

BEYOND THE SCALPEL: ELEVATING JUVENILE NASOPHARYNGEAL ANGIOFIBROMA SURGERY WITH PRE-OPERATIVE EMBOLIZATION - QUANTITATIVE ANALYSIS OF OPERATIVE TIME AND BLOOD LOSS

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ABSTRACT

Juvenile nasopharyngeal angiofibroma (JNA), constituting 0.05-0.5% of all head and neck tumours, predominantly affects males aged 10-18, with recent reports highlighting its increased incidence in the Indian subcontinent. (1) This study explores the role of preoperative embolization in mitigating intraoperative blood loss, reducing the risk of massive haemorrhage, shortening operation time, and enhancing intraoperative visibility during surgical excision. The research aims to provide a thorough understanding of the impact of preoperative embolization on surgical outcomes, emphasizing its significance in managing this highly vascular tumor. The investigation involves a detailed analysis of cases using imaging techniques, including nasal endoscopy, contrast-enhanced CT scans, and MRI, coupled with a focus on the clinical presentation and non-surgical management options. The study seeks to contribute valuable insights for optimizing the treatment of JNA, addressing its unique challenges, and improving patient outcomes.

INTRODUCTION

Historical Perspective: Nasopharyngeal angiofibromas have been recognized since ancient times, with Hippocrates performing early surgical interventions.^[1,2] Evolution of surgical techniques, from midline incisions in 470 BC to modern advancements, has shaped the management of JNA.^[3]

Etiological Theories: Tumor origin is crucial for surgical planning. Proposed origins of angiofibromas vary: periosteum of the nasopharyngeal vault (Ringertz, 1938); inequalities in skull base bone growth (Som and Neffson, 1940); embryonic fibrocartilage between basiocciput and basisphenoid (Bensch and Ewing, 1941); or conjoined pharyngobasilar and buccopharyngeal fascia (Brunner, 1942). Osborn (1959) considered hamartomas or residues of fetal erectile tissue. Girgis and Fahmy (1973) noted paraganglioma-like cell nests ('zellballen'). The pituitary androgen-estrogen axis alteration theory is supported by JNA prevalence in adolescent males, though pituitary-gonadal axis studies found no abnormalities.

The widely accepted theory implicates embryologic chondrocartilage.^[4] JNA originates from the sphenopalatine foramen's superior margin, extending

along natural foramina and fissures. It invades the nasopharynx, nasal cavity, sinuses, and pterygopalatine fossa, potentially eroding pterygoid plates to reach the infratemporal fossa or orbit. Superior extension involves foramina rotundum, ovale, and lacerum, reaching the middle cranial fossa and parasellar region, typically extradural and lateral to the sphenoid sinus. Intracranial extension patterns include destruction of the skull base at the pterygoid process attachment.

Clinical Presentation: Recurrent severe epistaxis often leading to life-threatening bleeding. Chronic anemia, hyposmia, anosmia, nasal intonation, and facial deformities characterize advanced cases. The tumor's slow growth results in delayed presentation, with substantial morbidity evident upon examination.^[5]

Gross and Histological Features: Macroscopically, JNA presents as a lobulated, pink to purplish mass with a fibrous appearance and rich vascularity. Histologically, it comprises fibrous stroma and irregularly shaped blood vessels, lacking smooth muscle and elastic fibers.^[6]

Diagnosis:

Imaging Modalities: CT and MRI are crucial for evaluating tumor extent, with CT revealing bone erosion and MRI providing detailed staging.

Angiography aids in preoperative assessment of vascular supply, guiding embolization strategies.^[7]



Figure 1: The compelling visual representation illustrates Juvenile Nasopharyngeal Angiofibroma (JNA) as a lobulated, pink to purplish mass, showcasing a discernible fibrous texture and notable vascularity. The clinical presentation is marked by patient-reported hyposmia, nasal intonation, and facial deformities, providing valuable insights into the intricate clinical characteristics of this condition.



Figure 2: Clinical imaging reveals an axial section contrast-enhanced CT, unveiling the presence of a left-sided Juvenile Nasopharyngeal Angiofibroma (JNA) extending to the contralateral side.

Staging Systems: Tumour size and extra nasopharyngeal extension typically exhibit a linear relationship, but measuring size consistently proves challenging. In the case of malignant neoplasms in the nasopharynx, staging relies on the sites of involvement. This staging is crucial for both individual assessment and comparisons of treatment across institutions. Multiple staging systems have been proposed to address this complexity.

Chandler et al (1984).^[8]

Stage I	Tumour confined to the nasopharyngeal vault
Stage II	Tumour extending into nasal cavity and/or sphenoid sinus
Stage III	Tumour extending into antrum, ethmoid sinus, pterygomaxillary fossa, infratemporal fossa, orbit and or cheek
Stage IV	Intracranial tumour

Sessions et al (1981).^[9]

Stage I A	Tumour limited to posterior nares/or nasopharyngeal vault
Stage I B	Tumour involving posterior nares and/or nasopharyngeal vault with involvement of at least 1 paranasal sinus
Stage II A	Minimal lateral extension into pterygomaxillary fossa
Stage II B	Full occupation of pterygomaxillary fossa with or without superior erosion of orbital bones
Stage II C	Infratemporal fossa with or without cheek
Stage III	Intracranial extension

Chandler's staging system was based on a system proposed for nasopharyngeal cancer. JNA, however, is a benign lesion with a predictable growth pattern that differentiates it from malignant nasopharyngeal tumours. In Chandler's staging system stage III includes all extra nasopharyngeal sites except sphenoid sinus and intracranial extension. The inclusion of multiple sites with variable resectability into a single stage limits the clinical usefulness of the system.

Treatment Approaches:

Surgical Interventions: Various surgical approaches, including endoscopic transnasal, transpalatal, medial maxillectomy, facial translocation, and infratemporal fossa, are employed for Juvenile Nasopharyngeal Angiofibroma (JNA) excision, contingent on surgeon expertise.^[10] Critical considerations involve prioritizing optimal tumor exposure, effective hemostasis, and minimizing postoperative facial sequelae, while ensuring no

interference with facial skeleton growth. This necessitates a balance between aggressive transfacial approaches and the imperative to minimize both functional and cosmetic deficits in alignment with contemporary treatment philosophies. Surgeries are conducted within 24-72 hours post-embolization, utilizing endoscopic approaches for stages I/II and open approaches (Lateral Rhinotomy/Maxillary swing) for stages III/IV. Quantification of blood loss, transfusion requirements, operative time recording, and documentation of tumor resection extent are integral, with routine postoperative CT scans at six months facilitating comprehensive assessment.

Role of radiation in JNA: Although radiation has been effective in treating Juvenile Nasopharyngeal Angiofibroma (JNA), concerns about long-term complications, including secondary malignancies in the head and neck, and deleterious effects on the visual system, have limited its adoption. Recent advancements in radiation therapy offer better

precision, but worries persist about impacts on facial growth centers, particularly in adolescents. Due to these concerns, many institutions still prioritize surgery, even for cases with intracranial extension.^[11] Both extracranial and combined neurosurgical/otolaryngologic techniques have been used, but surgeries come with serious complications such as hemorrhage, optic neurovascular bundle injury, cranial nerve damage, meningitis, and motor nerve deficits.

Preoperative chemotherapy: Preoperative chemotherapy for Juvenile Nasopharyngeal Angiofibroma (JNA) is explored, with estrogen therapy showing variable effects but unwanted feminizing effects in adolescent boys and surgery delays. A study on the nonsteroidal androgen receptor blocker, flutamide, reported tumor shrinkage up to 44%, but temporary side effects like nausea, breast tenderness, and gynaecomastia occurred. However, a pilot study with seven stage IV patients showed only a 7.5% mean shrinkage, deemed insignificant.^[12] Despite potential in preoperative preparation, further research is needed, especially in advanced cases with intracranial extension.

Preoperative embolization: Preoperative embolization is a common strategy to minimize intraoperative bleeding in highly vascular Juvenile Nasopharyngeal Angiofibroma (JNA). Introduced by Robertson in 1972, embolization aims to achieve tumor devascularization while preserving normal vascular supply to surrounding tissues. Advances in techniques and materials, such as small particles and microcatheters, have improved safety. The vascular supply of JNA depends on tumor size and extension, involving both internal and external carotid arteries.^[13] Selective obliteration of intratumoural vascular beds with agents like PVA particles has shown efficacy. Notably, Onyx, a novel embolic material, offers benefits, including preventing migration and reducing intraoperative blood loss. Direct percutaneous embolization with Onyx, while requiring traditional angiography, allows targeted efforts and decreases the time needed for the procedure.^[14] Despite concerns about complications, recent studies highlight its potential advantages, particularly in reducing blood loss and transfusion requirements during surgical resection. Refinements in techniques and the availability of new materials contribute to minimizing residual risks. The transarterial embolization technique involves angiography, selective catheterization, and the use of PVA particles, ensuring maximal vascular tumor embolization with technical success determined by safety and absence of non-target embolization or vascular occlusion.

Technique of Percutaneous embolization: The lesion is usually punctured with a 19-ga 10-cm needle using CT, fluoroscopy, biplane transarterial roadmaps, or bone landmarks. Needles will be placed into the lesion from transzygomatic, transnasal, transoral, and transbuccal approaches. Needle

position within the lesion will be considered correct when blood reflux from the needle was slow but continuous. A small amount of contrast is injected through the needle, and a parenchymogram is obtained to confirm needle position and assess the neovascular compartment of the lesion filled by the needle position.^[15] Biplane angiography is also performed to confirm needle position in the lesion and to make sure the needle did not transgress the ICA or a branch of the ECA. The dead space of the needle is primed with dimethyl-sulfoxide. EVOH is injected through the needle to embolize the tumor.

MATERIALS AND METHODS

Study Design: A retrospective study at Lokmanya Tilak Municipal General Hospital involved 40 patients diagnosed with Juvenile Nasopharyngeal Angiofibroma (JNA). The study aimed to compare outcomes between patients undergoing preoperative embolization and those who did not, specifically focusing on blood loss and surgery duration.

Study Area and Source of Data: The study was conducted at the Lokmanya Tilak Municipal General Hospital, with data sourced from patients referred to the Interventional Radiology (IR) department with a diagnosed JNA from the Department of Ear, Nose, and Throat (ENT) for pre-operative embolization.

Sample Size and Inclusion/Exclusion Criteria

The sample size comprised 40 patients meeting inclusion criteria, including being newly diagnosed with JNA, scheduled for excisional surgery, and willing to undergo pre-operative embolization. Exclusion criteria involved non-consent for pre-operative embolization, being unfit for surgery, and having significant External Carotid Artery (ECA) – Internal Carotid Artery (ICA) collateral.

Statistical Methods: Variables considered for analysis included age, side, JNA stage, main feeding artery, blood loss, and surgery duration. Statistical analysis involved the use of appropriate methods such as the Chi-square test and independent T-test. Mean, standard deviation, and p-values were calculated to assess the significance of the results.

Procedure for Data Collection: Data collection included assessing 40 patients based on intraoperative blood loss, operation time, and intraoperative visibility. Cases were categorized into JNA stages according to FISCH classification. Intraoperative blood loss was calculated using blood-soaked gauze packs and suction machine collection. The chosen surgical approach was endoscopic transnasal surgery, and the duration of surgery was measured from incision to closure. This comprehensive approach aimed to provide valuable insights into the impact of preoperative embolization on surgical outcomes in JNA cases.

RESULTS

Group A: preoperatively embolized patients
Group B: non embolized patients (data before 2002).

Table 1: Age Distribution of study participants

Age (in years)	Group A	Group B
6 to 10	01(03.4)	02(18.2)
11 to 15	14(48.3)	05(45.4)
16 to 20	10(34.5)	02(18.2)
21 to 25	04(13.8)	02(18.2)
Total	29(100.0)	11(100.0)
Mean±SD	16.03±3.39	14.36±5.14

Mean age of patients in group A is 16 and group B is 14. Commonest range of age in group A and group B is 11-15yrs.

Table 2: Side of JNA distribution of study participants

Side (epicenter)	Group A	Group B
Right	11(37.9)	07(63.6)
Left	18(62.1)	04(36.4)
Total	29(100.0)	11(100.0)

Total 29 patients were in group A, out of which 18 patients had the lesion predominantly on the left 11 patients were in group B, out of which 7 patients had the lesion predominantly on the right

Table 3: Stage distribution of study participants

Stage	Group A	Group B
1	06 (20.7%)	02 (18.2%)
2	13 (44.8%)	04 (36.3%)
3	06 (20.7%)	03 (27.3%)
4	04 (13.8%)	02 (18.2%)
Total	29 (100.0%)	11 (100.0%)

In group A, out of 29 patients, 13 were of stage 2 which constituted 45 % of cases. In group B, out of 11 cases, 4 were of stage 2 which constituted 36 % of total non embolized patient.

Table 4: Main feeding artery (MFA) Distribution of study participants

MFA	Number	Percentage
IMA	17	58.6
BL IMA	06	20.7
IMA & APA	06	20.7
Total	29	100.0

The most common main feeding artery (MFA) in group A was found to be Internal Maxillary Artery (IMA). In almost all the cases, IMA had its supply to the lesion.

Table 5: Comparison of blood loss by stage and group

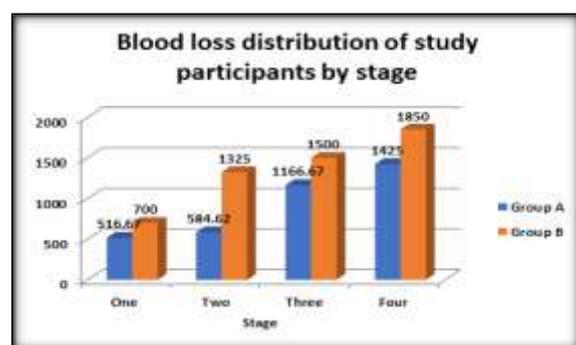
Stage	Group A (in ml)	Group B (in ml)	P Value
One	516.67	700.00	0.046
Two	584.62	1325.00	0.009
Three	1166.67	1500.00	0.147
Four	1425.00	1850.00	0.054

P<0.05 Considered statistically significant

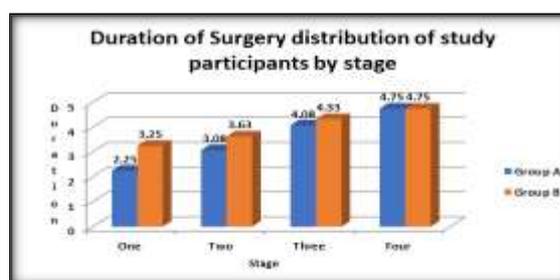
Table 6: Comparison of duration of surgery by stage and group

Stage	Group A (in hr)	Group B (in hr)	P Value
One	2.25	3.25	0.024
Two	3.08	3.63	0.184
Three	4.08	4.33	0.685
Four	4.75	4.75	1.000

P<0.05 Considered statistically significant



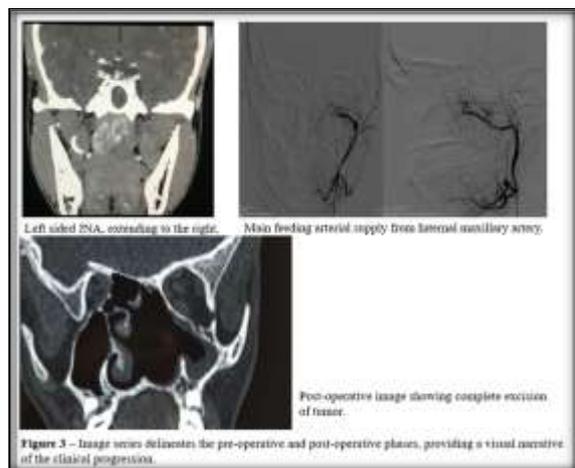
Blood loss was found to be lesser as compared to the same stage in group B, more so for stage 2 patients.



Duration of surgery reduced significantly in group A, compared to group B of same stage. However, in stage 4, no significant change in duration of surgery was seen.

DISCUSSION

Juvenile nasopharyngeal angiofibromas (JNA), an affliction predominantly affecting adolescent males, frequently extend intracranially, relying on the internal maxillary artery for primary vascular supply. The gold standard treatment is surgical resection, yet the formidable challenge lies in mitigating substantial intraoperative hemorrhage. Historically, external carotid artery ligation sought to address this concern. Our study, encompassing 40 patients, underscores the transformative impact of preoperative embolization on surgical outcomes, markedly reducing blood loss during surgery (Group A: 922 ml vs. Group B: 1343 ml). Group A's distinct advantage, fostering meticulous operative field visualization and expediting tumor assessment, led to a noteworthy reduction in overall surgery duration. Despite the documented lower recurrence rates associated with preoperative embolization, cases with intracranial extension in Group A demonstrated a noteworthy paradox, exhibiting significant intraoperative blood loss. Postoperative blood transfusions were judiciously administered based on the extent of blood loss. Importantly, the study illuminated the remarkable safety profile of preoperative embolization, with no discernible long-term complications, reinforcing its pivotal role in advancing the therapeutic landscape of JNA.



CONCLUSION

Juvenile nasopharyngeal angiofibromas (JNAs), benign and highly vascular tumors primarily affecting adolescent males, often exhibit intra-cranial extensions. The internal maxillary artery is the primary feeding vessel, with other arteries contributing as the tumor enlarges. Treatment modalities, including waiting for spontaneous regression, radiation, sclerosing agent injections, and surgical resection, have evolved. Surgical resection, while widely accepted, poses challenges, notably significant intraoperative hemorrhage leading to incomplete removal and higher recurrence rates.

Preoperative embolization has emerged as an effective strategy, reducing blood loss, and improving surgical outcomes. Studies report varying reductions in blood loss, with potential benefits in terms of clear visualization, easier resection, and shorter surgery duration. Preoperative embolization may also correlate with lower recurrence rates. In cases with internal carotid artery supply, there may be increased intraoperative blood loss. While complications like stroke or blindness are rare in embolization, no serious long-term complications were observed in this study. Overall, preoperative embolization proves to be a valuable adjunct to surgical management of JNAs, enhancing safety and efficacy.

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