a successfully managed juvenile nasal angiofibromas by purely endoscopic resection.[41] The Surgical advancement in the endoscopic field have revolutionized the management of juvenile angiofibroma, allowing the surgeons to improve surgical outcomes for extensive lesions. [42]

In our study, the majority of patients presented initially with nasal obstruction followed by epistaxis (100 % & 93.8% respectively) and the most frequent sign was nasal mass (100%) followed by proptosis (25%) which coincides with the findings of Allan et al (2020).[43]

In our study, 100% of JNA cases had involvement of nasal cavity, pterygopalatine fossa choanal and nasopharynx, while sphenoid sinus, infratemporal fossa, petrous-ICA, and cavernous sinus were involved areas in 87.5%, 68.8%, 37.5% and 12.5% patients, respectively. This is in agreement with that of **Allan et al (2020)** who found that all of the JNA cases had involvement of choana and nasopharynx, distorted pterygoid plates, and 50% and 25% patients had nasal septal displacement, and erosion of medial maxillary wall, respectively. [43]

In our study, 31.25% of patients were in stage II, 25% of patients were in stage IIIa, and 31.25% patients were in stage IIIa whereas 12.5% patients were in stage IV b. **Devang et al (2020)** reported earlier stages in their study, 32% patients were in stage II, and 36% patients were in stage IIIa, whereas 32% patients were in stage III b. [42]

In our study, for patients with JNA Wide MMA was the most frequent utilized corridor in (50%). This corridor was efficiently used not only for all patients with stage II, but also could be utilized in 2 patients with stage III a and one patient with stage IV b in spite of extension of the lesion up to ITF with no recurrence or major morbidity or any late serious complications. Medial maxillectomy with or without modified Denker's corridor was done in (8/16) cases (50%) with stage III & IV. **Devang et al (2020)** stated that the modified Denker's procedure only is an effective approach for excision of juvenile angiofibroma. They added that this approach was used up to Fisch stage III b without major recurrence or any serious complications. [42]

In our study, in additional to the endonasal transantral approach, trans-

sphenoid assisted approach was used in only 2(12.5%) of cases of JNA. [44]

Different EETAs were performed based on precise preoperative surgical mapping. In JNA cases, 14/16 cases (87.5%) were managed via combined EETAs. The combined EETA type B, C, D approaches were used in all advanced cases (7/16) with stage III b and IV while the combined EETA type A, B, D approaches were utilized in (4/16) patients with stage IIIa and the EETA type AB approaches were utilized in 3 out of 4 patients with stage II. However, others stated that lesions with ITF or intracranial extension need external approach that is technically more difficult. [4,8]

In our study, drilling or thinning of the pterygoid wedge in JNA was done to reduce the incidence of residual and recurrent disease. The clinical and radiographic follow up revealed that the incidence of recurrence was 18.8% of cases of JNA.

Janakiram et al (2017) and **Liu et al (2019)** found that the pterygoid wedge, followed by the lateral part of the ITF are the most common site for recurrence. [45-46] They added that a key element to any successful removal must involve drilling of the cancellous bone of the pterygoid base, especially around the vidian canal, which is the most common area of residual lesion (58%) and recurrence (45%), but the residual/recurrence rates decrease markedly (3.07%) when the pterygoid base was drilled. [45]

Conclusion:

The endoscopic transpterygoid approach is a minimally invasive approach to gain access to pterygopalatine fossa, infratemporal fossa, petrous apex, lateral recess of sphenoid, and other regions of the paramedian skull base with using the vidian canal and foramen rotundum as landmarks to plan the EETA.

Declaration of interest:

There are no interests to declare

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