ORIGINAL RESEARCH PAPER

Gynaecology

Pre-Surgical Embolization of Juvenile Nasal Angiofibroma: Our Experience

KEY WORDS: JNA – Juvenile Nasal Angiofibroma, SD – Standard deviation, INR – Indian Ruppee

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ABSTRACT

- **Context:**JNA is a rare tumour, accounting for up to 0.5% of all head and neck tumours, show features like high vascularity and local aggressiveness.
- **Aims:**To evaluate the utility of pre-surgical embolization of JNA and comparison of mean blood loss, total duration of surgery, and the cost of treatment between embolized and non-embolized groups.
- **Methods and Material:** In this retrospective study, records of patients from 2005 to 2015 were reviewed. A record of 60 patients was reviewed. Patients with Radkowski stage IIB and greater were included in the study. During this period, 20 patients were non-embolized while 15 were embolized. Both the groups were compared and assessed in terms of tumour stage, total duration of surgery, mean blood loss and cost of treatment.
- **Results:**Higher mean blood loss was seen in non-embolized group (1159 ml) in comparison to embolized group (524 ml). Mean duration of surgery was higher in non-embolized group (398.61 ± 99.76 min) than in embolized group (252.10 ± 88.51 min). While higher cost of treatment was seen in embolized group (Rs. 37800 ± 2500) in comparison to non-embolized group (Rs. 29600 ± 1500).
- **Conclusions:**From the present study, we conclude that if cost of the treatment is kept apart, then benefits of performing pre-surgical embolization of JNA outweighs that of non-embolization in terms of lower blood loss and lesser operative time, especially in high grade tumours, which serves as a boon for the operating laparoscopic surgeon and also improves the surgical outcome.

Introduction:

JNA is a rare tumour, accounting for up to 0.5% of all head and neck tumours.[1]Typically presenting in males between age group of 5 to 25 year, with a mean age of 15 years.[1, 2] This tumour being one of the most common benign nasopharyngeal tumours with pathological features like high vascularity and local aggressiveness, with rare malignant transformation.[1]An association of JNA with familial adenomatous polyposis has been established.[3]

Most common clinical symptom includes unilateral or bilateral nasal obstruction and recurrent spontaneous epistaxis however other symptoms includes facial swelling, proptosis, hyponasal speech and diplopia.

The tumour origin is believed from either the lateral wall or the roof of the nasopharynx especially the sphenopalatine foramen, which serves as communication between the pterygopalatine fossa and the nasal cavity.[1, 4,] 5They have tendency to occupy various nasopharyngeal spaces, example maxillary, ethmoid, and sphenoid sinuses. Laterally, the tumour expands into the pterygomaxillary fossa leading to protuberance of the maxillary sinus. The tumour extensions can be seen in various local regions like infratemporal fossa, orbit and base of skull.

However in rare cases the tumour may destroy the posterior sphenoid sinus resulting in invasion to the optic chiasm, pituitary, and/or cavernous sinus. [6,7], 8Hence pathways of tumour extension is key to proper evaluation and staging of these lesions. Newer modalities like CT and MRI provide a better resolution and insight into defining tumour stage and therefore helping in deciding the required method of treatment.

We have conducted this study in order to evaluate the benefits of preoperative embolization in cases of JNA and compare it with a non-embolized group in terms of blood loss, duration of surgery and cost of overall treatment.

Subjects and Methods:

Patient criteria:

Retrospectively records of the patients from 2005 to 2015 were reviewed. During this period, a total of 60 patients were operated for JNA, of which 35 patients were directly operated, while 25 patients were pre-surgically embolized and then operated to remove the tumour mass.

For the present study, Radkowski stage IIB and greater were selected, thus finally 20 non-embolized and 15 embolized patients were included, as 15 from non-embolized and 10 from embolized groups came under exclusion criteria.

Comparison of stage of tumour, total duration of surgery, mean blood loss and overall cost of treatment between the two groups was done.

Staging Considerations:

Many authors like Fisch, Bremer, Antonelli, Andrews, and Radkowski have introduced multiple staging criteria to help determine the surgical resectability of tumour and planning the best possible surgical approach. [9-13]

Staging criteria described by Radkowski et al in 1996 was used for the present study, as it is the most recent adaptation and defines the tendency of JNA to extend posteriorly towards the pterygoid plates and also focuses on the degree of presence of skull base erosion.[9]

No universally acceptable classification system has been developed, and hence, various authors have used different classification systems.

No classification system has been universally accepted, and therefore it is common for various articles to discuss their results using different systems.

Chances for residual and/or recurrence of tumour increase with a higher stage of tumour. However sphenoidal extension leads to an increased risk of residual and/or recurrence as the region is difficult to access surgically.[2,14]

Staging chart of juvenile nasopharyngeal angiofibroma of Radkowski et al,199614 (Table 1).

Patients were further sub-categorized in different stages using the above criteria. Pre- and post-operative CT and/or MRI were done in both the categories.

Based on the above criteria the patients were then sub-categorized into different stages. Both pre and postoperative CT and/or MRI was done in both the category. (Figure – 1, 2)

Evaluation of the Data:
Outcome measures in terms of mean blood loss, duration of surgery, overall cost of treatment, staging of tumour, requirement of blood transfusion, duration of hospital stay were evaluated.

Procedure Done:
A team of interventional radiologist, anesthetist, otolaryngologist and residents doctors had performed the procedure. Patient selection for embolization was based on the combined consensus of the interventional radiologist and otolaryngologist.

Local anesthesia was administered under the supervision of an anesthetist and then puncture in the femoral artery was made by the interventional radiologist to establish femoral access. Clinical and hemodynamic changes were continuously monitored.

Conventional transfemoral arterial access was obtained for all patients and 5-Fr head hunter (H1) catheter from cook was used to take angiograms of both internal and external carotid artery bilaterally in order to identify the feeder vessels of the benign mass. The feeder arteries were identified as blush in the region of tumour.

Thereafter super selective catheterization of the feeding arteries arising from external carotid artery were performed with a 2.7-Fr micro catheter (Progreat micro catheter from Terumo) and were embolized using gelform (Figure - 3). Only gelform was used in all the patients in order to reduce the procedure cost. We never embolized the branches of internal carotid artery even if they were feeders for the tumour mass like in stage IIIA and IIIB.

Tumour excision surgery was performed 24 - 48 hours after embolization.

All patients in category IIB, IIC, IIIA underwent endoscopic removal of the tumour mass in both the groups however the patients with stage IIIB due to the large tumour mass and extension underwent open surgery by lateral rhinotomy via midfacial degloving.

Blood loss during the surgical resection was estimated during the procedure by measuring the amount of fluid collected from the operative field and subtracting the amount of saline applied during surgery.

Ethical approval:
Consent was taken from the medical record department to use the data of the patients from 2005 to 2015. Care was taken to conceal all the personal information of the patients. The approval to conduct the study was also obtained from the institutional ethical committee.

Results:
All the 35 patients included in the study were male. In all the 15 patients who had undergone embolization of JNA before surgery, the embolization procedure went uneventful without any local or general neurological complications.

8 patients in embolized group and 10 patients in non-embolized group were stage IIB, 4 patients in embolized and 5 patients in non-embolized group were stage IIC, 2 patients in each group were stage IIIA, 1 patient in embolized group and 3 patients in non-embolized group were stage IIIB. (Figure 4, Chart 1)

The mean of blood loss was 524 ml in embolized group as compared to 1159 ml in non-embolized group whereas subgroup analysis revealed mean blood loss in embolized group and non-embolized group was 350ml (SD 100ml) and 790ml (SD 80ml) in stage IIB, 470ml (SD 80ml) and 1160ml (SD 70ml) in stage IIC, 880ml (SD 70ml) and 1590ml (SD 60ml) stage IIIA respectively. Stage IIIB had only a single patient in embolized group with a blood loss of 950ml and mean blood loss was 2100ml (SD 180ml) in non embolized group. (Figure 4 – Chart 2)

The mean duration of surgery was 252.10 minutes (SD 88.51 minutes) in embolized group and 398.61 minutes (SD 99.76 minutes) in non-embolized group. (Figure 4- Chart – 3)

The mean cost of treatment was INR 37800 (SD 2500) in embolized group and INR 29600 (SD 1500) in non-embolized group. (Figure 4 – Chart 4)

Discussion:

As JNA is a highly vascular mass, therefore blood loss during surgical procedure is inevitable. What add-on to this trouble is that the feeder vessels consist of a single endothelial lining and lack the capacity of vasoconstriction as there is lack of smooth muscle layer.[15,16]

This in turn leads to extensive blood loss encountered during surgical resection and as a consequence obscures the surgical field leading to complications like incomplete resection and/or injury to normal surrounding structure. [17,18]

In all the patients of JNA with mass supplied with feeders only from external carotid artery achieved a near total devascularisation and sub-total grades were achieved in whom feeder vessels were also from internal carotid artery.

Presence or absence of tributaries from the internal carotid artery becomes a matter of concern as it increases the risk of performing the embolization. The embolizing material may flow into brain through the internal carotid artery and can cause stroke and encephalic damage.[19]

Significant reduction in blood loss in embolized group was seen in comparison to non-embolized group.

We have observed that there was a significant reduction in the blood loss in embolized group compared from that of non-embolized group.

Douglas and Wormald reported that preoperative embolization can reduce the intraoperative blood loss to less than 1000ml.20 It is well known that there is a significant reduction of blood loss in patients with preoperative embolization of JNA, which is a major cause of morbidity.[21] Other research also reported nearly 60% reduction in mean blood loss during operative procedure in pre-embolized patients.[22]

There was also a reduction in the transfusion requirements. Moulin and Chagnaud have also reported a similar finding.[23]

In our study there was a significant reduction in the total duration of surgery. Other studies have also shown that occlusion of surgically inaccessible feeders helped cut short the blood loss and thereby increasing the visualization, allowing much easier identification and differentiation of normal anatomical structures from pathological structures which in turn helped in reducing the total duration of surgery.[24]

Surgeon's confidence level was assessed when operating an

embolized patient in comparison to operating a non-embolized patient. Though no scale was used for assessment, However, surgical team had reported higher confidence level when operating an embolized patient in comparison to non-embolized patient.

The best treatment to date remains surgical removal of the tumour.[24]

Multiple embolic agents are available to effect vessel occlusion, including gelform, polyvinyl alcohol particles, trysacryl microspheres (Embospheres; Bio- Sphere Medical, Rockland, Maine), microcoils, N-butyl 2-cyanoacrylate (Trufill; Cordis, Miami Lakes, Florida), and Onyx (ethylene vinyl alcohol copolymer; ev3). In the present study, gelfoam alone was used due to financial constraints.

Finally we also looked for the total cost borne by the patient throughout treatment including medication charges, hospital stay, transfusion charges and procedure cost in both the groups and there was significant difference in both the groups, with a higher cost in embolized group.

Other benefits like lesser blood loss, lesser operating time, requirement of lesser medications, better surgical viewing and lesser surgical complications because of pre-surgical embolization, cost of the treatment can be kept apart, as other benefits outweigh the costs of the treatment.

Limitations of the study was that follow-up regarding the tumour recurrence was not available for all the included patients, secondly, we had used only gelfoam, while other centers use newer methods like PVA, microcoils, onyx, etc. because of this effectiveness of other embolic agents could not be done.

Conclusion:

From the present study, we conclude that embolization of JNA has many benefits which outweigh the higher cost of the procedure, which improves the surgeon's confidence level and also the surgical outcome.

TABLE 1 - Juvenile Nasopharyngeal Angiofibroma Staging by Radkowski

Stage	Description
IA	Limited to nose or nasopharynx
IB	Same as IA with extension into 1 or more paranasal sinus
IIA	Minimal extension through the sphenopalatine foramen into and including a minimal part of the medial part of the pterygopalatine fossa.
IIB	Full occupation of the pterygopalatine fossa, displacing the posterior wall of the maxilla forward; lateral or anterior displacement of the branches of the maxillary artery; superior extension may occur, eroding orbital bones.
IIC	Extension through the pterygomaxillary fissure into the cheek and infratemporal fossa or posterior to the pterygoid plates.
IIIA	Erosion of the skull base with minimal intracranial extension.
IIIB	Erosion of the skull base with extensive intracranial extension with or without cavernous sinus involvement.

Source: Radkowski D, McGill T, Healy GB, et al. Angiofibroma changes in staging and treatment. Arch Otolaryngology Head Neck Surg 1996

Figures and Charts:-

Figure 1

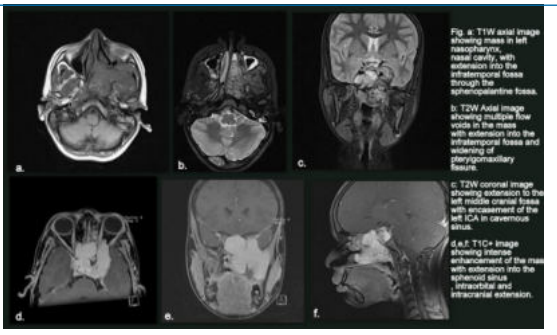


Figure 2

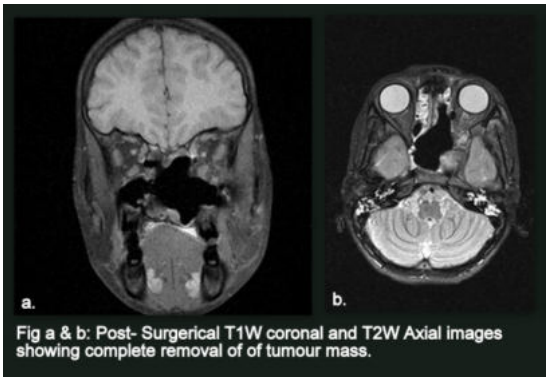


Figure 3

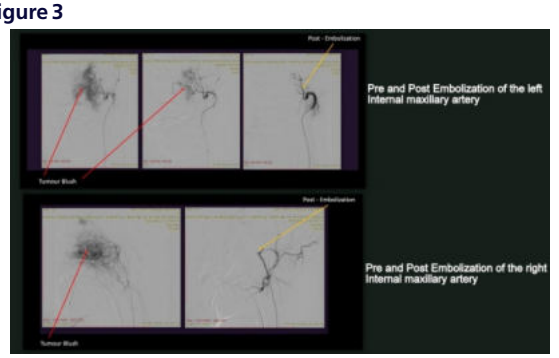
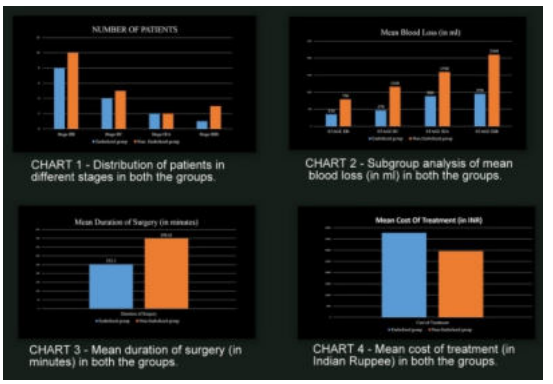


Figure 4



References:

1. Gullane PJ, Davidson J, O'Dwyer T, et al. Juvenile angiofibroma: a review of the literature and a case series report. Laryngoscope 1992;102:928–33
2. Riascos R, Lazor J, Squires JH, et al. Imaging and anatomic features of juvenile angiofibroma. Neurographics 2011;1: 84–89
3. Ponti G, Losi L, Pellacani G, et al. Wnt pathway, angiogenetic and hormonal markers in sporadic and familial adenomatous polyposis-associated juvenile nasopharyngeal angiofibromas (JNA). AppIImmunohistochemMolMorphol 2008;16:173–78
4. Bremer JW, Neel HB 3rd, DeSanto LW, et al. Angiofibroma: treatment trends in 150 patients during 40 years. Laryngoscope 1986;96:1321–29
5. Scholtz AW, Appenroth E, Kammen-Jolly K, et al. Juvenile nasopharyngeal

angiofibroma: management and therapy. Laryngoscope 2001;111(4 pt 1):681–87
6. Pryor SG, Moore EJ, Kasperbauer JL. Endoscopic versus Traditional Approaches for Excision of Juvenile Nasopharyngeal Angiofibroma. Laryngoscope. 2005;115:1201-7.
7. CarvalhoJúnior RS, Mendonça CA, D'Ávila JS, Santos ACG, Jesus EPF, Sobral PM. Abordagemendoscópica no tratamento do nasoangiofibroma: relato de caso. Braz J Otorhinolaryngol. 2005;71(3):4-19.
8. Bernal-Sprekelsen M, Alobid I, Guilemany JM. Angiofibroma Juvenil. ActaOtorrinolaryngol. 2007;58(1):84-95.
9. Radkowski D, McGill T, Healy GB, et al. Angiofibroma changes in staging and treatment. Arch Otolaryngology Head Neck Surg 1996;122:122–29
10. Onerci M, Og□retmenog□lu O, Yu¨cel T. Juvenile nasopharyngeal angiofibroma: a revised staging system. Rhinology 2006;44:39–45
11. Chandler JR, Goulding R, Moskowitz L, et al. Nasopharyngeal angiofibromas: staging and management. Ann OtolRhinolLaryngol 1984;93(4 pt 1):322–29
12. Sessions RB, Bryan RN, Naclerio RM, et al. Radiographic staging of juvenile angiofibroma. Head Neck Surg 1981;3:279–83
13. Andrews JC, Fisch U, Valavanis A, et al. The surgical management of extensive nasopharyngeal angiofibromas with theinfratemporal fossa approach. Laryngoscope 1989;99:429–37
14. Howard DJ, Lloyd G, Lund V. Recurrence and its avoidance in juvenile angiofibroma. Laryngoscope 2001;111:1509–11
15. Khalifa MA, Ragab SM (2008) Endoscopic assisted antral window approach for type III nasopharyngeal angiofibroma with infratemporal fossa extension. Int J PediatrOtorhinolaryngol 72(12):1855–1860
16. Midilli R, Karci B, Akyildiz S (2009) Juvenile nasopharyngeal angiofibroma: analysis of 42 cases and important aspects of endoscopic approach. Int J PediatrOtorhinolaryngol 73:401–408
17. Pryor SG, Moore EJ, Kasperbauer JL (2005) Endoscopic versus traditional approaches for excision of juvenile nasopharyngeal angiofibroma. Laryngoscope 115:1201–1207
18. Roberson GH, Biller H, Sessions DG et al (1972) Presurgical internal maxillary artery embolization in juvenile angiofibroma. Laryngoscope 82(8):1524–1532
19. Petruson K, Rodriguez-Catarino M, Petruson B et al (2002) JNA: long-term results in preoperative embolized and non-embolized patients. ActaOtolaryngol 122:96–100
20. Douglas R, Wormald PJ. Endoscopic surgery for juvenile nasopharyngeal angiofibroma: Where are the limits? CurrOpinOtolaryngol Head Neck Surg 2006; 14: 1-5.
21. Santaolalla F, Araluce I, Zabala A, López A, Garay M, Sanchez JM. Efficacy of selective percutaneous embolization for the treatment of intractable posterior epistaxis and juvenile nasopharyngeal angiofibroma (JNA).ActaOtolaryngol. 2009;129:1456–62.
22. Antonelli AR, Cappiello J, Di Lorenzo D, Donajo CA, Nicolai P, Orlandini A. Diagnosis, staging, and treatment of juvenile nasopharyngeal angiofibroma (JNA). Laryngoscope. 1987;97:1319–25.
23. Guy Moulin, Christophe Chagnaud, Regis Gras, ErwannGueguen, Patrick Dessi, Jean-Yves Gaubert, and et. al. Juvenile Nasopharyngeal Angiofibroma: Comparison of Blood Loss During Removal in Embolized Group Versus Nonembolized Group. CardiovascInterventRadiol. 1995 18: 158 – 161.
24. J. Lutz, M. Holtmannspötter, W. Flatz, A. Meier-Bender, A. Berghaus, H. Brückmann, P. Zengel Preoperative Embolization to Improve the Surgical Management and Outcome of Juvenile Nasopharyngeal Angiofibroma (JNA) in a Single Center: 10-Year Experience. ClinNeuroradiol. 2015