

Clinicopathological Study of Juvenile Nasopharyngeal Angiofibroma; a Single Institution Study in a Tertiary Care Hospital in Odisha.

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ABSTRACT

Background: Aim- To study the presenting features correlates them with radiological finding & the management of JNA. **Methods:** A prospective study was done on 30 patients who were clinically & radiologically diagnosed as Juvenile nasopharyngeal angiofibroma & treated in department of ENT & HNS SCB Medical College Cuttack during the period of 1.8.2015 to 30.10.2017. All patients are examined & after stabilisation, CECT of nose and PNS done. Staging was done according to clinical & radiological finding by FISCH staging system. Patients were treated by surgical excision by various approaches. Most common procedure done is endoscopic guided excision by medial maxillectomy with lateral rhinotomy approach. Excised tissues send for HP study & patients are followed up for a period of one year. **Results:** All the patients in our study were males. Majority (70%) are in 11-18 yr age group with mean age of presentation 17.13yr. Most common presenting feature is epistaxis with progressive nasal obstruction. Palatal bulging & hearing loss is the commonest extra sinonasal manifestation. Hollman miller sign found in 50% of cases. Maximum patients presented in stage-2 disease. Post operatively 1 death occurred due to blood transfusion reaction. All patients HP study report is found to be nasopharyngeal angiofibroma. **Conclusion:** JNA is a benign tumour & should be suspected in all cases of adolescent male presented with epistaxis & progressive nasal obstruction. CECT, has a definite role in diagnosis, staging & plan of management of JNA & should be done within 1 wk prior of surgical management. Ram-Haran sign which is found most of cases of early stage, help in early diagnosis of JNA & can be a diagnostic criteria for JNA. Endoscope assisted external approach, is definitely beneficial in total disease clearance.

Keywords: angiofibroma, embolization, epistaxis.

Abbreviation used: JNA- juvenile nasopharyngeal angiofibroma; CECT-contrast enhanced computed tomography, PNS-paranasal sinus.

INTRODUCTION

Juvenile nasopharyngeal angiofibroma, recognized since ancient times by Hippocrates, is an uncommon, benign & extremely vascular tumour which arises in the tissue within the sphenopalatine foramen.^[1,2] It accounts for less than 0.5% of all head & head tumour.^[3] It occurs almost exclusively in adolescent male, though rarely found in children, elderly, young & may even pregnant women.^[4-6] It is locally invasive though few have been reported to behave in a more malignant fashion. As it grows, tumour extends to nasopharynx, PNS, pterygopalatine & infratemporal fossa. Larger tumour can involve the orbit & cavernous sinus.

JNA presents with recurrent severe epistaxis accompanied by progressive nasal obstruction, which is the classical symptoms. Other features includes, swelling in cheek, trismus, hearing loss secondary to ET obstruction, anosmia, nasal intonation of voice; proptosis, diplopia, visual loss, facial pain & headache in case of extensive tumor growth with orbit & cavernous sinus invasion.^[7]

The diagnosis of JNA is essentially based on a careful clinical history, detailed physical examination & radiological findings. Imaging techniques have led to major advances in the diagnosis and treatment of JNA. Nasal endoscopy, CECT and MRI help establish the site and extension of this tumor and the relations with blood vessels and nerves, all of which make it possible to precisely stage the tumor and help during surgery. Biopsies are not recommended because of highly vascular nature of this tumor. Arteriography is done to assess the vascular supply in larger tumors and to make it possible to embolize these vascular lesions to reduce intraoperative bleeding.^[8-10]

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Treatment modality depends mainly on the extent of the lesion. Surgery is the preferred modality of treatment for all stages of the mass up to stage Iva while radiotherapy is used for stage IVb. Several surgical approaches have been described for the treatment of JNA; these include the transpalatine approach, lateral rhinotomy approach, mid-facial degloving approach & endoscopic endonasal approach. In any of these techniques, the recommended treatment of choice is preoperative tumor embolization to decrease intraoperative bleeding when removing the angiofibroma.^[11,12] Furthermore, postoperative complications including cosmetic deformities, facial scars, facial asymmetry, and psychological trauma should also be taken into consideration. Considering that most JNA patients are adolescents and are still growing, these complications could be critical not only physically, but also emotionally. In recent years, endoscopic surgery has been used for resection of JNA.^[13]

Aims & objectives

1. To assess all cases of bleeding nasal mass; clinically & radiologically suspected to be JNA.
2. To study the incidence of these tumours in relation to age, sex, among overall Head & Neck tumours.
3. To study various Presentation of JNA.
4. To study the role of CT scan in diagnosis and management of JNA.
5. To study the Outcome of various technique & approaches for surgical resection of JNA.

MATERIALS AND METHODS

Source of Data

This is a prospective study of clinicopathological study of JNA cases, treated in the ENT Department of SCB Medical College & Hospital, Cuttack, during the period of 1.8.2015 to 30.10.2017 with proper consent. Total 30 patients included in this study.

Inclusion Criteria

Cases of JNA admitted to this institute from during the period of 1.8.2015 to 30.10.2017 with consent. Exclusion Criteria: Pt with some severe systemic disease or with any bleeding disorders.

Patients who meet the criteria are selected for the study. A detailed history taken for each patient including their symptoms, history of allergy, hypersensitivity, mucocilliary disorders, associated systemic disorders, and previous medical and surgical management.

Patients usually come with epistaxis, stabilisation of those patients were done with anterior nasal packing & blood transfusion given, if Haemoglobin level < 9 gm%. After stabilisation of patients, A complete otorhinolaryngological examination of the patients undertaken including external examination of nose, anterior rhinoscopy, posterior rhinoscopy, paranasal sinuses, test for nasal patency, and nasal endoscopic examination. A general and systemic examination was also performed.

Investigations include routine haematological examination, and CECT of nose and paranasal sinuses & pure tone audiometry. Staging of patient done according to clinical & radiological finding by FISCH staging system. After all required investigation & pre anaesthetic check-up, surgical excision done under general anaesthesia in all patients. Pre operative embolisation not given to any patient due to non availability of the facility in this institute. Two patients of stage-I, done by endoscopic transnasal method & rest 28 cases done by open approaches, out of which 2 patients of stage-I, done by transpalatal approach & 2 patients done by mid facial degloving approach & 24 cases done by lateral rhinotomy approach. In all cases endoscope used intra operatively to see any residual tumor mass left behind which is than cleared. The pterygoid wedge is drilled in all cases which play a vital role in reducing the recurrence of disease. In all cases intraoperative monitoring done, systolic BP kept between 90-100 mm of Hg & diastolic BP kept between 60-70 mm of Hg. Intraoperative blood loss measured & blood transfusion given if blood loss is more than 500ml. Anterior & posterior gauze nasal packing given. Post operatively IV fluid given for 3day in maintenance dose for fluid electrolyte balance, I/O chart, temperature chart maintain. Injectable antibiotics given for 5 days. Posterior & anterior nasal pack removed after 48 hour in stage- I, cases & after 72 hour in stage –II,III cases. Regular dressing done & stitches removed in case of lateral rhinotomy approach cases on 7th post-operative day. Patients discharge with proper advice (nasal douching) & asked to come for review after 1 month & then 6 monthly. Patients are then on regular follow up & see for any complication or recurrence.

RESULTS

Total 30 cases of JNA are admitted to the Department of ENT & HNS SCB Medical College, Cuttack, from 01.08. 2015 to 30.10.2017; which is 0.0137% of total patient attending ENT outdoor, 0.70 % of total patient admitted to our Department & 0.89% of head & neck tumour cases admitted. Incidence per OPD case is 1:7262 with annual incidence of 13.84.

In our study all patients are adolescent males belongs to age of 8-36 years with mean age of presentation is 17.13 years. Majority (70%) of patients fell in the age group of 11–18 years and young adult age group (23.3%) [Table 1]. Majority of patient belongs to low socioeconomic status, from rural area & student by occupation. Out of 30 cases, two recurrent cases are included in the study ; one patient of 23 yr had surgery 2 times, 2yr & 5 yr back respectively and one patient of 22yr has surgery 3yr back, presented as recurrent case .Both the case are at stage –II at the time of presentation. Surgical Excision of mass done by endoscope assisted open approach i.e. medial

maxillectomy with lateral rhinotomy approach, Endoscope used intraoperatively to see about disease clearance. Basisphenoid area drilled out to clear pterygoid wedge area, Both these cases are disease free since then and are under observation.

Table 1: Age distribution of JNA cases.

Age group(yrs)	Number	Percentage
Up to 10	1	3.33%
11-18	21	70%
19-25	7	23.3%
>25	1	3.33%



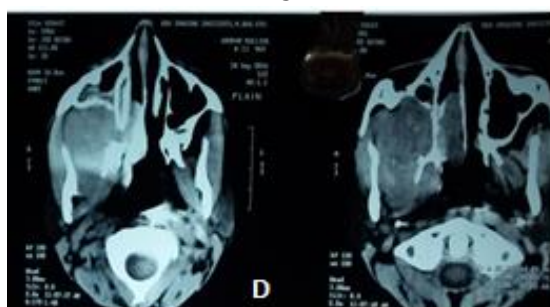
A



B



C



D



E

F



Figure 1: A. Mass in Nasal cavity with palatal bulging, B. Facial deformity by Broadening of nose with cheek swelling C. Palatal bulging with mass in oropharynx. D: Axial view of CT, showing “Holman-miller” sign B. “Ram-haran sign” E,F : Post-Operative picture of excised JNA Mass of stages I,II G-picture showing, Post-operative scar and Stenosis of nares.

Table 2: Presenting symptoms.

Symptom	Number	Percentage
Epistaxis without Nasal obstruction	6	20%
Nasal obstruction without epistaxis	3	10%
Both epistaxis & nasal obstruction	21	70%

Table 3: Duration of illness.

Duration	No. Of patients	Percentage
<6 month	5	16.6%
6 month-1yr	12	40%
1- 2 yr	6	20%
2-3yr	4	13.3%
>3yr	3	10%

In our study, most of patients (70%) presented with both epistaxis and nasal obstruction, than only epistaxis (20%) and nasal obstruction without epistaxis (10%) [Table 2].

In our study, most of patients present to the hospital within 1 yr of onset of symptoms out of which 12(40%) presented within 6 month-1 yr & 5 (16.6%) within 6 month .10% patient presented with more than 3yr of duration of symptoms [Table 3].

Table 4: Symptoms of JNA

Symptoms	No. of patients	percentage
Recurrent epistaxis	27	90%
Nasal obstruction	24	80%
Change in voice	18	60%
Nasal discharge	15	50%
Facial deformity	12	40%
↓hearing, pain in ear	15	50%
↓smell	12	40%
Orbital symptoms (proptosis/diplopia)	1	3.3%
Headache	12	40%

Of all the symptoms [Table 4], epistaxis and nasal obstruction are common, which were present in most of patients. Nasal obstruction without epistaxis seen in 10% cases. Change in voice (60%), Nasal discharge (50%), ↓hearing (50%), Facial deformity, smell & Headache(40%) are common associated symptoms. Most common sign is mass in nasopharynx (100%) followed by mass in both nasopharynx& nasal cavity (80%), pallor (70%), Retracted TM (50%), Palatalbulging (40%) Facial deformity (40%), Cheek swelling (33.3%), Mass in oropharynx (20%).

Table 5: CECT scan finding in JNA

Features	Number	Percentage
CE mass in nasopharynx	30	100%
CE mass in nasal cavity & nasopharynx	27	90%
CE mass invading PPF	21	70%
CE mass invading sphenoid sinus	21	70%
CE mass invading ethmoid sinus	21	70%
CE mass invading maxillary sinus	12	40%
CE mass invading ITF	12	40%
CE mass invading cranium	0	0%
Holman-miller sign	15	50%
Houndousa sign	8	26.6%
Ram-Haran sign	10	33.3%

In our study; most common CT scan finding is contrast enhanced mass in nasopharynx in all cases, followed by contrast enhanced mass in both nasopharynx& nasal cavity (90%), invading sphenoid sinus, invading PPF, ethmoid sinus (70% each), Holman-miller sign(50%), contrast enhanced mass invading maxillary sinus & ITF (40%), Ram-Haran sign (33.3%), Houndousa sign (26.6%) [Table 5]

Table 6: Stage of disease at time of presentation (according to FISCH staging)

Stage of disease	Number	Percentage
Stage -I	6	20%
Stage-II	12	40%
Stage-III	12	40%
Stage-IV	0	0%

Table 7: Surgical resection of JNA done through various approaches & Techniques

Approach	Stage of disease	No. Of pt	Percentage
Endoscopic transnasal open	I	2	6.6%
-Trans palatal	I	4	13.3%
-midfacialdegloving (sublabial)	II	2	6.6%
-lateral rhinotomy	II & III	18	60%
-Transmaxillary approach using Weber-Fergusson's incision	III	4	13.3%

In our study; Stage of disease at time of presentation is mostly stage-II (40%) & stage-III (40%) then stage-I(20%),no patient was found at stage -IV [Table 6].

In our study most of patient (93.4%) underwent external approach; most common procedure being medial maxillectomy by lateral rhinotomy approach, followed by Transmaxillary approach using Weber-Fergusson's incision & Trans palatal approach. midfacial degloving (sublabial) approach in 2 cases. Endoscopic transnasal approach in 2 cases [Table 7]. In our study endoscopic assisted external approach for stage -I disease take approximately 2.5 hr & average blood loss is 500 ml due to limited extension of disease. In stage-II time taken is 3hr with 750ml average blood loss. In stage-III time taken is 3.5 hr with 800ml average blood loss. [Table 10]. In case of blood loss of more than 500ml we give 2 unit of blood transfusion one unit intra operatively& 1 unit post operatively.

Table 8: Post-operative complication

Features	No of patients	Percentage
Haemorrhage	2	6.6%
Septicaemia	1	3.3%
CSF leak	0	0%
Synechia formation	6	20%
Wound dehiscence	2	6.6%
Stenosis of nares	3	10%
BT reaction	1	3.3%
Death	1	3.3%
Recurrence	Till date nil	

In our study synechia formation (20%) is the most common complication followed by stenosis of nares (10%), haemorrhage (6.6%) & wound dehiscence (6.6%). Deaths occurred due to blood transfusion reaction in one patient. Other complication like CSF leak was not found in our study. No recurrence seen till date with cure rate of 96.6 % [Table 8].

Postoperatively, all patients were called for regular follow up for nasal endoscopic examination at monthly interval, and a repeat scan was performed at six monthly intervals in whom it was felt necessary. In our study we have not found patients with recurrence, although minimum 5yr follow up require to declare disease free. All patients are on regular follow up.

DISCUSSION

In our study, incidence of JNA in relation to head & neck tumour cases is relatively high(0.89%), Incidence per OPD case is 1:7262 with annual incidence of 13.84,as it is a tertiary care referral hospital in Odisha; compaired to study of A E Terzian et al 1999 & Prusty NM et al.^[14,15]

In our study all the patients were males&most of the patients are belongs to juvenile age group (i.e.11-18yr) (70%) which happens to be age of most rapid growth of the patient as well as the JNAand young adult age group (23.3%), youngest patient is of 8yr

age & oldest is of 36 yr. Mean age of presentation is 17.13 yr. Finding in our study is similar to study by Gaillard A L et al, Prusty NM et al,^[15,16] Tang IP et al;^[22] corroborative to study of Mistry RC et al,^[9] Terzian et al,^[14] Alexandar Valkov et al & contrast to study of Dr RK pandey et al,^[18,19] as we found 4 cases JNA of age more than 20 yr age.

In our study majority(70%) of patients present with both epistaxis & nasal obstruction along with other symptoms corroborative to study of dr RK pandey et all & A Eterzian;^[14,18] contrast to study by Prusty NM et al & Paris J et al,^[15,17] where nasal obstruction is the most common presentation. 20% of our case present with epistaxis without nasal obstruction, which is due to initial stage of disease, mass not occluding choana or nasal cavity & more vascular component of tumor. 10% cases present with nasal obstruction without epistaxis which may be due to more fibrous component of the tumor, like study of A E. TERZIAN,^[14] where nasal obstruction is common symptom.

In our study Recurrent epistaxis & Nasal obstruction is the most common presentation similar to study of Gaillard A L et al,^[15] Tang IP et al,^[22] prusty NM et al & Tiwari P K et al.^[15,20] Epistaxis was the most distressing symptom of all which made the patients to seek medical advice early. 12 patients came for ENT consultation on first episode of epistaxis though other symptoms were already present for a long duration. The amount of bleeding varied from 10 to 100 ml in each episode and was spontaneous, self-limiting. Patients tolerated nasal obstruction better than epistaxis. Nasal discharge was seen in 15 out of 30 cases but only 7 patients have actually complained about it. It was mucoid to mucopurulent. A history of head ache was given by 10(33.3%) patients, but it was secondary to sinus infection of either maxillary or sphenoid sinus. Nasal intonation of voice, rhinolalia clausa, was observed in 18(60%) cases. It was due to mass in nasopharynx obstructing choana. History of hyposmia was evident in 12 patients, though it was not a presenting complaint. 15 patients had conductive hearing loss because of serous otitis media caused by eustachian tube block. In these patients mass was encroaching on to tubal openings causing physical obstruction. True facial deformity was not a complaint in any of the patients but 10 cases had cheek swelling due to infratemporal spread of JNA & 8 cases have broadening of nose. One Case had orbital proptosis due to intra orbital extension of JNA. He complained of diplopia on lateral gaze. His visual acuity was normal.

In our study tumour was extending into nasopharynx in all the patients, thus it is the most common site of extension. Nasal Cavity extension of JNA was seen in 27(90%) patients with attachments to posterior end of turbinates in 18 cases and posterior end of septum in 6 patients. After occupation of nasopharynx the tumour invaded sphenoid sinus in 70% patients. Pterygopalatine fossa was invaded in

70% patients. 12(40%) Cases also had Infratemporal fossa extension out of which 8 had cheek extension. True maxillary sinus involvement was seen in 12 patients & soft tissue density seen on CT scan in 6 cases is because of infection caused by osteomeatal block due to mass effect. Orbital involvement through infraorbital fissure is seen in one patients. 15(50%) cases found to have "Holman-miller sign" i.e. anterior bowing of posterior wall of maxillary sinus due to mass effect & 8(26.6%) cases found to have "Hondousa sign" i.e. widening the gap between ramus of mandible & maxillary body & 10(33.3%) cases shows "Ram-Haran" sign i.e. triangle shaped area of pterygoid wedge appears quadrilateral due to presence of tumor in early cases. No cases found to have cavernous sinus or intracranial extension.

In my study most of the patient presented at stage II & III; similar to study by Gaillard A L et al & corroborative to study by Paris J et al,^[16,17] Piero Nicolai et al,^[21] Prusty NM et al,^[15] Tang IP et al.^[22] patients presents at later stage may be due to ignorance & low socioeconomic status. Staging was also done again based on extension of JNA found intraoperatively and had to be upstaged in 3 of the 30 patients. During surgery, it was found that one case had JNA extending into sphenoid sinus, two cases were having JNA extension into pterygomaxillary fossa hence they were upstaged accordingly. However, this upstaging did not prevent total excision surgically with the approach that was planned prior to surgery. Incidentally these three patients were having a CT scan which was taken more than a month prior to surgery and JNA should have expanded by the time they were taken up for surgery. They could not obtain a repeat scan within 1 week prior to surgery because of financial constraints that they had.

In our study 2 patients were treated with endoscopic transnasal approach. These two patients were in stage I disease. Preoperative embolization could not be done in these cases (due to non-availability of facility) which seem to be helpful in reducing intraoperative hemorrhage. Everything was kept ready preoperatively to convert endoscopic approach to transpalatal approach as and when it is felt necessary. The advantages felt with endoscopic approach were better and magnified view of various extents and attachments of JNA and the surgical dissection can well be limited to subperiosteal plane. This approach obviates the need for any skin incision and hence no cosmetic defect is expected. The amount of bleeding was considerably less compared to other approaches. Post operatively the follow up was easier with endoscopic approach. The duration of surgery reduced considerably and there was no need for a prolonged post nasal pack. Hence the patient can take oral feeds early and with less morbidity. The disadvantages are it is technically difficult in untrained hands and visibility drastically reduces if endoscope comes in contact with blood.

Four patients having Fisch stage I disease, were treated by Wilson's transpalatal approach and intraoperatively one case found to have extension to sphenoid sinus. This approach gave excellent visualization of entire nasopharynx, and gave good exposure of sphenoid sinus. Thus this approach is best utilized for JNA confined to nasopharynx and sphenoid sinus.

Eighteen patients with stage II&III disease was taken up for lateral rhinotomy as the mass was extending into pterygopalatine fossa, infratemporal fossa and sphenoid sinus. This approach with medial maxillectomy can give good exposure of pterygopalatine fossa and easy manipulation of maxillary artery.

Transmaxillary approach using Weber-Fergusson's incision was used in 4 cases with stage III disease. One case had orbital extension. A modified Weber-Fergusson's incision was used to approach the JNA in pterygopalatine fossa, orbit, and sphenoid. Partial maxillectomy was done in one case. Orbital decompression was done to resolve proptosis.

In the present series 2 patients were treated with Midfacial degloving approach which provides good exposure to the maxillary antrum, nose, pterygopalatine fossa and infratemporal fossa. There will be no deforming scar on face because of the use of a sub labial incision, but needs extensive removal of bones from the anterior, posterior, medial and lateral walls of maxillary antrum.

In our study there is death of one patient on 3rd post-operative day due to blood transfusion reaction. Hemorrhage occurred in two patients immediate post-operative period which was controlled by ligation of bleeding vessel & nasal packing. Synechia formation occur in 6 cases which is released and wax plate given & kept for 21 days. Wound dehiscence seen in 2 case, which improved with secondary stitches & regular dressing. Other complication like CSF leak not found in our study. No recurrence of disease seen till date with cure rate of 96.6%.

CONCLUSION

Juvenile nasopharyngeal angiofibroma or nasopharyngeal angiofibroma is an uncommon benign tumour of adolescents male should be suspected in all cases with recurrent epistaxis and progressive nasal obstruction. It has great potential for growth in all directions through natural foramina & also eroding bony confines. The planning of surgical approach for excision of JNA is based on extent of the lesion or stage of disease. Radiological investigations, like CECT, have definite role in diagnosis, staging & plan of management of JNA & should be done within 1 wk prior of surgical management. Ram-Haran sign which is found most of cases of early stage, help in early diagnosis of JNA & can be a diagnostic criteria for JNA.

Endoscope assisted external approach, is definitely beneficial in total disease clearance & drilling of pterygoid wedge area help in reducing recurrence of disease.

Declaration

- Ethical committee approval and consent of the patients has been taken.
- No Conflict of interests regarding publication of this article.
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