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Evaluation of the Impact of Preoperative Transarterial Particle Embolization of Juvenile Nasopharyngeal Angiofibroma on the Surgical Outcome on 30 Egyptian Patient.

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Abstract: Background: Juvenile nasopharyngeal angiofibroma (JNA) is a highly vascular benign tumor with potentially fatal intraoperative haemorrhage. The aim of this study was to evaluate the impact of preoperative embolization of JNA on the surgical outcome including intraoperative blood loss, transfusion and endoscopic feasibility. **Results:** Preoperative embolization used PVA particles in all cases and resulted in lowering the mean intraoperative blood loss and transfusion to 665 and 928.57 ml respectively. Pure endoscopic resection was used in 19/30 (63.3%) patients. There was a significant correlation between blood loss and endoscopic use, residual tumor blush and internal carotid artery supply to the tumor. No major, but only minor post-operative complications were encountered. **Conclusions**: Preoperative embolization of nasopharyngeal angiofibroma is a safe and effective technique to reduce intraoperative blood loss and facilitate endoscopic use. Angiography enables better surgical planning. Minor complications may be encountered, but major complications are rare if done using the correct technique.
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1. Introduction:

Juvenile nasopharyngeal angiofibroma (JNA) is a highly vascular benign nasopharyngeal tumor with a locally destructive behaviour. Although rare accounting for only 0.5% of all head and neck tumors, it is considered the most common benign nasopharyngeal neoplasm. JNA typically affects adolescent males with a mean age at presentation of 15 years [1,2].

The site of origin of JNA has been debatable, however currently it's thought to originate from the postero-superior part of the sphenopalatine foramen, an important foramen connecting the nasal cavity to the pterygopalatine fossa. The tumor classically spreads via the natural anatomical connections and foramina of the pterygopalatine fossa into the nasal cavity, paranasal sinuses, infratemporal fossa, orbit and intracranially [1,3].

Affected patients usually present with nasal obstruction and recurrent epistaxis early in the disease [4]. Facial deformity, cranial nerve affection or proptosis may develop later with advanced disease progression [5].

Cross sectional imaging modalities play a major role in the JNA diagnosis, staging and preoperative planning. Typically a mass is seen centred upon the sphenopalatine foramen with widening of the pterygopalatine fossa. A characteristic forwards bowing of the posterior wall of maxillary sinus may be seen and is known as "Holman-Miller sign". Vivid enhancement is seen after IV contrast injection. Magnetic resonance imaging (MRI) is superior to computed tomography (CT) for evaluating the extent of skull base, intracranial, intraorbital and bone marrow invasion [6,7].

JNA is supplied by ipsilateral branches of the external carotid artery (ECA), mainly the internal maxillary (IMax) and ascending pharyngeal branches. With further tumor growth more arterial feeders are recruited from the ipsilateral or contralateral internal and external carotid arteries [1,8].

Digital Subtraction Angiography (DSA) is helpful in preoperative planning to detect the supplying vessels and preoperative embolization can be performed in the same setting.

The characteristic angiographic finding is tumoral blush noted in the arterial and capillary phases [1,9].

Multiple staging systems for JNA have been proposed and most are based on tumor extension. The most widely used now are those proposed by Radkowski et. al. (1996) [10] and the University of Pittsburgh Medical Centre (UPMC) staging system proposed by Snyderman et. al. (2010), Table 1[11]. The UPMC staging is the most recent and is unique in adding the residual tumor blush and direction of spread relative to internal carotid artery (ICA) as prognostic factors [12], **Table 1.**

Currently surgery is the method of choice for the definitive treatment of JNA. The high risk of intraoperative bleeding and the extensive approaches needed for tumor resection have been the main obstacles against surgery. Radiotherapy is now reserved for non-operable cases or postoperative cases with residual intracranial parts. Other treatment options, including hormonal and chemotherapy were used in the past but with controversial results [13,14].

The purpose of this study was to evaluate the impact of preoperative embolization of juvenile nasopharyngeal angiofibroma on the surgical outcome.

2. Methods:

In this study, we retrospectively analysed the clinical, imaging, angiographic and operative data of 30 male patients with clinical, radiological and histopathological proven JNA, at our institution during the period from May 2018 to May 2019. The embolization procedures were carried out at the Interventional Radiology Unit, Radiology Department, while surgical procedures were done at the Otorhinolaryngology Department of our institution. The study was carried out after obtaining ethical committee approval and an informed consent from each patient.

Method of preoperative angiography and embolization:

The procedures were performed under either general anaesthesia (n=19) or conscious sedation (n=11). Femoral arterial access was obtained using Seldinger technique and a 5- or 6-French (Fr) femoral sheath was secured in place. A 5-Fr Bern® (SE, Boston Scientific, Natick, MA, USA) guiding catheter was used to perform bilateral common carotid, external carotid and internal carotid angiography utilizing digital subtraction techniques (DSA).

The feeding vessels of the tumor were identified by the presence of tumoral blush, vessel hypertrophy and larger number of branches leading to the tumor. Super-selective catheterization and angiography of the tumor supplying ECA branches was then performed using a 2.9-Fr Renegade® (SE, Boston Scientific, Natick, MA, USA) microcatheter. Careful analysis of the angiographic anatomy was performed to exclude vascular anomalies especially abnormal origin of the ophthalmic artery or anastomosis to the intracranial circulation.

Embolization of the tumor feeding vessels was then performed by placing the microcatheter as close as possible to the tumor and the embolic material was then injected. Injection was done in short pulses, synchronized with cardiac systoles to avoid reflux of embolic material into proximal vessels. Embolic materials used were polyvinyl alcohol (PVA) with sizes of (250-355 µm) and (355-500 µm). Post embolization angiography was then performed to confirm stasis of contrast in the feeding vessel and absence of tumor blush from it.

Evaluation of tumor devascularization:

The percent tumor devascularization and residual blush were determined subjectively by 3 different observers by comparing the pre and post embolization angiograms and graded as low (0-9%), moderate (10-19%) and high (20-29%). The remaining feeding vessels of the tumor were also determined and recorded.

Evaluation of the impact on surgery:

The amount of intraoperative blood loss, estimated by the operating surgeon from the suction tubes and soaked dressings, was determined for each patient from the surgical sheet, together with the amount of intraoperative blood transfusion and type of operative approach used for tumor resection.

Statistical analysis:

Statistical analysis of the collected data was done using SPSS 23 (IBM corp., Chicago, IL) software. Descriptive statistics were calculated for different datasets. Several comparisons were made between the intraoperative blood loss in categories with different criteria using Student's T-test for normally distributed data and Mann-Whitney U test for non-normal distributed data. Several correlations were made between intraoperative blood loss and tumor stage, residual tumor blush, and operative approach used using either Pearson or Spearman's Rank correlations according to the type of data. In all cases, ap-value of <0.05 was considered statistically significant, and <0.01 was considered highly statistically significant.

3. Results:

All patients were male, with a mean age of 17.1 years (range 10-33 years). 7/30 patients (25%) were at stage I, 5/30 patients (15%) at stage II, 1/30 patient (5%) at stage III, 11/30 patients (35%) at stage IV and 6/30 patients (20%) at stage V_M. The most common presenting symptoms were epistaxis (86.7%) and nasal obstruction (80%), **Figure 1.**

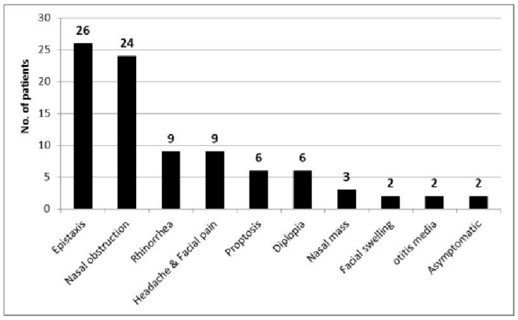

Figure 1: Symptoms of the studied patients.

The procedure was done under general anesthesia in 19 patients (63.3%) and under conscious sedation in 11 patients (36.7%). The angiography and embolization procedures were technically successful in all patients (100%).

PVA 355-500 μm was used in all patients (100%), in addition to PVA 250-355 μm which was used in 16/30 patients (53.3%). Total embolization procedure duration had a mean of 56.5 ± 28.9 minutes (range 25 – 120 minutes), while the total fluoroscopic duration had a mean of 31.6 ± 16.2 minutes (range 16 – 69 minutes). Embolization was done at a mean of 35.4 ± 14.81 hours before surgery (range 24-72 hours).

In 13/30 patients (43.3%) the tumors were supplied by branches of the ipsilateral ECA only. While in 17/30 (56.7%) the ICA was additionally contributing to the tumor's vascular supply, **Table 2.**

Table 1: Pattern of vascular supply of JNA encountered in the study.

Pattern of vascular supply	No. (%)
Ipsilateral ECA only	13 (43.3%)
Bilateral ECA + Bilateral ICA	8 (26.7%)
Ipsilateral ECA + Ipsilateral ICA	6 (20%)
Bilateral ECA + ipsilateral ICA	3 (10%)
Bilateral ECA supply	0

The post-operative residual tumor blush was found low in 15 patients (50%), moderate in 10 patients (33.3%), and high in 5 patients (16.7%).

No reported intra-procedural complications in any of the patients. Neither any of the patients encountered any neurological complications. However, minor post procedure complications were reported in 9/30 patients (30%). These were treated medically with no need for surgical intervention or leaving permanent sequel, **Table 3.**

Table 2: Complications encountered post-procedure:

Complications	No. (%)
Groin pain	3 (10%)
Facial pain	3 (10%)
Headache	2 (6.7%)
Puncture site hematoma	1 (3.3%)

Concerning the surgical approach used, 19/30 patients (63.3%) underwent pure endoscopic procedures, 6/30 patients (20%) underwent open surgery while 5/30 patients (16.7%) underwent combined endoscopic plus open surgery, **Table 4.**

The intraoperative blood loss for all procedures had a mean of 665 ml ± 464.55. Procedures performed using endoscopy had a mean blood loss of 388.46 ml ± 251.78, while procedures performed using open surgery had a mean of 1275 ml ± 340.34. Procedures where combined open and endoscopic approaches were used had a mean of 1050.33 ml ± 180.27. There was a highly significant negative correlation between endoscopic use in JNA resection and the amount of intraoperative blood loss ($r = -0.674$, $p=0.001$), **Figure 2.**

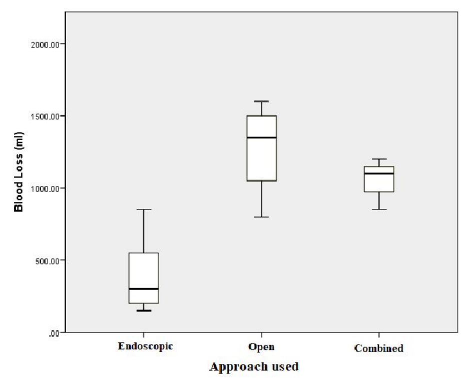

Figure 2: Intraoperative blood loss for different surgical approaches used in tumor resection.

Table 3: Operative approach used for the studied cases

Approach	No (%)
➢ **Endoscopic Surgery**	19 (63.3%)
➢ **Open Surgery (Midfacial Degloving + Craniotomy)**	6 (20%)
➢ **Combined (Endoscopic + Open)**	5 (16.7%)
• **Endoscopic + Midfacial Degloving**	3 (10%)
• **Endoscopic + Lateral Rhinotomy**	2 (6.7%)

Additionally, there was a highly significant correlation between the intraoperative blood loss and ICA contribution to tumor supply (p= 0.001), were tumors with both ECA and ICA supply had a mean blood loss of 1013.63 ml versus 238.89 ml tumors with ECA supply only. Another highly statistically significant correlation was found between the tumor's UPMC stage and the corresponding intraoperative blood loss (r=0.895, p<0.001).

Lastly, a highly significant correlation was found between the intraoperative blood loss and the residual tumor blush (r=0.905, p=<0.001).

Blood transfusion was required in 9/30 patients (30%) with a mean of 928.57 ml ± 345.03 and range 500-1500 ml. None of stage I, II or III patients required blood transfusion, while 3 patients at stage IV had a mean transfusion of 833.33 ml and 6 patients at stage V had a mean transfusion of 1000 ml, Figure 3.

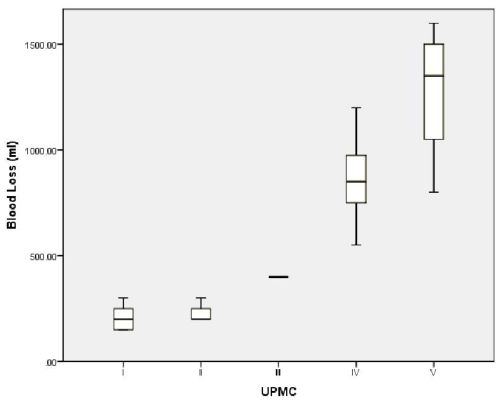

Figure 3: Relation of UPMC and the intraoperative blood loss.

Additionally, none of the patients operated via endoscopy required intraoperative blood transfusion, while 6 patients with open surgery required a mean blood transfusion of 1000 ml and 3 patients with combined open and endoscopic surgery had a mean blood transfusion of 833.33 ml.

4. Discussion:

JNA is a highly vascular benign tumor with a locally aggressive behaviour, affecting young adolescents with distinct male prevalence. It arises in the postero-lateral wall of the nasopharynx near the sphenopalatine foramen. The tumor then extends to involve the nasal cavity, paranasal sinuses, infratemporal fossa, orbit and intracranially [6].

The bloody nature of the tumor and the young age of patients poses a challenge during its surgical removal to avoid potentially fatal haemorrhage, extensive dissections and unintended injury to vital structures. Management thus requires proper preoperative evaluation, control of intraoperative bleeding and appropriate choice of surgical approach [15]. Modern imaging techniques have permitted accurate delineation of tumor extension and invasion of nearby skull base and intracranial structures. Contrast enhanced CT and MRI are now considered a gold standard for preoperative JNA evaluation especially in cases with intracranial extension [16].

Despite the drawbacks of surgery, yet it remains the primary modality for treatment of JNA. Surgical techniques have been developed in the last few decades with endoscopy replacing most of traditional techniques in the management of JNA. Endoscopic use is associated with less haemorrhage, no external scars and better magnification of the surgical field to facilitate tumor margin excision. Yet, the major disadvantage of endoscopy is its vulnerability to bleeding and the requirement of bloodless field for clear visualization. Any minor amount of bleeding can preclude the surgical field [17,18].

Different techniques were tried in the past in an attempt to decrease the intraoperative blood loss including preoperative radiation, external carotid ligation, injection of sclerosing agent and systemic administration of female hormones. Yet these methods were proven ineffective [19]. The idea of internal maxillary artery embolization to reduce blood loss was introduced by **Roberson et. al.**[20].

The aim of this study was to evaluate the impact of preoperative embolization of juvenile nasopharyngeal angiofibroma on the surgical outckome.

One of the prominent findings in the current study was the marked reduction of intraoperative blood loss to a mean of 665 ml. The results are close to those reported by **Ballah et. al., Siniluoto et. al., Waldman et al. and Roberson et al.,** [21–24] who reported a mean blood loss of 655 ml, 510 ml, 775 ml and 800 ml respectively in their studies patients.

Authors reported a much higher blood loss in tumor resection without preoperative tumor embolization as **Petruson et. al., Li et. al., Moulin et. al. & Siniluoto et. al.** [19,24–26] reported a mean intraoperative blood loss of2167 ml, 1136 ml, 5380 ml and 1510 ml respectively.

Additionally, in the current study endoscopy had a lower mean blood loss of 388.46 ml compared to 1275 ml of open surgery and 1050 ml of combined procedures. A significant negative correlation was found between endoscopic use and the intraoperative blood loss (r= -.674, p=0.001). These findings are close to those reported by **Lutz et. al.** [27] who reported a mean loss of 548 ml for endoscopy compared to 1088 ml for open surgeries.

In the current study, preoperative embolization reduced the need for blood transfusion (9/30 patients) and was only required in open surgeries and in tumors with intracranial extension with a mean of 928.57 ml. These results are consistent with **Garofalo et. al.** [17] who reported a mean blood transfusion of 750 ml required only in patients operated via open surgery. Similarly, **Oliveria et. al.** [28] reported 9/37 of their patients requiring transfusion, 8 of them underwent open surgery. **Gaillard et. al.** [29] also reported 3/10 of patients required transfusion, 2 of them were not preoperatively embolized.

In the current study, endoscopy could be used in a total of 24/30 patients (63.3%). Of them, 19 patients underwent pure endoscopy with surgeons reporting good visualization of the surgical field and tumor borders and no need for other open procedures. Other authors; **Nicolai et. al**., **Oliveria et. al. & Ferraria et al.** [18,28,30] reported successful use of endoscopy after preoperative embolization in 40/46 patients, 20/37 patients and 9/9 patients respectively.

None of the patients experienced e reported in literature during to embolization in ECA territory as facial paralysis, stroke, necrosis of tip of tongue, or facial atrophy. However, 9/30 patients (30%) experienced minor post procedure complications which were treated medically with no surgical intervention or leaving permanent sequelae. These findings are superior to those reported by **Ogawa et. al. (2012)** [31], who reported an overall complication rate of 46.5%, however most of them were minor and only 2.4% of them were major.

Preoperative angiography allowed the accurate determination of tumor's supplying vessels before and after embolization, the vascular contribution of ICA and the estimated residual tumor blush post embolization. These determinants complement the modern endoscopic surgical approach proposed by **Snyderman et. al.** dividing the tumor into different vascular territories and dealing with them sequentially [11].

Conclusion:

Preoperative transarterial embolization of JNA is an effective method for reducing intraoperative blood loss and transfusion and allowing wider shift towards endoscopic resection. Preoperative angiography is an essential step for preoperative planning and determination of tumor vascular supply. Minor complications may be encountered, but major complications are rare and most can be treated without leaving permanent sequelae.

Abbreviations:

JNA= Juvenile nasopharyngeal angiofibroma, **DSA**=Digital Subtraction Angiography, **ECA**= External carotid artery, **ICA**= Internal carotid artery, **IMax**= Internal maxillary artery, **UPMC**= University of Pittsburgh Medical Center, **MRI** = magnetic resonance imaging, **CT**= Computed tomography, **Fr**=French, **PVA** = polyvinyl alcohol.

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