

Original Article

Surgical management of Tympanomastoid Paraganglioma: Experience in BSMMU, Dhaka

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Abstract :

Objective: To characterize the clinical presentation, surgical management, and outcomes of a consecutive cases of patients with tympanomastoid paraganglioma (TMP) tumors managed at a single tertiary referral center with 5 years experience.

Study Design: Retrospective review.

Setting: Bangabandhu Sheikh Mujib Medical University, a tertiary referral center in Bangladesh.

Methods: Between November 2014 and May 2019, 10 patients with histologically confirmed TMP tumor underwent surgical treatment. Tumor stage was described using the Sanna modified Fisch and Mattox's classification system.

Results: Distribution of tumors according to modified Fisch and Mattox classification was as follows: type A2 1 case (10%); B1 2 cases (20%), B2 6 cases (60%) and B3 1 case (10%). Class A2 tumour was safely removed via postauricular-transcanal approach. Two patients with Class B1 tumors were operated on through canal wall up mastoidectomy approach. Six patients including five Class B2 and one Class B3 tumors were managed by canal wall down mastoidectomy approach. One Class B2 underwent a subtotal petrosectomy with blind sac closure of the external auditory canal and middle ear obliteration. Gross total tumor removal was achieved in 9 cases (90%). One patient developed facial weakness (HB grade III) after one week of postoperative period which recovered completely by conservative treatment. No recurrence was noted in follow-up period.

Conclusion: Early diagnosis of tympanomastoid paragangliomas are very rare because of its benign and slow-growing nature. Clinical differentiation between tympanojugular and tympanomastoid paragangliomas are difficult. Surgery is the recommended primary modality of treatment for tympanomastoid paragangliomas with minimum morbidity and recurrence rate.

Key words: Paraganglioma, Tympanomastoid, Tympanojugular, Glomus, Middle ear tumor.

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Introduction:

Parangliomas are tumors that arise from proliferation of paraganglionic chief cells derived embryologically from the neural crest and found throughout the body associated with vascular and neuronal adventitia. Parangliomas of the head and neck accounts for 0.6% of head and neck tumors with most frequent carotid body tumour followed by vagal, tympanojugular and tympanomastoid paraganglioma.¹ Stacy R. Guild first noticed the presence of paraganglionic tissue or glomus formation within the temporal bone.² Parangliomas arise from glomus body along the Jacobson's nerve and Arnold's nerve are known as glomus tympanicum or tympanomastoid paraganglioma (TMP), and those arise from paraganglionic tissue along the adventitia of jugular bulb are known as glomus jugulare or tympanojugular paraganglioma (TJP).³ The term glomus is a misnomer. Earlier it was believed that the chief cell of glomus or paraganglioma derived from specialized pericytes or from blood vessel walls that are seen in true arteriovenous or glomus complexes which was proved false.⁴ So Paranglioma is being used instead of glomus tumour in the most of current literature. Though rare in incidence tympanic or tympanomastoid paraganglioma is the most common primary neoplasm of middle ear.⁵ This tumour confines to middle ear and mastoid

compartment without erosion of jugular plate or involvement of jugular bulb. The tumour may be asymptomatic and undiagnosed in very early stage of its origin from tympanic plexus. When the growing tumour touches the umbo, it begins to transmit pulsations to tympanic membrane and patient develops pulsating tinnitus.⁶

The patient usually presents with conductive hearing loss, pulsatile tinnitus and aural fullness in early stage. Otoendoscopic or otomicroscopic examination reveals a dark red or purplish pulsatile mass behind the intact tympanic membrane. High resolution CT scan is the investigation of choice for identification of possible tumour origin and its extension to adjacent structures.⁵⁻⁷ It helps in designing surgical approach before surgery. Surgery is the definitive primary treatment option in most of the cases of tympanomastoid paraganglioma.^{5,7,8,9} In the present study we report our experience of surgical treatment of 10 cases of tympanomastoid paraganglioma from Bangabandhu Sheikh Mujib Medical University, Dhaka, Bangladesh. To the best of our knowledge this may be the first reported large series of paraganglioma from a single center in Bangladesh. The authors followed the Modified Fisch and Matttox classification of tympanomastoid paragangliomas for operational definition.

Modified Fisch and Matttox classification of tympanomastoid paragangliomas:

Class Description

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- A. Tumors limited entirely to the middle ear
 - A1 Tumors completely visible on otoscopic examination
 - A2 Tumor margins are not visible on otoscopy. Tumor may extend anteriorly up to the Eustachian tube and/or to the posterior mesotympanum
 - B. Tumors limited to the tympanomastoid segment (middle ear cleft) of the temporal bone
 - B1 Tumors filling the middle ear with extension into the hypotympanum and tympanic sinus
 - B2 Tumors filling the middle ear with extension into the mastoid and medially to the mastoid segment of the facial nerve
 - B3 Tumors filling the middle ear with extension into the mastoid with erosion of carotid canal
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Methods:

The medical records of 10 patients with a diagnosis of Tympanomastoid Paraganglioma (TMP) who were managed at the Bangabandhu Sheikh Mujib Medical University, Dhaka, Bangladesh

between November 2014 and May 2019 were reviewed. Patients age, sex, presenting symptoms and the duration of these symptoms prior to diagnosis, clinical findings, side and site of the tumors were noted. Pre-operative and post-operative Otoendoscopic and or Microscopic findings were documented with photograph and video recording system. Audiological and radiological data were documented and analyzed. All patients underwent preoperative CT scan with iodinate contrast medium and four patients also underwent gadolinium (Gd) enhanced magnetic resonance imaging (MRI) to exclude jugular bulb and carotid involvement.

Urinary vanilylmandelic acid (VMA) was tested in four cases. Pure tone averages (PTA) for airconduction (AC) and bone conduction (BC) were calculated before and after surgery as the mean of 500, 1000, 2000 and 4000 Hz thresholds. The pre-operative and postoperative facial nerve (FN) function was graded according to the House-Brackmann (HB) grading system.¹⁰

The tumor was staged according to the classification proposed by Sanna known as modified Fisch and Mattox's classification.⁷⁻⁸ The type of procedure and the operative findings were recorded. Any operative complications, both immediate and delayed, were noted. Follow-up was defined as the period of time from surgery to the most recent office visit.

Surgical technique

Controlling bleeding and getting adequate exposure is the prime issue when excision

of tympanomastoid paragangliomas is executed. A very good nonstick bipolar diathermy is very useful in this regard. We followed the surgical technique proposed by Mario Sanna.⁷ In case of Class A2 tumour the retroauricular-transcanal approach was applied. A transcanal approach was established via a retroauricular incision. The tympanomeatal flap was elevated. The tympanomeatal flap consists of meatal skin, annulus and tympanic membrane was detached carefully from the bony meatus, the tumor and the malleus, and preserved in saline solution. This is known as glove finger flap technique. A wide canalplasty was performed after removal of the glove finger flap in order to get complete exposure of the tumor. The technique of tumor removal comprised bipolar coagulation and blunt dissection. The vascular pedicle (inferior tympanic branch of the ascending pharyngeal artery) is coagulated carefully. The temporal fascia was placed under the bony annulus as in the underlay technique after packing the tympanic cavity with gelfoam. The glove finger flap was carefully replaced over the graft adapting it to the enlarged external auditory canal through some radial skin incisions. All ossicles were preserved in one case. The incus was removed and repositioned in another case. In case of Class B1 tumour canal wall up mastoidectomy (CWUM) approach was used. Tympanomeatal flap was managed as glove finger flap technique manner. Epitympanotomy was done for better exposure of epitympanum. In case of B2 tumours conventional canal wall down mastoidectomy was performed. In one revision case subtotal petrosectomy with blind sac closure technique was applied. In one Class B3 tumour canal wall mastoidectomy with tympanoplasty was done to preserve hearing

Results:

Out of 10 patients with tympanomastoid paraganglioma (TMP) who were managed at BSMMU, a tertiary referral center in Bangladesh, 6 (60%) were female and 4 (40%) were male providing female/male ratio 3:2. The patients' ages ranged from 15 to 60 years with mean age of 37.5 years. 6 of the 10 (60%) patients were in the age range of 25 to 50 years. The tumour was found in the right ear in 7 cases (70%) and in the left ear in 3 cases (30%). Presenting symptoms ranged from just fullness in the ear to frank bleeding from the ear. Pulsatile tinnitus and hearing loss were the most common symptoms found in all 10 (100%) cases of our patients. 5 (50%) of our patients had discharge or fresh bleeding from ear when they

first presented to us suggesting a relatively late presentation with aural polyp or with previous history of biopsy or surgery. A pulsatile reddish mass was seen behind the intact tympanic membrane in 3 (30%) of the patients while remaining 7 (70%) patients presented with polypoidal pulsatile mass in external auditory canal. (Table II)

Table I :
Presentation and Examination Findings.

Finding	n (%)
Presenting symptoms	
Pulsatile tinnitus	10 (100)
Subjective hearing loss	10 (100)
Aural fullness	10 (100)
Bleeding /Otorrhea	6(60)
Facial paralysis	0 (0)
Asymptomatic	0 (0)
Examination findings	
Pulsatile mass with intact tympanic membrane	3(30)
Violation of tympanic membrane	7 (70)

The right was involved in 7 (70%) patients and the left ear in the other 3 (30%). None of the patients had symptoms or signs that can

be attributed to either neurosecretory tumor or involvement of facial cranial nerve or lower cranial nerves. Tumors were classified according to modified Fisch and Mattox's classification as follows: A2 1 (10%) case, B1 2 (20%), B2 6 (60%) cases and B3 1 (10%) case. (Table III).

Table II :
Baseline Population Characteristics.

Characteristic	Mean (Range) or n (%)
Age at diagnosis, years	37.5 (15 - 60)
Female sex	6 (60)
Male	4(40)
Malignant	0 (0)
Secretory	0(0)
Primary or recurrent disease	
Primary	8 (80)
Recurrent	2 (20)
Right-sided laterality	7 (70)
Tumor type (stage)	
A2	1(10)
B1	2(20)
B2	6 (60)
B3	1(10)

Eight patients underwent a primary operation and 2 patients presented with a recurrent tumor operated on elsewhere by a canal wall down mastoidectomy. The retroauricular-transcanal approach was applied for one patient with a tumour confined to middle ear (Class A2). A canal wall up mastoidectomy (CWUM) was used in two patients (Class B1). Total ossicular chain was maintained in two cases. Stapes suprastructure was preserved in two cases where in one case ossiculoplasty was done with repositioned autologous incus and other case with conchal cartilage. Six patients (Class B2) required canal wall down procedure. One patient underwent a subtotal petrosectomy with blind sac closure of the external auditory canal and middle ear obliteration.

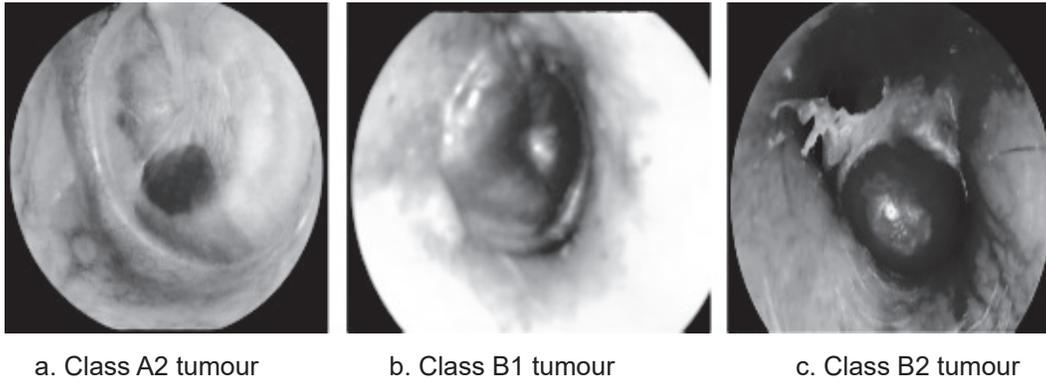


Fig. 1 (a,b,c) : *Otoendoscopic findings of different classes of Tympanomastoid Paraganglioma tumours*

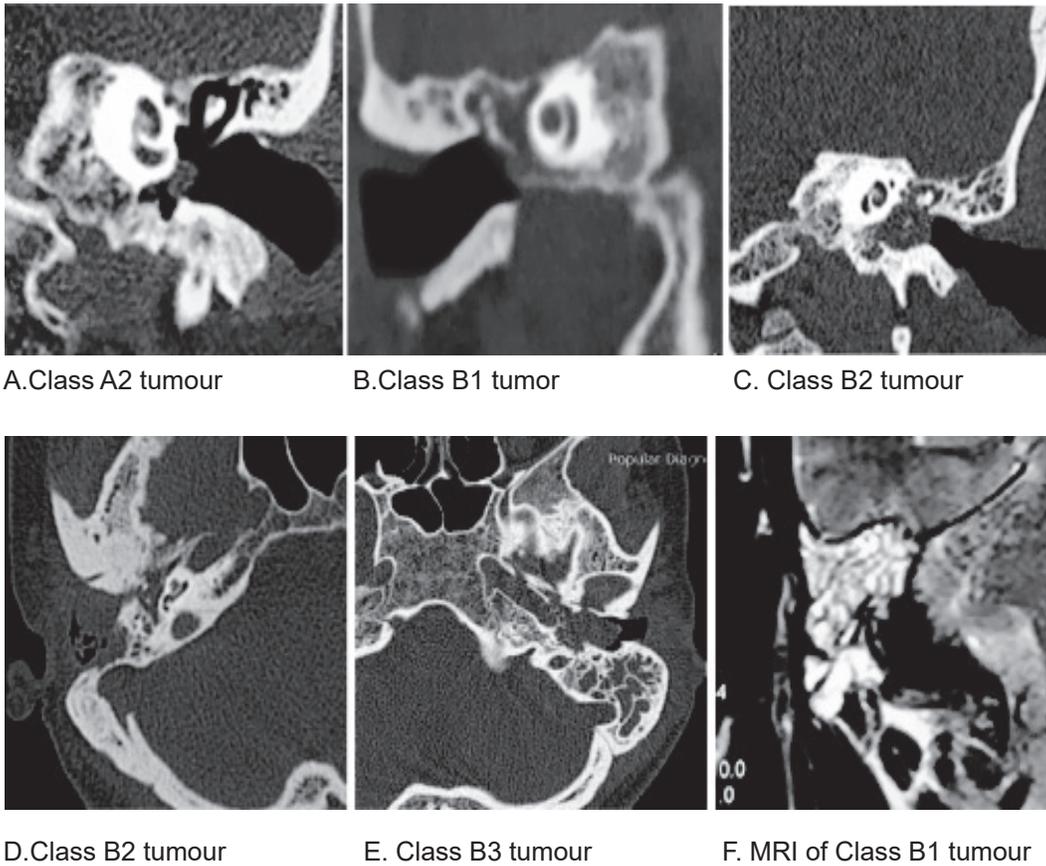


Fig. 2: *Computed tomography(CT) scans illustrating the different classes of tympanomastoid paraganglioma according to Sanna modified Fisch-Mattox classification. A,B,C –Coronal CT and C,D –Axial CT, F-MRI to see involvement of jugular bulb*

Gross tumor removal was achieved in 9 cases. Small residual tissue around the carotid canal in was left in one case. (Table III)

Table III :
Operative Details

Detail	n (%)
Surgical approach	
Postauricular-transcanal approach	1 (10)
CWU Mastoidectomy	2(20)
CWD Mastoidectomy	6(60)
STP with MEO	1(10)
Extent of resection	
Gross total	9 (90)
Near total	1 (10)

Abbreviations: CWD, canal wall down; CWU, canal wall up; STP with MEO Subtotal petrosectomy with middle ear obliteration

Postoperative complications were noted in 3(30%) cases. One patient developed facial weakness (HB grade III) after one week of postoperative period which recovered completely by conservative treatment with steroid and antibiotic over next three weeks. Non healing granulation tissue was seen anterior quadrant of middle ear in a case of radical mastoidectomy after 8 months of surgery. Examination under general anesthesia revealed a piece of bone wax in Eustachian tube area which was removed along with adjoining granulation tissue. Dry and epithelialized middle ear and mastoid cavity was found in follow up visit after one month. Another patient of CWUM developed pars tensa retraction pocket Cholesteatoma after 3 years of surgery. The case was managed by Canal wall down (CWD) mastoidectomy, and a dry and healthy well epithelialized middle ear mastoid cavity was achieved.

Pre-operative and post-operative hearing were analyzed. In pre-operative pure tone audiometry 5(50%) patients had moderate conductive hearing loss, 2(20%) severe mixed hearing loss and 3(30%) patients profound sensorineural hearing loss. Out of five conductive hearing loss cases three noticed improved hearing and two did not do post-operative audiometry.

Discussion:

Tympanomastoid paragangliomas (TMP) are slow-growing vascular middle ear mass. Because of its indolent nature, diagnosis is usually delayed until it reaches in significant size. Though it is benign in nature, it invades the adjoining bone through haversian systems and air cells.^{4,6,11} Pulsatile tinnitus (75-83%) and hearing loss (73-80%) are the most common presenting complaints of TMP patients in majority of reported literature.^{4-8,11} In our series 100% patients (10/10) presented with pulsatile tinnitus and hearing loss. A female preponderance of more than 90% with female/male ratio of 9:1 is reported in maximum literature.^{5,7,8,12} There were 6(60%) females and 4(40%) males with female/male ratio of 3:2 in our series. The mean age at diagnosis was 37.5 years in this study which is dissimilar with other studies where mean age was 51- 56.2 years^{5, 11,13,15}

Presence of a pulsatile reddish mass behind the intact tympanic membrane is the characteristic finding of tympanic paraganglioma. This finding is seen when tumour is confined to middle ear. When the tumour extends to external ear canal as polypoidal mass or if there is previous history of biopsy or surgery other differential diagnosis including inflammatory polyp, aberrant internal carotid artery, high jugular bulb, haemangioma, facial nerve neuroma etc. should be kept in mind. In this series 3

patients had pulsatile mass behind intact tympanic membrane and other 7 patients presented with pulsatile polypoidal mass in external ear canal. Unless the clear margin around the tumour is visible on otoendoscopy or microscopy, differentiation of tympanic paraganglioma from jugular paraganglioma is clinically impossible.^{6, 11, 14}

A slight left sided predominance (51.5-56.2%) was noted in some reported series^{7,15} and right sided in other studies.^{5,11} In our study it is predominantly lateralized to right (70%).

Histopathology is the confirmatory for diagnosis of paraganglioma but preoperative biopsy is contraindicated.^{5-8,11,14} In 8(80%) of our primary case preoperative biopsy was not performed.

Neurosecretory function is a very rare entity in tympanomastoid paraganglioma. Historically, preoperative venous sampling was encouraged in all cases to avoid inadvertent situation during operation.¹¹ In study of 115 cases of tympanic paragangliomas, only one tumour was associated with catecholamine secretion in a patient who had history of refractory hypertension¹³. In another large series of 133 such tumours, no single secretory lesion or intraoperative unwanted cardiovascular crisis was reported.¹⁵ So routine preoperative biochemical analysis and urinary VMA were not advocated in most literature in absence of high risk factors such as, younger age at presentation (<40 years), male sex, family history of disease, signs and symptoms of multicentric disease.^{5,8,11,13} Out of 10 cases, urinary VMA was tested in 4 cases(40%). None of them was positive for secretory function.

Pure tone audiometry may reveal conductive, mixed or sensorineural hearing loss depending upon the involvement of

middle ear or inner ear. In a study of 18 cases¹⁴ it was reported conductive hearing loss 9(50%), mixed hearing loss 3(30%) and severe to profound sensorineural hearing loss in 4(40%) cases. In our series conductive, mixed and sensorineural hearing loss are 50%, 20% and 30% respectively. Out of five conductive hearing loss cases three noticed improved hearing and two did not do post-operative audiometry.

High resolution CT scan with contrast is an essential imaging tool for diagnosis and extension of tumour to surrounding vital structures including jugular bulb, carotid artery and facial nerve. Differentiation of tympanomastoid paraganglioma from jugular paraganglioma is of critical element for preoperative surgical planning as latter requires quite different surgical approach – Infratemporal Fossa Type A (IFTA).^{5,7} If there is a thin plate of bone or air between tumour and jugular bulb, it confirms the confinement of tumour in the middle ear.^{7,8,16} CT scan can help to rule out other less common conditions such as high-riding jugular bulb, an aberrant internal carotid artery, invasive intracranial tumors, encephalocele, endolymphatic sac tumors centered on the posterior petrous face, or facial nerve schwannomas, facial nerve haemangioma with a dilated fallopian cana.¹¹³ If the tumour mass in the middle ear reveals a soft tissue density surrounded by air in CT scan, no further imaging is necessary.^{5,16} Gadolinium-DTPA enhanced Magnetic Resonance Imaging(MRI) is necessary if the tumour involves the hypotympanum or suspected erosion of jugular bulb, carotid canal.^{5,7,8,11,16} In our series CT scan was done in all 10(100%) cases and MRI was done in 4(40%) patients to see carotid and jugular bulb involvement. Angiography is not recommended for tympanomastoid paraganglioma.^{5,7,8,11} We did not advice angiogram in any of our patients.

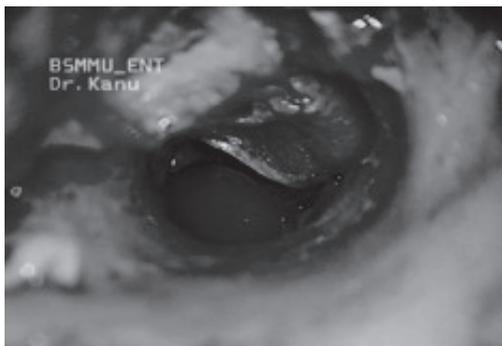
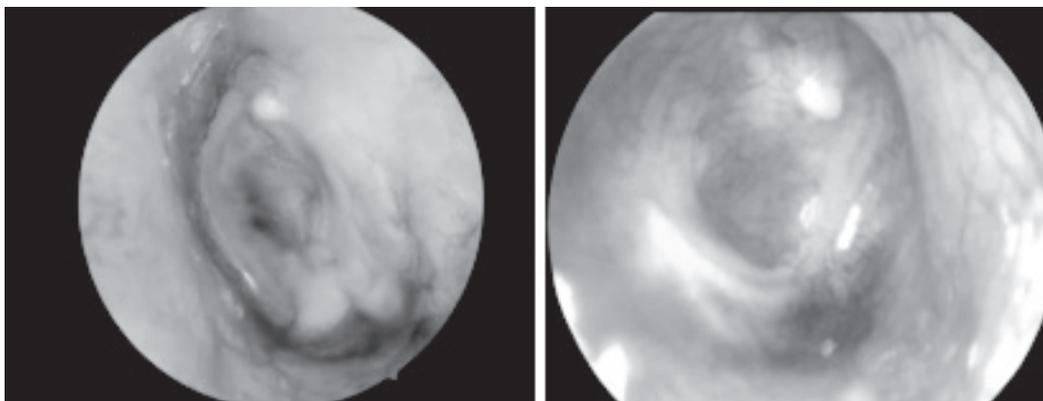


Fig. 3: Peroperative picture

workup. According to proposed algorithm followed at Gruppo Otologico one Class A2 case was managed by postauricular transcanal-transcanal approach (PA-TCA), two cases Class B1 tumours were excised through canal wall up (CWU) mastoidectomy. In this study five (50%) tumours (Class B2) including one revision were managed by canal wall down mastoidectomy and one by subtotal petrosectomy due to difficult exposure and bleeding although algorithm proposed canal wall up mastoidectomy with



a. Class A2 tumour post-op

b. Class B1 tumour post-op

Fig. 4 a,(b): Otoendoscopic post op follow up findings of Tympanomastoidparaganglioma tumour

Total surgical resection is the only curative treatment of tympanomastoid paraganglioma except few unwanted situations.^{5,7,8,11,15} With advancement of modern microsurgical technique and neuroimaging, total removal of tumour is possible with minimum or no postoperative sequelae.^{8,11} The TMP tumours are staged preoperatively for selecting appropriate surgical approach. There are different staging systems -Fisch-Mattrox, Glasscock-Jackson and Sanna modified Fisch-Mattrox for tympanic and paraganglioma.¹¹ We have followed Sanna modified Fisch-Mattrox classification for staging and preoperative

posterior tympanotomy and subfacial recess tympanotomy for B2 tumours. In a study 44% of patients with B2 tumours had to manage with subtotal petrosectomy.⁸ Usually B3 tumours need subtotal petrosectomy (STP) with middle ear obliteration (MEO).^{7,8} One B3 tumour in our series underwent canal wall down mastoidectomy with type III tympanoplasty to preserve or improve hearing. In this case the preoperative conductive hearing loss was improved in postoperative period.

Literature reported that complete resection of tympanomastoid paragangliomas was possible in 94-100% of Class A and B

tumours with 0% to 5% recurrence rate.^{5,7,17} In our series total tumour removal was achieved in 9 (80%) cases. Seven(70%) cases are in regular follow up, three lost to follow up after one year of surgery and on recurrence has been detected.

Radithery has been suggested as primary alternative to surgery in tympanic paraganliomas some few studies.^{18,19,20} Most authors agreed that radiotherapy is not recommended in tympanomastoid paragangliomas limited to middle ear and mastoid rather it may develop osteo-radionecrosis, stenosis of external ear canal, radiation induced neoplasm.^{5,7,8,11-14,17} Wait and scan or radiotherapy may be an alternative option in recurrent or residual tumor , patients with risks for general anesthesia and in elderly patients.^{5,7,8,11,21,22}

Conclusion:

Early diagnosis of tympanomastoid paragangliomas are very rare because of its benign and slow-growing nature. Clinical differentiation between tympanojugular and tympanomastoid paragangliomas are difficult as both have same clinical findings of reddish pulsatile mass and tinnitus. High resolution CT scan of temporal bone is the investigation of choice for preoperative evaluation, staging and selecting definitive surgical approach. MRI is reserved for selective cases of suspicion of jugular bulb and/or carotid artery involvement. Surgery is the recommended primary modality of treatment for tympanomastoid paragangliomas with minimum morbidity and recurrence rate.

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