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# Chronic Suppurative Otitis Media : A Comparative Microbiological Review of India with other Developing Countries.

# Saurabh Varshney<sup>1</sup>, Pratima Gupta<sup>2</sup>, Aroop Mohanty<sup>3</sup>, Shyam Kishor Kumar<sup>4</sup>, Mithilesh Kumar Jha<sup>5</sup>, Ankita Kabi<sup>6</sup>

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### Abstract

Chronic infection of the middle ear cleft for at least 2 weeks or more is known as Chronic Suppurative otitis media. It usually manifests as a complication of acute otitis media. It is more prevalent in developing countries due to various predisposing factors like malnutrition, overcrowding, poor hygiene, inadequate health care, and recurrent upper respiratory tract infection. Due to advancement in medical facility, India still falls under high prevalence zone. In India, average prevalence of chronic Suppurative otitis media is 5.2%. *Pseudomonas aeruginosa* and *Staphylococcus aureus* are the predominant isolates in most of the studies conducted in India as well as in other developing countries of the world. In case of fungal aetiology *Candida spp* and *Aspergillus spp* were the predominant isolates.

Keywords: Chronic Suppurative otitis media; Aerobic isolates; Developing countries.

### Introduction

"Chronic suppurative otitis media (CSOM) as stage of disease in which there is chronic infection of the middle earcleft, in which a nonintact tympanic membrane (e.g, perforationor tympanostomy tube) and discharge (otorrhoea) are present for at least 2 weeks or more".<sup>1</sup> It is also known as chronic active mucosal otitis media, chronic otomastoiditis or chronic tympanomastoiditis. This is usually a disease of childhood occurring as asequel of acute otitis mediapresenting most commonly with hearing loss and suppurative drainage.<sup>2</sup> It is more prevalent in developing countries due to malnutrition, overcrowding, poor hygiene, inadequate health care, and recurrent upper respiratory tract infection.<sup>3</sup> It can lead to

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irreversible complications like persistent otorrhoea, mastoiditis, labyrinthitis, and facial palsy to more serious intracranial abscesses or thromboses.<sup>4</sup> Hence an early diagnosis of the causative agent and prompt effective treatment are necessary to avoid such complications.

The most common aerobic bacterial isolates found in CSOMare *Pseudomonas aeruginosa*, *Escherichiac oli, Staphylococcus aureus, Streptococcus pyogenes, Proteus mirabilis, Klebsiella spp Candida spp* and *Aspergillus spp*. are the common fungal isolates. However, this may vary according to geographical areas and other factors.<sup>5</sup>

Topical antibiotics in combination with aural toilet are the mainstay of therapy for uncomplicated CSOM.<sup>6</sup> However, widespread irrational use

of antibiotics has led to multidrug resistance organisms which in turn pose complications making treatment of CSOM more difficult. In developing countries, multidrug resistance organisms are rapidly increasing due to indiscriminate use of antibiotics, overcrowding, poor hospital hygiene and lack of resources and personnel trained in infection control.<sup>4</sup> Thus, there is urgent need of speciation of isolates along with their antibiotic resistance pattern.

This systemic review aims to compare the aetiology of CSOM of India with that of other developing countries. We also investigated for any changing trends of CSOM aetiology in our country.

# Materials and Methods

# Data sources and searches

Published original articles and abstracts in English were searched through google, scholar and PubMed search engine. Data sources were included from all parts of India and other developing countries from 2009 to 2019 and included studies on CSOM. Exclusion criteria were:

- i) Study of CSOM with complications and
- ii) Case reports, letter to editors and intervention articles.

We have searched original articles from all parts of India and 10 from other neighboring developing countries ranging from 2009 to 2019 through various search engine. We had 32 Indian studies and 9 studies from 8 countries related to prevalence and microbiological profile of CSOM.

# Results

# Demographic profile

We found 13 studies which discussed prevalence of CSOM (Table 1) in school going children. The prevalence ranged from 1. 4% to 6.6%. Males outnumbered in most of the studies

# Microbiological profile

A wide range of aerobic bacteria were isolated in different studies of India (Table 2 and 3). *Pseudomonas aeruginosa* and *Staphyococcus aureus* were the predominant isolates (19.9%–67.5% and 11.3%-51.9% respectively), others were *Klebsiella*  *spp*, *Proteus spp*, *Escherichia coli* and few studies also isolated *Serratia marcescens* and *Shewanella spp*.<sup>14-16</sup>

*Candida spp* and *Aspergillus spp* were the common fungal isolates in most of the studies (Table 4). Only Juyal et al. reported *Penicillium spp and Mucor* in their study.<sup>17</sup>

In other developing countries, Pseudomonas aeruginosa andStaphyococcus aureuswere the predominant isolates. However, some studies isolated Proteus vulgaris and Proteus mirabilis as predominant isolates.<sup>18,19</sup>*Aspergillus spp* and *Candida spp* were the common fungal isolates (Table 5).

# Discussion

# Prevalence

According to WHO/CIBA workshop of otitis media experts in 1996, countries having prevalence rate of 1-2% were considered under low prevalence zone while countries having 3-6% prevalence rate were considered as high prevalence zone.13In India, since average prevalence of CSOM is 5.2%, it falls under high prevalence zone.1In most of the other developing countries, prevalence of CSOM falls more than 3% as like in India which is indicative of high prevalence zone. Only Latin America Andeanregion shows less than 2% of prevalence rate (Figure 1).44 In this systemic review a study from 1961 showed a high prevalence of 14.7% <sup>45</sup>, whereasin the last decade7 studies taken from different parts of India found prevalence to have decreased to 6.6%8. Based on these studies, it may be concluded that prevalence of CSOM has changed in past many years probably due to improved health care seeking behavior and more awareness. There are many other factors which determine prevalence of CSOM like- younger age, male sex, parental or sibling history of otitis media, early onset of otitis media, low socioeconomic strata, reduced duration of breast feeding, over crowding, day-careat tendance, recurrent upper respiratory infections, allergic rhinitis, ignorance regarding ear diseases, lack of medical facility, passive smoking and different geographical location.7,11. In India, as like other developing countries ( Nepal, Bangladesh, Pakistan etc.), low socioeconomic condition is associated with progression of CSOM. Lower socioeconomic class people generally live in congested room and maintain poor personal hygiene which favor transmission of infectious agents. Poor living condition is also associated with malnutrition resulting in depressed immune system which causes increased risk of disease.46

Author	Study year	Place of study	Prevalence
Sophia et al. 2010 <sup>7</sup>	July 2006–April 2007	Tamil Nadu	1.4%
Abhinav et al. 2014 <sup>8</sup>	1 yr. period	Maharashtra	6.6%
Chadha et al. 2015 <sup>9</sup>	2010 - 2011	Delhi	3.6%
Parvez et al. 2016 <sup>10</sup>	Aug 2010 – July 2011	Aligarh	6.1%
Garud et al. 2016 <sup>11</sup>	Jan 2012 – Jan 2013	Maharashtra	6.4%
Ray et al. 2017 <sup>12</sup>	2011	Delhi	3.6%
Singhal et al. 2018 <sup>13</sup>	July 2016 – Sep 2016	Uttar Pradesh	6.5%

**Table: 1** Showing prevalence of CSOM in India.

Table: 2 Aerobic isolates in CSOM in India (data are expressed in percentage).

Reference No/year	20 2009	21 2010	22 2010	23 2011	24 2012	25 2012	26 2013	27 2013	28 2013
Isolates									
P aeruginosa	32.4	67.5	28.3	45.5	32.2	45.2	33	19.9	54
S aureus	21.2	-	29.2	37.7	17.4	22.2	25.8	48.7	11.3
CONS	-	-	-	-	-	-	-	-	8.1
P vulgaris	5.2	-	-	-	0.9	4.8	-	2.1	6.5
P mirabilis	-	-	7.5	1.3	2.6	6.4	20.6	-	8.1
E coli	7.6	17.5	-	1.3	1.7	4.8	-	7.3	3.3
E aerogenes	-	-	2.8	-	0.9	-	4.1	-	-
Klebsiella spp	10	-	3.7	9.1	6.9	6.4	4.1	9.4	8.1
ß hemolytic Streptococcus	-	-	3.7	2.9	-	-	4.1	-	1.6
S pneumonie	-	-		-	1.7	-		1.1	-
Acinetobacter spp	3.2	-	1.9	-	1.7	2.4	1	3.1	-

Table 3: Aerobic isolates in CSOM in India. (data are expressed in percentage)

Reference No/ year	14 2015	29 2015	15 2016	30 2016	31 2016	16 2017	32 2018	33 2018	5 2018	34 2018	35 2018
Isolates											
P aeruginosa	37.9	33.1	30.8	34.3%	35	31.1	37.5	56.1	38.5	20	34.9
S aureus	32.6	21.6	29.8	28.6%	20.5	31.8	5	24.03	51.9	51.5	27.2
CONS	-		2.9	-	10.8		-		1.9	-	-
P vulgaris	6.3	-	-	-	-	2.2	10	4.1	-	3.1	12.6
P mirabilis	-	5.8	6.7	7.6%	4	0.7		-	-		-
E coli	4.2	3.6	1.9	-	3.6	7.4	5	6.01	-	7.7	7.8
E aerogenes/spp	-	2.2		-		-	-	-	-	-	-
Klebsiella spp	-	9.4	10.6	7.6%	8.4	2.2	5	6.01	3.8	8.5	9.7
ß hemolytic Streptococcus	-	-	0.9	3.8%	-	-	-	-	-	-	-
S pneumonie	2.1	-	-	-	5.6	0.7	-	-	3.8	0.8 (alpha)*	4.9
Acinetobacter spp	3.2	1.4	-	1.9%	-	6.75	-	1.7	-	3.4	-

Reference no/year Isolates	23 2013	24 2013	27 2016	26 2015	17 2014	29 2018	36 2013	5 2018	35 2017
Candida spp	-	-	2.6	-	-	-	2.3	-	-
Candida albicans	9.8	0.9	1.9	1.03	23	4.3	-	3.1	4.3
Candida glabrata	-	1.7	-	-	-	-	-	-	-
Candida parapsilosis	-	-	-	-	4.8	-	-	-	-
Candida tropicalis	-	-	-	-	10.8	-	-	-	-
Candida krusei	-	-	-	-	2.4	-	-	-	-
Aspergillus spp	-	-	-	-	-	-	5.4	-	-
Aspergillus niger	2.2	3.5	5.8	2.1	14.5	6.5	-	10.8	4.3
Aspergillus flavus	1.1	3.5	-	3.1	20.5	2.9	-	4.6	1.7
Aspergillus fumigatus	3.3	-	5.2	-	8.4		-	1.5	0.8
Aspergillus terreus	-	-	-	-	3.6		-	-	-
Penicillium	-	-	-	-	9.6		-	-	-
Mucor	-	-	-	-	2.4				

Table 4: Fungal isolates in CSOM in India. (data are expressed in percentage)

 Table 5: Aerobic isolates in CSOM in other developing countries (data are expressed in percentage).

Country / Reference No	Sri Lanka 37	Bangladesh 38	Pakistan 39	Nepal 40	Malawi 41	Iran 42	Luanda 18	Ethiopia 43	Ethiopia 19
Isolates									
S aureus	20.5%	29.1%	<b>65.2</b> %	36.1%	20.1%	29.2%	2.6%	15.8%	16.2%
CoNS	-		-	8.8%	4%	35.7%	7.3%	-	16.2%
S pyogenes	-	14.9%	-	-	-	-	1.1%	7.9%	-
S pneumonie	-	1.6%	-	-	-	-	-	8.9%	-
Viridans Streptococci	-	-	-	-	-	-	-	9.9%	-
Streptococcus spp	-	-	-	-	-	-	2.2%	-	-
Enterococcus	2.5%	-	-	-	-	-	8.8%	6.3%	-
P aeruginisa	29.5%	22.8%	15.2%	33.3%	20.8%	10.3%	13.2%	10.9%	-
Pseudomonas spp	-	-	-	-	-	-	1.9%	-	-
Escherichia coli	-	9.4%	0.65%	2.3%	8.4%	-	2%	7.4%	-
K neumoniae	-	4.7%	-	6.01%	4%	12.9%	-	7.4%	-
Klebsiella spp	-	-	-	1.4% (oxytoca)	-	-	3%	-	13.5%
P mirabilis	-	6.3%	13.1%	4.2%	28.6%	-	-	7.9%	21.6%
P vulgaris	-	-	-	0.4%	4%	-	-	-	2.7%
Proteus spp	-	-	-	-	-	10.3%	14.7%	-	-
E cloace/spp	-	-	-	0.9%	2.6%	-	2%	3.2%	5.4%
coliforms	16.7%	-	-	-	0.6%	-	-	2.1%	-
Citrobacter spp	-	-	-	0.4%	-	-	3.4%	-	6.8%
Acinetobacter spp	-	-	-	3.2%	-	-	-	7.9%	-
A fumigatus	-	6.3%	-	-	-	-	-	-	-
Candida	7.7%	4.7%	-	-	-	-	-	-	-

Male dominance was seen in most studies which may be due to males reporting more than females in the hospital or because malesare more exposed to outdoor activities.



CSOM incidence rate estimates for the year 2005 per thousand people, by the 21 WHO regions.44doi: 10.1371/journal. Pone. 0036226.g004

# Microbiological profile

Pseudomonas aeruginosa and Staphylococcus aureus are the most common isolates in most of the studies from different geographical regions of India as well as other developing countries. There are many favorable factors which help colonization of Pseudomonas aeruginosa over other bacteria like minimum nutritional requirement for survival, relative resistant to antibiotics and its antibacterial products i.e., pyocyanin and bacteriocin. One postulate by Vartiainen and Vartiainen that Pseudomonas is capable of maintaining a niche for itself in local infection through necrotising activities of its extracellular enzymes. The niche consists of damaged epithelium, interrupted circulation and devitalized tissue that protects the organism from normal host defence mechanisms and antibiotic agents. Besides these, the organism acts as an opportunistic pathogen, flourishes in the external auditory canal and may cause suppurative disease in contiguous sites.<sup>21,22</sup> Staphylococcus aureus was the second most common organism in most of these studies where as few described Staphylococcus aureus as the most common isolate. Other bacterial isolates were Klebsiella spp, Proteus spp, Escherichia coli, Enterobacter, Acinetobacter spp, Citrobacter spp, beta hemolytic Streptococci and Streptococcus pneumoniae. The variations in bacteria isolation rates of different organisms reported by different workers may be as an effect of inappropriate antibiotic uses, climatic and other geographical factors.33 Few studies also isolated Serratia marcescens and Schewanella sppwhich may be due to improved isolation/identification techniques.14,16,21

et al. found highest incidence of fungal infection in second and third decade of life.17 Jitendranath et al. also found in third and fourth decade of life. 29 Probably, these age groups are more exposed to fungal spores as compare to extreme age groups. Aspergillus spp were common isolates in most of the studies. Aspergillushas capability of growing rapidly and produces a large number of small, dry, easily aerosolized conidia. These conidia can easily contaminate the air. Their growth is further facilitated by cerumen and the slightly acidic pH of the ear canal.<sup>29</sup>Candida spp were the second common isolates. Non albicans Candida, which are less pathogenic than Candida albicans, were also isolated. Jitendranath et al. found higher incidence of fungal infections during rainy season followed by summer season. Hot and humid climate and presence of dust in the environment facilitates the fungal growth.47

# Conclusion

*Pseudomonas aeruginosa* and *Staphylococcus aureus* are the common isolates in India. There is no significant change in prevalence for these two bacteria. Even outside India, these two bacteria are more common in most of the studies. There are some other isolates in different zones which may indicate local geographical or climatic variation or impact of inappropriate antibiotic use. *Candida spp* and *Aspergillus spp* are only fungal isolate in almost all studies.

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# Sinonasal Mucormycosis: A Cryptic Paradox

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#### Abstract

Mucormycosis is one of the sinister variant of fungal rhinosinusitis. An immunocompromised case with mucormycosis requires prompt diagnosis, in order to prevent a fatal outcome. Though the primary cause lies in the nose and sinus, due to lack of awareness, patients usually visits a physician, an ophthalmologist and may be then the otorhinolaryngologist, during this period, patient would have progressed to an advanced stage missing the early intervention. This case report of 70 year old male with mucormycosis of left side of the nose and sinus with paradoxical involvement of right eye and has been successfully treated with repeated endoscopic clearance of the crusts and diabetic control. Since the disease has a stormy course and rapid progression, an early diagnosis and aggressive management is imperative and all the more so when the presentation is paradoxically elusive to avoid complications.

Keywords: Mucormycosis; Amphotericin; Fungal

# Introduction

Sinusitis or more accurately rhinosinusitis is a common disorder, affecting approximately there are 20% of the population at some time of their lives. 400,000 known fungal species of which 400 are human pathogens and 50 of which cause systemic or CNS infection. Once a rare disorder, is now being identified and reported with increasing frequency over the last two decades.<sup>1,2</sup> Increased diagnostic methods enables us for more frequent recognition. It carries a high residual morbidity and mortality due to the angioinvasive property of fungi, causing vascular occlusion and extensive tissue

necrosis.<sup>3</sup> Functional endoscopic sinus surgery with repeated suction clearance still remains the main stay of treatment with intravenous antifungal Amphoterecin.

# Case Report

We present a 70 year old male, reporting to physician for generalized weakness and was incidentally diagnosed with diabetes mellitus. He was referred to ophthalmologist for right eye swelling and reduced vision since 10 days and was subsequently referred to us for Left sided nose block since 10 days and headache since 8 days. He had associated symptoms of foul smelling discharge. *Nose examination*: Revealed thick crusts with foul smelling discharge filling both the nasal cavities.

*Diagnostic Nasal Endoscopy*: Thick crusts with tenacious, foul smelling discharge in the left nasal cavity, left maxillary and sphenoid sinus and with bony septal perforation.

Right nasal cavity, maxillary and sphenoid sinus normal.

*Ophthalmic examination*: On the contrary to the presence of disease on left side of nose and paranasal sinuses, in this case on examination, Right eye showed mild proptosis, chemosis, extraocular movement (right lateral) was restricted, with right eye reduced vision. Left eye Normal.

*CT Scan: PNS plain with contrast*: Showed crusts filling the entire Left nasal cavity, osteomeatal complex and involving entire Maxillary, Ethmoid and Sphenoid sinus with no breach in the lamina papyracea on the left side. Right nasal cavity and sinus appeared normal but there was soft tissue edema around the right orbit with no breach in the lamina papyracea.[Fig. 1]

# Management

Under general anesthesia. 0 degree endoscope was used to visualize bilateral nasal cavities



**Fig. 1**: A,B,C: Preoperative CT scan PNS images of Axial, Coronal and Sagittal views respectively, it shows thick tenacious discharge filling the left nasal cavity, maxillary, ethmoid and sphenoid sinus with no breach in the lamina papyracea. Right nasal cavity- Clear



D,E,F: Postoperative CT scan PNS images of Axial, Coronal and Sagittal views respectively images with bilateral clear nasal cavity and sinus.



**Fig. 3**: A-KOH mount - positive for fungal filaments. B-HISTOPATHOLOGY: mucosa showed edema, congestion and diffuse inflammation with necrotic material showing numerous hyphae branching at right angles suggestive of mucormycosis. A black eschar was seen involving the left osteomeatal complex, left maxillary sinus and left anterior ethmoidal cells and sphenoid sinus. Underlying bone was necrosed.

Hence, the diseased bone and the unhealthy mucosa were excised, the specimen was sent for KOH and histopathological examination.

He underwent repeated DNE and suction clearance of the crusts formed. His post op repeat CT scan showed clear left nasal cavity left maxillary sphenoid and ethmoid sinus as compared to pre op imaging [Fig. 2]

KOH: Positive for fungal filaments.[Fig. 3]

*Histopathology:* Mucosa showed edema, congestion and diffuse inflammation with necrotic material showing numerous hypha branching at right angles suggestive of mucormycosis. [Figure 3]

He was treated with IV antifungal of liposomal amphotericin B and fluconazole. Also, nasal douching with fluconazole twice a day.

His diabetes mellitus was managed with insulin.

He was asymptomatic following 2 months and 6 months follow up. His eye swelling was completely resolved [Fig. 4].



Fig. 2: Endoscopic Images

A: Shows thick white tenacious discharge filling the entire left maxillary sinus

B: Shows white discharge in the sphenoid sinus with erosion of inter sphenoidal septum

C: Bony septal perforation

D,E: Shows postoperative clear maxillary and sphenoid sinus respectively.



Fig. 4: A: Preoperative right eye swelling.B: Postoperative reduced eye swelling at 2 month follow up.

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# Discussion

Fungal infections of the NOSE and PNS have been recognized since the beginning of this century. Rhino/sino-orbital mycosis- most often caused by the saprophytic moulds - aspergillus and Mucorales<sup>6,7,9-12</sup>. Found worldwide in a variety of habitats in soil, on decaying vegetation, in the air, and in water supplies. Their thermotolerance permits a wide range of suitable host conditions.<sup>4,5,8,9</sup>. They are encountered more frequently in patients with immunocompromised status.4 They usually present with unilateral or bilateral nasal cavity involvement with ipsilateral or bilateral orbital involvement respectively, with or without intracranial involvement. Unlike in this case where the patient has presented paradoxically with contralateral eye involvement and ipsilateral eye being normal.

Amphotericin B remains the only antifungal agent approved for the treatment of this infection.<sup>4</sup> The lipid formulations of amphotericin B are significantly less nephrotoxic than amphotericin B deoxycholate and can be administered at higher doses for a longer period of time. Several case reports of patients with mucormycosis documented successful treatment with up to 15 mg/kg/day of a lipid formulation of amphotericin B.

Concurrent therapy: Azoles and Amphotericin-B better than monotherapy.

Though the primary cause lies in the nose and sinus, due to lack of awareness, patients usually visits a physician, an ophthalmologist and may be then the otorhinolaryngologist, during this period, patient would have progressed to an advanced stage missing the early intervention.

# Conclusion

An immunocompromised case with mucormycosis requires prompt diagnosis, in order to prevent a fatal morbidity. Delay in the treatment and risk of associated intracranial and intraorbital complications are major determinants of the survival outcome in these patients.<sup>5,6</sup> In these cases along with the ipsilateral eye examination, *it is important to examine the contralateral eye* also for the signs of endogenous spread or systemic spread of the disease, while monitoring it closely for progression, 7. It also needs to be emphasized that, high clinical suspicion from physician, otolaryngologist and ophthalmologist is a must, so that, the patient receives early intervention to avoid the dreaded complications.

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# Castleman Disease: An Unheard Entity in the Neck

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# Abstract

Castleman Disease (CD) is a nonclonal lymphoproliferative disorder that can affect either a single lymph node called unicentric or, can be generalized, multicentric. Though the disease has a wide spectrum of presentation, still there are no studies describing its natural history. Unicentric type is usually a chronic and asymptomatic swelling with neck being the unusual site as compared to multicentric type which presents with mediastinal lymph node involvement and constitutional symptoms. Here we report a 24 year male, presenting with long standing history of painless solitary gradually progressive swelling in the posterior triangle of the neck, which was treated successively with excisional biopsy and was diagnozed with histopathology which showed unicentric type of Castleman disease. He was evaluated further to rule out multicentric type of disease, no other swellings seen in the body.

Keyword: Castleman disease; Excisional biopsy; Unicentric.

# Introduction

Castleman Disease (CD) is a rare, poorly understood lymphoproliferative disease. It is otherwise known as "giant lymph node hyperplasia", "lymph node hamartoma", "angiofollicular mediastinal lymph node hyperplasia", and "angiomatous lymphoid hyperplasia"<sup>1</sup>. It is a rare disease of unknown aetiology that affects lymphatic tissues in diverse locations.<sup>2</sup> Castleman disease is classified into unicentric and multicentric type clinically, based on the number of lymph nodes involvement and histologically further classified into hyaline vascular variant, plasma cell variant, mixed cellular or plasmablastic variant. Unicentric type is usually chronic and asymptomatic whereas multicentric type usually presents with constitutional symptoms. The definitive treatment is based on pathological findings postoperatively. The gold standard management is by complete surgical excision. The main aim of our study is to describe a case of unicentric type of Castleman's disease, its presentation, investigations, and the intraoperative management.

# **Case Presentation**

A 24 year old male presented with a 5x4 cm ovoid, painless, gradually progressive swelling in the left

posterior triangle of the neck since 10 years, Fig. 1 with well - defined borders, which was non tender, firm in consistency and mobile in both horizontal and vertical plane, the plane of the swelling was above the sternocleidomastoid muscle. The skin over the swelling was normal and pinchable with no other associated symptoms. No other swellings palpated elsewhere.

# Fig. 1: Clinical photograph

He was subjected to routine investigations, *ultrasound neck* and *fine needle aspiration cytology* of the swelling which was *inconclusive*, he then underwent excisional biopsy of the swelling and it was sent for histopathological examination.



**Fig. 1**: Clinical photograph – Showing the swelling in the left lateral aspect of the neck

# Management

Under General Anesthesia, a horizontal skin incision around 5 cm was taken over the swelling in the posterior triangle of the neck. Skin, superficial fascia and sternocleidomastoid muscle was separated, capsule over the mass identified and delineated completely all around and the mass was separated from all the adjacent structures and from the base which was anterior border of the trapezius muscle and removed en-mass Fig.2 A and B. He was followed up for one year and he was asymptomatic.



**Fig. 2**: Globular soft tissue mass measuring 5x4.5x3 cm, external surface capsulated, Cut section – homogenous, greyish- white in color.

# B - Cut section

# Histopathology

10x- shows large or normal-sized lymphoid follicles with germinalcentres. It has conserved polarity and paracortical vascular proliferation with some vessels of the paracortex penetrating radially towards the germinal centers, forming lesions that resemble a spiral palette (lollipop appearance). [Fig 3A]

100x-shows magnified image of lymphoid follicles with two or more germinal centers referred to as "twin germinal centers" (twinning). The germinal center has central fibrosis and small lymphocytes with concentric ring formation giving an "onion skin"appearance. [Fig 3B]



**Fig. 3:** a – 10 X – Lollipop appearance 3b- 100 X- Twinning and onion skin appearance

### Discussion

In 1954, Benjamin Castleman first reported, on 2 patients with isolated mediastinal lymph node involvement with lymphoid follicles showing germinal-center involution and increased capillary proliferation with hyperplasia of the endothelium in both follicular and interfollicular regions<sup>1</sup>. The etiopathogenesis is unknown, but based on few studies, evidence points toward faulty immune regulation, resulting in increased B-lymphocyte plasma-cell proliferation in lymphoid and tissue. 1-3 Interleukin 6 (IL-6) plays a pivotal role <sup>2,3</sup>.Mediastinum is the most common site of presentation,<sup>2,4,5</sup> Other sites include the neck, axilla, mesentery, pelvis, pancreas, adrenal gland, and retroperitoneum. <sup>2,4,5</sup> The disease in most cases has aninflammatory background, with predominent levels of interleukin-6 (IL-6) and vascular endothelial growth factor (VEGF).6,7

The lymph nodes with small, prominent, hyalinized follicles with marked interfollicular vascular proliferation are categorized into hyaline vascular type and is further classified into Unicentric type of Castleman's Disease (UCD).<sup>8</sup> Although the incidence of the disease is unknown, but because of its association with Human Immunodeficiency Virus (HIV) and Human Herpes Virus (HHV-8), MCD is also seen to be associated with few malignancies, particularly lymphomas and Kaposi's sarcoma. It is seen that nowadays Multicentric type of disease is resulting in fatality in few patients due to disease progression, disseminated infection, or related malignancies.<sup>10</sup>

A wide variety of treatment modalities have been tried for MCD along with surgery, steroids, specific antibodies, antiviral agents, radiation, cytokine inhibitors, and even chemotherapy. Usually, in these conditions, surgery has no role in the treatment of MCD, although splenectomy has shown symptomatic improvement temporarily in few patients. Unicentric type of disease is the commonest type among the two and is characterized by a localised benign lymphoproliferative disorder in young adults and is not associated with Human Herpes virus-8 infection and usually managed with surgical resection.<sup>10</sup>

# Conclusion

Achieving the diagnosis is quite complicated as there is no proper laboratory investigations or imaging modalities specific for this disease except for histopathologic examination. The gold standard mode of diagnosis is excisional biopsy from an affected lymph node.<sup>8,9</sup> as done in our case. The treatment mainly depends on the type of Castleman disease. Unicentric type has a good outcome to excisional biopsy.<sup>9</sup> Radiotherapy can also be considered with successful outcomes in patients with either residual disease after incomplete resection or unresectable lesions. Chemotherapy is usually not indicated for unicentric type of disease. To conclude prognosis of unicentric Castleman's disease is excellent.

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# An Easier Approach for Nasal Vestibule Recanalization: Endotracheal Tube Tailored Nasal Stents

Nayak Puneeth S<sup>1</sup>, Debnath Paramita<sup>2</sup>, Harugop Anil S<sup>3</sup>, Anand Anshika<sup>4</sup>

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#### Abstract

*Introduction:* Following surgical procedures in the region of nasal vestibule and nasal valves majority of patients have risk of developing vestibular stenosis and face lifelong complications with the external and internal nasal valves; sequels of the repair. The use of endotracheal tube as nasal stent has been mentioned in various literature but it is not validated yet. In the present study we highlighted the use of this endotracheal tube tailored nasal stents for recanalization, prevention of recurrences vestibular stenosis and preserving nasal valves.

Keywords: Nasal vestibular stenosis; Endotracheal tube; Nasal stents.

# Introduction

Majority of patients who undergo surgical procedures involving the nasal vestibule and nasal valves are at risk of developing vestibular stenosis and it affects their day to day life adversely.

Nasal vestibule is the narrowest portion of the nasal cavity hence it is the commonest area to undergo stenosis. The various causes that leads to its stenosis include trauma, burns, tumour, surgery infections. In cases of nasal trauma involving the nasal vestibule and internal and external nasal valves stent placement avoids sequels, adhesions,

valves stent placement avoids sequels, adhesions, © Red Flower Publication Pvt. Ltd.

contractures, synechia vestibular stenosis and fibrosis that occurs surrounding this anatomical structure. The use of endotracheal tube as nasal stent has been mentioned in various literature but it is not validated yet. The advantages of this endotracheal tube tailored nasal stents over the traditional readymade ridged nasal stents, and composite aural grafts are: a) Technical simplicity of use, b) Safety, c) Less morbidity, d) Custom moulded and e) Economical. In the present study we highlighted the use of this endotracheal tube tailored nasal stents for prevention of recurrences vestibular stenosis and preserving nasal valves.

# Case report

A 55 year old male presented to the ENT OPD with complaints of bilateral nasal obstruction for 5 years, insidious in onset, gradually progressive associated with decreased perception of smell which progressed to complete loss of smell perception since last 2 years. Patient also had complaints of mouth breathing. For the same complaint's patient had undergone surgery, following which symptoms recurred within 2 months. On examination, the external appearance of the nose revealed bilateral vestibule stenosis. On anterior rhinoscopy, (Fig.1) synechia were present between inferior turbinate and septum in bilateral nasal cavities. Posterior rhinoscopy and the rest of the ENT examination were found to be normal.

Diagnostic nasal endoscopy revealed synechiae in both nasal cavities.

The patient underwent excision biopsy(Fig.2) which revealed features suggestive of chronic inflammatory disease. The endotracheal tube was cut to appropriate lengths and placed in each naris extending minimally posterior to the bony piriform aperture in bilateral nasal cavity and sutured. (Fig. 3). Patient was started on antibiotics and rifampicin and antihistaminic postoperatively and called for regular follow up, the stents were removed on postoperative day 7. Patient was followed up for a month on weekly basis for removal of crusts and then on monthly intervals. After 3 months of follow-up bilateral vestibules were found to be patent and no signs of re-stenosis present.

# Discussion

The nasal vestibule forms the lower most part of the nasal fossae, limited superiorly by the caudal edge of upper lateral cartilage. The vestibule is lined with skin consisting of squamous epithelium which contains hair and sebaceous glands. It is the narrowest portion of nasal cavity. Nasal vestibular Stenosis is a rare deformity that causes significant aesthetic and functional problems for the patient which in turn affects his day to day life.<sup>1</sup> It affect sthe upper and lateral aspects of the nasal vestibule and due to the lack of support of the alae compared with the more rigidseptal and lower lateral cartilages. Injury to the vestibular lining is the main pathogenesis seen in this condition.<sup>2</sup> The various causes of nasal vestibular stenosis include: infection, trauma, burns, tumours, chemical agents, and surgery to the vestibular line. Traumatic injury to the nose as seen in cases of burns, fractures and lacerations can lead stenosis. Infectious

conditions like chicken pox, tuberculosis, leprosy, syphilis, rhinoscleroma and atrophic rhinitis has also been implicated to result in nasal vestibular stenosis.<sup>3</sup> Surgical procedures in the past including septoplasty and rhinoplasty, unnecessary use of chemical and electrocauterization and traumatic placement of nasal packing for control of epistaxis can also lead to the development of vestibular stenosis. The most common presenting complaint is unilateral or bilateral nasal obstruction. Diagnosis is mainly accomplished based upon physical examination.<sup>4</sup>

Excision of the stenotic tissues followed by splitthickness skin graft remains the easiest technique for recanalization. The skin graft is wrapped around a moulded stent and is left in place for up to 6 months to prevent relapse.<sup>5</sup>

Various techniques have been mentioned in the literature for the repair of nasal vestibular stenosis. A study conducted by Karen et al. showed that excellent results were obtained by using composite aural graft without any nasal stent in cases of vestibular stenosis.6,7 Jablon and Hoffman in their study reported the treatment of case of nasal vestibular stenosis caused by obstetric forceps by successfully using mucosal graft from the hard palate.<sup>2</sup> Reconstructive options available include the modified Z-plasty skin grafts, local myocutaneous flaps, and septal cartilage flaps, mucosal flaps from the palate and composite cartilage grafts. Such techniquere quires general or prolonged monitored anesthesia.8 Another study done by Salvado and Wang concluded that vestibular stenosis can be corrected by using Mitocmycin C as it inhibits fibroblast proliferation and collagen synthesis. The use endoscopic lasers as a treatment option was described by Smith and Roy, the lasers cause lyses of the scar tissue along with application of mitomycin C and nasal stent placement.9

The demerits of above-mentioned procedures include – time consuming, delayed healing, not cost effective, longer healing curve, morbidity, recurrences. The recurrence rates are quite high and are adversely affecting the quality of life. However, recent literature supports management by endoscopic lysis, mitomycin application and stenting.

Endotracheal tubes moulded as nasal stents can be used as effective method to prevent such recurrences following surgeries. The advantages include: a) Sparing the patient from prolonged anesthesia, and b) Major surgery with a better surgical outcome. Additionally, the stent placement is technically simpler and it provides support to the external and internal nasal valves, and also helps achieve maximal anesthetic and functional results in a shorter duration. However, the use of such stents is not validated yet.

In this current case report, we highlighted the use of endotracheal tube tailored stents which is a very simple and inexpensive technique to attain nasal vestibule recanalization and also to prevent its recurrances.

# Figure legends:

**PreOperative Findings** 



Fig. 1: Figure showing synechia present between septum and inferior turbinate in both nasal cavities.



Fig. 2: Figure showing intraoperatively after taking excision biopsy.



Fig. 3 :Figure showing endotracheal tube tailored stents placed in bilateral nasal cavities.

# Conclusion

Thus, we conclude that endotracheal tube tailored nasal stents can be used successfully for avoiding sequels; adhesions, contractures, synechia, vestibular stenosis and fibrosis involving these anatomical structures, also they areeasily available, cost effective and can be custom moulded, patient compliance is better as healing time is considerably less as compared to the traditional methods.

# Conflict of Interest: None

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# **Carcinoma Temporal Bone**

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#### Abstract

Temporal bone carcinoma is a rare entity. Incidence of Sqamaous Cell Carcinoma(SCC) of temporal bone is very less accounting as less than 6 cases per million per year which is 0.3% of all head and neck tumors. Temporal bone carcinoma is frequently misdiagnozed before surgery. Delay in treatment as a result of misdiagnosis has a poor prognosis. Five year disease specific survival is only 19% to 48%<sup>1-3</sup>. In this article we are reporting two cases of temporal bone carcinoma. In these cases High Resolution Computed Tomography (HRCT) and MRI findings revealed soft tissue density in external auditory canal with involvement of middle ear and mastoid area. The disease was in filtrating the surrounding bony substance which was ill defined, there was destruction of ear ossicles with limited erosion of facial canal wall. Both cases were initially misdiagnozed as chronic otitis media with extensive cholesteatoma which were later proved to be SCC temporal bone. Both cases underwent surgical resection with adequate marginsto eliminate the disease. HRCT reports were consistent with the intraoperative findings and post surgery histopathology reports. Patients underwent radiotherapy postoperative and followed up for average 6 months without recurrence.

**Keywords:** Temporal bone carcinoma; Head and neck cancers; Lateral temporal bone resection; Radiotherapy.

#### Introduction

Temporal bone carcinoma is very rare amongst all head and neck cancers.<sup>1</sup> These can be of various types. Squamous cell carcinoma of temporal bone is the most commonly occurring tumor. Other less frequently types are adenocarcinoma, adenocystic carcinoma, mucoepidermoid carcinoma, basal cell carcinoma, ceruminous carcinoma, and rhabdomayosarcoma. Associated risk factors includeprevious radiotherapy treatment and chronicsuppurative otitis media (CSOM) or cholesteatoma within external auditory canal and middle ear.<sup>4-5</sup> Evidence has emerged to suggest human papillomavirus (HPV) can be linked to a subset of this disease group.<sup>6-7</sup> The survival rate at

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5 years is 27% with a high operative mortality rate of 5%[8]. The long-term prognosis of SCC temporal bone malignancies is correlated to the tumor stage, biology of the tumor and the initial treatment.

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Fig. 1a. 1b.

A 47 year old male patient was admitted with chief complaints of bilateral ear discharge since childhood which was insidious in onset, initially mucoid later mucopurulent, non foul smelling, blood stained, which was unresponsive to the medications, with no aggravating factor. It was also associated with decreased hearing since 2 years, earache (recurrent ) and headache since 6 months. There was also a history of mass in right ear since 1 month. It was a post operated case of Right Modified Radical Mastoidectomy done at a local hospital in 2017. Biopsy was taken at the same time on suspicion which later proved the tumor as moderately differentiated SCC temporal bone. On examination tragal cartilage prominence was absent. Pinna was protruding outwards. Postauricular region there was apostaural scar of previous surgery and mastoid region was erythematous, tender, with raised local temperature. In right external auditory canal a pink colored mass was seen, which was occluding whole of the canal, single, soft in consistency and having smooth appearance. On palpation it was non tender, sensitive to touch, probe could be passed all around and mass did not bleed on touch. Tuning fork tests showedrinne test bilateral negative, webers test lateralised to right, absolute bone conduction tests were reduced bilaterally . Facial nerve function was normal bilaterally. On neck examination no palpable lymph nodeswere there and other cranial nerve examination was normal. Audio metric evaluation showed mixed hearing loss of 50 dB in right ear and left ear mixed hearing lossof 40 dB. Preop HRCT and MRI temporal bone showed right external auditory canal soft tissue extending from aditus to antrum with destruction of ear osscicles, thinning of tegmen and bony defect on lateral aspect of temporal bone (Fig: 1a,1b). The choice of surgical approach was based on the extent of tumor

determined via physical examination and imaging studies. Tumor was categorized as University of Pittsburgh (T4N0Mx) Stage 4.

A lateral temporal bone resection with superficial parotidectomy with cavity obliteration Cul de sac closure of the right mastoid was performed under general anesthesia. Revealing a defect in the lateral bony wall of the mastoid process. On further dissection it was seen that mastoid cavity was filled with tumor tissue, which bleed easily . The tumor tissue was confirmed as a squamous cell carcinoma during the subsequent frozensection pathology reports. The tumor had grown upwards destroying the mastoid and tympanic cavity and infiltrating the dura of the middle cranial fossa. It had also destroyed the bonystructure of the sigmoid sinus but had not infiltrated into soft tissues. Destruction of the anterior wall of the external auditory canal and the posterior wall of the tympanic cavity was also seen. But the facial nerve bony canal and labyrinth medial to the tumor were intact. Post operative histopathology reports were suggestive of moderately differentiated SCC temporal bone. Patient was in regular follow up in radiotherapy department after 2 weeks for intensity modulated radiotherapy (IMRT) (66 Gy at 2 Gy per fraction for four weeks).

A 53 year old female patient presented in otorhinolaryngology out patient department with complaints of discharge from left ear for 1.5 years



Case 2 Fig. 2

which was insidious in onset, mucopurulent, scanty, foul smelling, blood stained. There was pain in left infra auricular region for one year which was also associated with decreased hearing.

On otoscopic examination there was anir regular ulcero-proliferative growth involving the left external auditory canal, firm in consistency, which did not bleed on touch (Fig 2). On tuning fork tests, rinne test was negative on left side and positive on right side, webers test was lateralized to left ear and absolute bone conduction tests were reduced on both sides. Bilateral facial nerve examination was normal. On neck examination there was no significant lymph nodes or abnormality seen. On pure tone audiometry there was mixed hearing loss of 50 dB in left ear and 40 dB in right ear. MRI of temporal bone showed soft tissue mass 1cm anterior, 1.5 cm posterior, 1cm superior and 1cm inferior-away from external opening of pinna. Mass engulfing the ossicles but the medial wall of middle ear free from the growth (Fig. 3a,3b). Biopsy was done at local hospital that reported moderately differentiated carcinoma of temporal bone. Tumor was categorized as University of Pittsburgh (T2N0Mx) stage 2.



# Case 2 Fig. 3a, 3b

A left lateral temporal bone resection with superficial parotidectomy was done under general anaesthesia, U shaped incision given from preauricular to postauricular, extending inferiorly to hyoid bone, excising the skin, superficial fascia and temporalis muscle. Circular incision with adequate margins from the tumor given in external auditory canal. Flaps elevated, cortical mastoidectomy done. Epitympanic dissection done upto temporomandibular joint. Extended facial recess approached isolating the facial nerve mastoid segment. Incudo-stapedial joint disarticulated. Tensor tympani tendon was cut. Facial recess extended and complete separation of specimen was done. Superficial parotidectomy done and branches of facial nerve were preserved. Fat and tensor fascia lata harvested from left anterolateral thigh. Eustachian tube opening obliterated using bone wax. Middle ear and mastoid cavity obliterated using fat, tensor fascia lata and temporalis muscle flap. Postoperative HPE documented well differentiated squamous cell carcinoma in the resected tumor, attached salivay gland showed

normal morphology, four intraparotid lymph nodes showed feaures of reactive lymph node hyperplasia. Patient followed up in radiotherapy department for futher radiotherapy (60Gy at 2 Gy per fraction for four weeks) management after 2 weeks.

# Discussion

SCC temporal bone are rare and have low incidence rates. Risk factors for SCC temporal bone are mostly unknown but few of the possible etiologies are chronic suppurative otitis media, cholesteatoma or previous radiotherapy exposure.

Approximately 75 % to 85 % of malignant tumors of temporal bone are secondary to CSOM.<sup>9</sup> As chronic inflammation of malignant tumors of middle ear mucosa may lead to metaplasia. In our experience both cases had history of CSOM.

Symptomatology of SCC temporal bone are mostly atypical. Most commonly presenting complaints in these cases are blood stained otorrhoea, mass in external auditory canal, decreased hearing and otolagia in affected ear. Further as tumor extends to middle ear, inner ear or petrous apex patient may presents with vestibulopathy, cranial nerve neuropathies in which facial nerve involvement is the most commonly encountered nerve.

As the presentation is obcure there are more chances of misdiagnosis hence causing the treatment delay. So, high suspicion of carcinoma temporal bone is necessary and should be kept in mind in all cases of CSOM. For confirmation of diagnosis tissue biopsy is necessary. If first tissue biopsy is negative and clinically external auditory canal mass is of high suspicion it is advisable to repeat biopsy in those cases.

In both of our cases tissue biopsy were found to be moderately differentiated SCC. HRCT and magnetic resonance imaging (MRI) should be done in all cases of diagnozed malignancy of temporal bone for staging the disease and determining the extent of surgical limits. HRCT would help in assessing the tumor invasion into bony structures and MRI would give details of tumor with dural and brain involvement. In one of our case tumor destroyed the mastoid and tympanic cavity infilterating dura of middle cranial fossa and sigmoid sinus. Other case had only limited involvement of external auditory canal and middle ear.

Surgery with postoperative radiotherapy is the main treatment approach in our both

cases of SCC temporal bone.<sup>10</sup> In one case with University of Pittsburgh (T4N0Mx) tumor stage 4, surgical approach was lateral temporal bone resection with superficial parotidectomy with mastoid and tympanic cavity obliteration and Cul de sac closure. In other case with university of Pittsburgh (T2N0Mx) stage 2 lateral temporal bone resection with superficial parotidectomy was done. Post surgery histopatological report confirmed moderately differentiated SCC temporal bone in both cases. Patients were followed up in radiotherapy department after 2 weeks for further management.

# Conclusion

High index of suspicion is required for early diagnosis and treatment of SCC temporal bone which carries a better prognosis. Surgical treatment with lateral temporal bone resection with superficial parotidectomy followed by radiotherapy appears to be the best treatment modality.

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#### Standard journal article

[1] Flink H, Tegelberg Å, Thörn M, Lagerlöf F. Effect of oral iron supplementation on unstimulated salivary flow rate: A randomized, double-blind, placebo-controlled trial. J Oral Pathol Med 2006; 35: 540-7.

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#### **Reference from electronic media**

[9] National Statistics Online – Trends in suicide by method in England and Wales, 1979-2001. www. statistics.gov.uk/downloads/theme\_health/HSQ 20.pdf (accessed Jan 24, 2005): 7-18. Only verified references against the original documents should be cited. Authors are responsible for the accuracy and completeness of their references and for correct text citation. The number of reference should be kept limited to 20 in case of major communications and 10 for short communications.

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