

Bengal Journal of Otolaryngology and Head Neck Surgery

Official Publication of The Association of Otolaryngologists of India, West Bengal

Volume 25 No. 3 - December, 2017

From the desk of the Editor

EDITORIAL BOARD

<u>Editor</u> Dr Saumendra Nath Bandyopadhyay

> *Editorial Secretary* Dr Swagatam Banerjee

<u>Members</u> Dr Ranjan Roychowdhury Dr Swagatam Banerjee

> <u>Treasurer</u> Dr Snehasis Barman

Ex-Officio Members Dr Sumit Kumar Basu (President) Dr Dwaipayan Mukherjee (Hony. Secretary)

<u>Co-ordinators (Journal Operations)</u> Dr Shaoni Sanyal Dr Titas Kar

This journal is indexed in Index Copernicus, DOAJ, OCLC WorldCat, Google Scholar, WHO Hinari, OAIster, BASE, CiteFactor, SIS, OAJI, JournalTOCS, EZB and listed in AcademicKeys and UIUC Repository. It is also registered in the OAI database of conforming repositories.

> p-ISSN: 2395-2393 e-ISSN: 2395-2407 RNI No.: 62551/95



CONTENTS

Main Article	
Evaluation of Results of Cartilage Augmentation in Type III Tympanoplasty Netra Aniruddha Pathak, Vidya Vasant Rokade	119
Does Nasal Obstruction Increase Heart Rate? Venkatesha Belur Keshavamurthy, Munish Kambathatti Shekharappa, Yogeesha Beesanahalli, Nagaraj Maradi, Priya Rani Kori	124
Evaluation of Diagnostic Criteria for AFRS: A Hospital Based Study Krishna V Chaitanya, Lakshmi C Kalavathi	130
Comparison of Various Graft Materials in the Reconstruction of Ossicular Chain in Patients with Chronic Otitis Media: A Prospective Hospital Based Study <i>Niaz Fakhruddin, Chethan Kumar Gangaiah, Gopakumar KP, Geogin</i> <i>George Thottan, Mon Noufal</i>	136
Does Anterior Tunnelling Really Help in Underlay Type I Tympanoplasty? Sohag Kundu, Bhaskar Ghosh, Bijan Kumar Adhikary, Mainak Dutta	142
Role of Polidocanol as Sclerosant in Treatment of Hemangiomas of Head and Neck Region <i>Rupanjita Sangma, Mukul Patar</i>	148

Case Series

Cas

Midline Nasal Dermoid - A Series of Thirteen Cases and Review of Literature Vedula Padmini Saha, Debangshu Ghosh, Santanu Dutta, Somnath Saha, Sumit Kumar Basu	154
Congenital Midline Nasal Mass: Four Cases with Review of Literature Sambhaji Govind Chintale, Sonali Prafull Jatale, Vilas Rambhahu Kirdak, Kaleem Azeem Shaikh	160
e Reports	
Skull Base Langerhans Cell Histiocytosis with Diabetes	166
Insipidus and Panhypopituitarism - A Rare Clinical Entity Anirban Ghosh, Mithun Chaudhury, Abhishek Mukherjee	
	170

Hydatid Cyst of Tongue: A Diagnostic Challenge	170
Neeraj Aggarwal, Tanaya Panja, Khuku Biswas, Titas Kar	
Venous Malformation in the Neck	173
Bijan Kumar Adhikary, Sohag Kundu, Bhaskar Ghosh, 1 Ramanuj Sinha 1	

Correspondence

How We Do It: A DIY Smart Phone based ENT Endoscopy 176 Shyam Duvvi, P Neelapala, A S Duvvi, B Nirmal Kumar

Book Review

Endoscopic Ear Surgery - A New Horizon *by Dr Saurav* 180 Sarkar (Reviewed by Swagatam Banerjee)

Bengal Journal of Otolaryngology and Head Neck Surgery

(Incorporating and directly descended from State Journal of Otolaryngology and Otolaryngology, Calcutta)

Published by

The Association of Otolaryngologists of India, West Bengal CMC House, 91B Chittaranjan Avenue, Kolkata - 700073, West Bengal, India.

Copyright Notice

Copyright © Bengal Journal of Otolaryngology and Head Neck Surgery 2017

The Bengal Journal of Otolaryngology and Head Neck Surgery (hereafter referred to as "BJOHNS") is published by The Association of Otolaryngologists of India, West Bengal (hereafter referred to as "AOIWB") as a triannual scientific journal. All matters published herein (in printed, web or CD format) are copyright of BJOHNS and its publisher AOIWB unless stated otherwise and are governed by the Creative Commons Attribution-NonCommercial 4.0 International Public License. Complete reproduction without alteration of the content, partial or as a whole, is permitted for non-commercial, personal and academic purposes without a prior permission provided such reproduction includes full citation of the article, an acknowledgement of the content, partial or as a whole, is being reproduced on a website, intranet or any other electronic media.

Legal Disclaimer

The views expressed in the articles are entirely of the author(s). The Bengal Journal of Otolaryngology and Head Neck Surgery (hereafter referred to as "BJOHNS"), editorial board or publisher bears no responsibility about authenticity of the articles, references, copyright or otherwise any claim whatsoever. Neither BJOHNS nor its publishers nor anyone else involved in creating, producing or delivering BJOHNS (in printed, web or CD format) or the materials contained therein, assumes any liability or responsibility for the accuracy, completeness, or usefulness of any information provided in BJOHNS (in printed, web or CD format), nor shall they be liable for any direct, indirect, incidental, special, consequential or punitive damages arising out of the use of BJOHNS. All material published in BJOHNS undergoes peer review to ensure fair balance, objectivity, independence, and relevance to educational need. The editors of the material have consulted sources believed to be reliable in their efforts to provide information that is in accord with the standards accepted at the time of publishing. However, in view of the possibility of error by the authors, editors, or publishers of the works contained in BJOHNS, neither BJOHNS, nor its publishers, nor any other party involved in the preparation of material contained in BJOHNS represents or warrants that the information contained herein is in every respect accurate or complete, and they are not responsible for any errors or omissions or for the results obtained from the use of such material. Readers are encouraged to confirm the information contained herein with other sources. Patients and consumers reading articles published in BJOHNS should review the information carefully with their professional healthcare provider. The information is not intended to replace medical advice offered by the physicians. BJOHNS and its publishers make no representations or warranties with respect to any treatment, action, or application of medication or preparation by any person following the information offered or provided within or through BJOHNS. BJOHNS and its publishers will not be liable for any direct, indirect, consequential, special, exemplary, or other damages arising therefrom. The advertisers who purchase advertising space in BJOHNS have no influence on editorial content or presentation. Moreover, the publishing of particular advertisements does not imply endorsement by the BJOHNS or its Editors; they are purely commercial in nature. All legal matters pertaining to BJOHNS (in printed, web or CD format) shall be governed by the laws of India and fall strictly and solely under Kolkata jurisdiction.

<u>Correspondence</u> Dr Saumendra Nath Bandyopadhyay *Editor* Bengal Journal of Otolaryngology and Head Neck Surgery C/O The Association of Otolaryngologists of India, West Bengal CMC House, 91B Chittaranjan Avenue, Kolkata - 700073 Email: <u>editor@bjohns.in</u>

The Association of Otolaryngologists of India West Bengal

OFFICE BEARERS 2017-18

<u>President</u>

Dr Sumit Kumar Basu

President Elect

Dr Debasish Mukherjee

Immediate Past President

Dr Manjula Bhattacharya (Dey)

Vice-Presidents

Dr Subhajit Banerjee Dr Mrinal Kanti Acharya

Honorary Secretary

Dr Dwaipayan Mukherjee

Honorary Treasurer Dr Snehasis Barman

Editor *Dr Saumendra Nath Bandyopadhyay*

Honorary Joint Secretaries

Dr Saumitra Kumar Dr Diptanshu Mukherjee

Executive Committee Members

Dr Utpal Jana Dr Debasish Guha Dr Ajoy Kumar Khaowas Dr Swapan Kumar Ghosh Dr Manoj Mukherjee Dr Tushar Kanti Ghosh Dr Sarmistha Bandyopadhyay Dr Debangshu Ghosh

Editorial Board Members

Dr Ranjan Roychowdhury Dr Swagatam Banerjee

Trustee Board Members

Dr Tarun Palit Dr A M Saha Dr Dulal Kumar Basu Dr Debratan Nandi Dr Haradhan Bora BJOHNS

Bengal Journal of Otolaryngology and Head Neck Surgery

The Official Publication of The Association of Otolaryngologists of India, West Bengal

NOW ONLINE at www.bjohns.in



EASY ONLINE SUBMISSION OF ARTICLES

> DOUBLE-BLIND PEER REVIEW PROCESS

ZERO SUBMISSION AND PUBLISHING CHARGES

PUBLISHED IN PRINT AND ONLINE VERSIONS

INDEXED IN POPULAR INDEXING SERVICES

OPEN ACCESS PROVIDED FOR ALL READERS

FOR AUTHORS

- 1. Visit <u>www.bjohns.in</u> on your internet browser
- (BJOHNS recommends Google Chrome or Mozilla Firefox)
- 2. Click '**Submit Article**' under *Publish with Us* from the right navigation bar
- 3. Registered users can log in to the system with their passwords
- 4. New users must complete a *one-time Registration Process* by selecting 'Not a user? Register with this site'
- 5. Follow the instructions for the easy 5-step submission process
- 6. Track the status of your submission online as it is processed

FOR READERS

- 1. Visit <u>www.bjohns.in</u> on your internet browser (BJOHNS recommends Google Chrome or Mozilla Firefox)
- 2. Prior registration is *not mandatory* to browse articles but is preferred as all users are notified about new issues
- 3. Readers can access *all articles* published in the journal absolutely free of charge
- 4. Select '**Current**' for the current issue and '**Archives**' for previous issues from the top navigation bar
- 5. Use 'Article Tools' from the right navigation bar to cite the article, email the article hyperlink to a colleague or contact the corresponding author by email

From the Desk of the Editor

Menière's disease remains an enigma despite extensive research on the pathogenesis of endolymphatic hydrops. The uncertainty over control of endolymphatic hydrops and consequent uncertainty over control of recurrences of acute episodes have prompted researchers to explore other probable aetiological factors of Menière's disease outside the patho-physiologic hypotheses.

The high incidence of psychological disturbances like anxiety, agoraphobia, depression, avoidance of social interaction and other troubling mood states may be secondarily associated with the uncertainty, discomfort and extreme disability of acute episodes. But Patrice Tran Ba Huy, in an editorial in Otology & Neurotology (2005; 27:1-3), reviewed numerous articles to suggest that psychological factors might disturb the endolymphatic homoeostasis to induce endolymphatic hydrops.

Kessén-Söderman et al. (Laryngoscope 2004; 114:1843-48) demonstrated that emotional rather than physical or mental stress increased the risk of an attack within the next few hours. Ba Huy, while recounting his experience in the research on Menière's disease, had "little doubt that anxiolytic agents, antidepressants and psychotherapy are as efficient as antivertigo, vasodilator or anti-inflammatory drugs."

We still do not know if Menière's disease is caused by psychological factors or if the psychological manifestations are secondary to the disease. "A vicious circle of interaction seems to exist between the somatic organic symptoms of Menière's disease and resultant psychological stress" (Orgi FT. Ann Med Health Sci Res. 2014; 4:3-7).

A patient suspected to be suffering from Menière's disease should be assessed for psychological disturbance including its physical, behavioural and cognitive symptoms to find a relationship between the vestibular symptoms and the psychological status. But it is practically not possible to explore the entire field of human psychopathology and the full range of situations. The best possible option may be to allow the patient to talk freely. "If the patient feels at ease; may reveal marital disharmony, sexual abnormalities, socio-economic problems or other stressful events concomitant with the onset of the symptoms." Talking and listening should be considered to be an integral component of management of Menière's disease in addition to clinical, investigative and conventional therapeutic procedures.

Dr Saumendra Nath Bandyopadhyay Editor, Bengal Journal of Otolaryngology and Head Neck Surgery

This page is intentionally left blank

Evaluation of Results of Cartilage Augmentation in Type III Tympanoplasty

Netra Aniruddha Pathak,¹ Vidya Vasant Rokade¹

ABSTRACT

In conventional Type III tympanoplasty, post operative air-bone gap (ABG) is around 10-60dB. This study aimed to assess the hearing results in patients undergoing canal wall down mastoidectomy with cartilage augmented type III tympanoplasty. <u>Materials and Methods</u>

Patients of 6-50 years of age with the diagnosis of Chronic Otitis Media (Squamous) with conductive or mixed hearing loss, requiring canal wall down mastoidectomy and with intact and mobile stapes suprastructure at surgery who underwent cartilage augmentation were included in the study. Pure tone audiometry (PTA) was performed and evaluated. Post- operative hearing was assessed in terms of average air bone gap (ABG) and size of ABG closure.

<u>Results</u>

The results concluded that mean of pre and post operative air bone gap were 37.5db and 29.7db respectively with net gain of 7.8db. These differences were significant. Also ABG closure was within 30 db in 28 cases (70%).

Discussion

Introduction

The results of this study were compared with other reported series. The mechanical and acoustical aspects of canal wall down surgeries as also the probability of variation in results due to differences in surgical procedures and post-operative fibrosis have been mentioned.

Conclusion

There was significant improvement in postoperative hearing after stapes head augmentation in type III tympanoplasty. <u>Keywords</u>

Tympanoplasty; Mastoid; Cartilage; Audiometry, Pure-Tone; Hearing

hronic otitis media (COM) is a common condition, affecting 0.5–30% of any community.¹ A conservative estimate of the number of people in the world suffering from COM is over 20 milion.² The objective of tympanomastoid surgery for chronic otitis media, in decreasing order of priority are elimination of disease to produce safe and dry ear; alteration of anatomy to prevent recurrent disease, optimization of cleaning and reconstruction of the middle ear to achieve serviceable post-operative hearing.

The goal of tympanoplasty is to restore sound pressure transformation at the oval window by coupling an intact tympanic membrane with a mobile stapes footplate via an intact or reconstructed ossicular chain and to provide sound protection for the round window membrane by a closed, air containing, mucosa lined middle ear.³ The modern era of tympanoplasty was ushered in by Wullstein and Zollner. In classical type III tympanoplasty or myringostapediopexy, disease is removed from tympanomastoid compartment and advancement of the tympanic membrane (TM) or placement of tissue graft is done on top of the stapes head. After this procedure, airbone gap (ABG) range is around 10-60dB. Merchant et al. in laboratory model demonstrated that improved hearing results could be achieved in myringostapediopexy by interposing a thin cartilage disc between the graft and stapes head.⁴

For augmented type III tympanoplasty, either cartilage or sculptured cortical bone can be kept between the intact stapes and the fascial graft. Cartilage disc was hypothesized to improve the "effective" vibrating area of the graft that was coupled to the stapes head.⁵

1 - Department of ENT, Smt Kashibai Navale Medical college, Narhe, Pune

<u>Corresponding author:</u> Dr Netra Aniruddha Pathak email: netra.pathak@gmail.com Main Article

Cartilage also offers the advantage of higher mechanical stability compared with membranous transplants, thus preventing retraction of tympanic membrane in the long run but others argue that it may alter the acoustic transfer characteristics of the graft due to its increasing mass and stiffness of the reconstructed tympanic membrane. The aim of this study was to compare pre and post operative hearing results after cartilage augmentation type III tympanoplasty.

Materials and Methods

A prospective, analytical and longitudinal study was performed after institutional ethical committee approval from October 2012 to April 2014. Patients between the ages of 6 to 50, with an intact and mobile stapes superstructure who underwent canal wall down mastoidectomy, were selected for this study. Pure tone audiometry was performed within seven days prior to the operation. Air and bone conduction thresholds were calculated by taking the averages of 500, 1000, 2000 and 4000 Hz frequencies.

The Air Bone Gap (ABG) was calculated by taking differences between air conduction and bone conduction threshold. air conduction and bone conduction threshold were recorded post-operatively at 6weeks, 6 months and at the end of one year. Postoperative ABG closure was calculated by taking the difference between preoperative and postoperative ABG of the average frequencies of 500, 1000, 2000 and 4000 Hz. For cartilage augmentation, thin disc of conchal cartilage of partial thickness of 4-6 mm in diameter was interposed between the stapes head and temporalis fascia graft. Cartilage disc did not touch the external auditory canal or facial nerve canal. The follow up was performed after a week then after six weeks postoperatively. Results were analyzed in terms of average postoperative ABG and ABG closure. The data analysis was performed with the help of SPSS 11.5 software package. P value was calculated using the independent samples test and P value of 0.05 was taken as significant.

Results

A total of 40 patients were enrolled in this study. Distribution of the patients according to age and gender are shown in Table I. All 40 (100%) patients had adequate follow up.

The preoperative ABG at frequencies at 500Hz, 1000Hz,2000Hz, and 4000Hz were found to be 47.5dB, 38.5dB, 29.7dB and 34.5dB respectively as compared to postoperative ABG of 36.3dB, 30.6dB, 22.4dB and 22.4dB respectively. The difference in four frequency average post-operative ABG was also found to be statistically significant with p value of <0.001. The four **Table I: Age and Gender distribution of patients.**

AGE (YEARS)	NUMBER OF PATIENTS	MALE	FEMALE	AVERAGE AGE (%)
< 10	3	2	1	7.5
11-20	7	4	3	17.5
21-30	18	12	6	45
31-40	8	4	4	20
41-50	4	3	1	10

frequency average preoperative ABG which was 37.5 dB was reduced to 29.9 dB postoperatively with net gain of 7.8 dB with p value <0.001 which is statistically highly significant. (Table II).

The ABG closure with cartilage augmented type III tympanoplasty at different frequencies (500Hz, 1000Hz, 2000Hz, and 4000Hz) was analyzed and their average was plotted and found to be statistically significant.

The ABG closure was again divided into different categories like 0-5dB, 0-10dB, 0-20dB, 0-30dB and 0-40dB. It was noted that 3(7.5%) cases fell within 0-Table II: Pre and post operative AB Gap (n=40)

PARAMETERS	FREQUENCY	MEAN	SD	P VALUE
Preop ABG	500HZ	47.5	13.75	<0.0001
Post op ABG		36.3	10.1	-0.0001
Preop ABG	1000HZ	38.5	15.5	0.0001
Post op ABG		30.6	10.25	0.0001
Preop ABG	2000HZ	29.7	15.15	0.0120
Post op ABG		22,4	9.8	0.0139
Preop ABG	4000HZ	34.5	10.42	<0.0001
Post op ABG		22.4	7.8	~0.0001
Preop ABG	AVERAGE	37.5	11.9	0.0011
Post op ABG	AVERAGE	29.9	7.7	0.0011

5dB , 11(27.5%) cases fell within 0-10 dB,12(30%) cases fell in 0-20dB bin , 13(32.5%) cases fell in 0

-30dB category and only 1case (2.5%) fell in .>30dB bin. (Table III).

Fable III: ABC	G Closures	in	different	categories	(n=40))
-----------------------	------------	----	-----------	------------	--------	---

0-5 DB	6-10 DB	11-20 DB	21-30 DB	>30 DB
3	11	12	13	1
7.5(%)	27.50%	30%	32.50%	2.50%

Discussion

The objectives of this study were to evaluate and analyze post-operative hearing results in terms of average ABG and the ABG closure in patients undergoing CWD mastoidectomy with cartilage augmented type III tympanoplasty. In each case, post-operative airbone gaps were calculated using post-operative air conduction and post-operative bone-conduction thresholds at frequencies 500, 1000, 2000 and 4000 Hz. None of the patient in the whole group had an acute worsening of bone conduction post operatively. During the length of follow up, there were no cases of cartilage extrusion.

Different methods have been used by different authors to report post-operative hearing results in middle ear surgery. Among these ABG closure, post operative ABG presented in 10 dB bins and air conduction threshold gain are commonly reported indicators of tympanoplasty outcome. We had applied average PTA-ABG and the ABG closure for audiological assessment. For calculation of the size of the post-operative PTA-ABG closure ABGs were divided into different bins of 0-5 dB,6-10 dB,11-20 dB, 21-30 dB, and >30 dB.

In our study, while comparing the average post operative air bone gaps at various frequencies the differences were found to be statistically significant (P<0.001). The difference in four frequency average post-operative air bone gap between these two groups

122 Main Article

was also found to be statistically significant.

Our findings are to some extent in agreement with those of Merchant et al who observed a 5 dB improvement at 250Hz, 500Hz and 2000Hz with interposition of thin disc of cartilage between the graft and the stapes head in both of their temporal bone model as well as in their clinical study.⁵ In their clinical study, cartilage augmentation was done after canal wall reconstruction and cavity obliteration. Variation in functional hearing results between the two studies may also have occurred due to this difference in the technique used.

While analyzing the frequency wise post-operative average Merchant et al in their review article state that a canal wall-down mastoidectomy poses several considerations from an acoustical and mechanical perspective when compared to a canal wall-up procedure. Firstly, the canal wall-down procedure i.e., radical or modified radical tympanomastoidectomy results in significant reduction in the size of residual middle ear air space. Secondly, a canal wall down procedure results in the creation of a large air space lateral to the tympanic membrane (TM), i.e., the air space within the mastoid bowl including the external auditory canal.

This mastoid bowl and ear canal air space generates resonances that can influence middle ear sound transmission favorably or unfavorably.⁶ Thirdly, after a canal-wall down procedure, the TM graft comes to lie in a more medial position compared to normal.

The mechanics of a TM graft coupled to the stapes / TORP are likely to be different from normal and also need to be characterized.⁷ The mechanics of such a TM graft and its coupling to the stapes/ TORP are likely to be different from normal and also need to be characterized.⁷ In our study, 28 (70%) cases fell within 30 dB ABG closure. Similar studies with some modifications in the technique have been published in the literature report varying proportions of PTA-ABG closure. Cheang et al in his myringolenticulopexy group (n= 20) achieved an ABG of less than 30 dB in 92 % and ABG of less than 20 dB in 64% of cases.⁸

Moustafa and Khalifa in their myringo-cartilagostapediopexy group (n= 95) achieved an ABG of less than 20 dB in 84%.⁹ Kyrodimos et al in their cartilage shield type III tympanoplasty (n=52) using a 0.8 mm thick cartilage piece with no capitulum for stapes head report that post-operative PTA-ABG of 25dB or less was achieved in 41 (79%) of patients and of 20 dB or less in 54% of patients.¹⁰ However their study included both canal wall up and canal wall down procedures.

Malafronte et al. in cases of both canal down and up procedures used modified folded double cartilage block with shallow acetabulum for stapes capitulum to augment their type III tympanoplasty procedure.¹¹ Another factor leading to failure of tympanoplasty is wide variability in the surgical techniques employed, criteria used to evaluate hearing results and a number of other anatomical, physiological and pathological events that occur post-operatively in the middle ear.

It must be remembered that fibrosis could be due to the underlying middle-ear or upper airway pathology that caused the disease and may not be caused by surgery.

Conclusion

The results concluded that mean pre and post operative air bone gap were 37.5dB and 29.7dB respectively with net gain of 7.8dB. These differences were statistically significant. Also ABG closure was within 30 dB in 28 cases (70%). Thus hearing results after cartilage augmentation in type III tympanoplasty showed improvement in mean post-operative PTA-ABG and also in ABG closure suggesting thin cartilage disc increased the effective vibrating area of tympanic membrane graft.

References

- Sadé J (1982) Introduction. In: Sade J (ed) Cholesteatoma and mastoid surgery. Kugler, Amsterdam, 1-3
- Adhikari P, Sinha BK, Pokhrel NR, Kharel B, Aryal R, Ma J. Prevalence of chronic suppurative otitis media in school children of Kathmandu district. Journal of institute of medicine 2007; 29(3):10-2
- Merchant SN, Rosowski JJ. Auditory Physiology. Surgery of the Ear. 5th Edition. New Delhi: Elsevier India; 2003.
- Merchant SN, McKenna MJ, Rosowski JJ. Current status and future challenges of tympanoplasty. Eur Arch Otorhinolaryngol.1998; 255:221-8
- Merchant SN, McKenna MJ, Mehta RP, et al. Middle ear mechanics of type III tympanoplasty (stapes columella): II clinical studies. Otol Neurotol. 2003; 24(2):186-94
- 6. Goode RL, Friedrichs R, Falk S. Effect on hearing threshold

of surgical modification of the external ear. Ann Otol Rhinol Laryngol.1977; 86:441-51

- Cook JA, Krishnan S, Fagan PA. Hearing results following modified radical versus canal-up mastoidectomy. Ann Otol Rhinol Laryngol. 1996; 105(5):379-83
- Cheang PP, Kim D, Rockley TJ. Myringostapediopexy and myringolenticulopexy in mastoid surgery. J Laryngol Otol. 2008; 17(3):1-5
- 9. Moustafa HM, Khalifa MA. Tympano-cartilagostapediopexy:

A method to improve hearing in open technique tympanoplasty. J Laryngol Otol. 1990; 104:942-4

- Kyrodimos E, Sismanis A, Santos D. Type III cartilage "shield" tympanoplasty: An effective procedure for hearing improvement. Otolaryngol Head Neck Surg. 2007; 136(6):982-5
- Malafronte G,Filosa B,and Mercone F. A new double- cartilage block ossiculoplasty: Long term results. Otol Neurotol. 2531-3008; 29:.3.

Main Article

Does Nasal Obstruction Increase Heart Rate?

Venkatesha Belur Keshavamurthy,¹ Munish Kambathatti Shekharappa,¹ Yogeesha Beesanahalli,¹ Nagaraj Maradi,¹ Priya Rani Kori¹

ABSTRACT

Introduction

Nasal obstruction is implicated in the etiopathogenesis of Obstructive Sleep Apnea (OSA). OSA is associated with mean heart rate (HR) variations in wakefulness and in sleep. Early intervention has proven to reduce cardiovascular morbidity in OSA patients. In spite of various confounding factors HR measurement has been utilised as an independent predictor of mortality. The influence of severity of nasal obstruction on HR has not been studied in the literature. This study aims to clarify the influence of severity of nasal obstruction on HR.

Materials and Methods

We examined 55 patients aged less than 50 years with no previous cardiac complaints, who underwent overnight oxygen saturation and HR monitoring. The patients were divided into Mild, Moderate and Severe Nasal Obstruction group depending on NOSE scale grading.

<u>Results</u>

There was no statistically significant difference in the Mean HR, Min HR, Max HR, and Max-Min HR in mild, moderate or severe nasal obstruction groups.

Discussion

The role of nasal obstruction in Obstructive Sleep Apnoea and the importance of HR as predictor of cardiovascular morbidity have been discussed. The studies on the heart rate in nasal obstruction and OSA were reviewed.

Conclusion

Nasal obstruction does not influence the heart rate.

Keywords: Heart Rate, Obstructive Sleep Apnea, Nasal Obstruction

Asal obstruction is implicated in the etiopathogenesis of Obstructive sleep apnea (OSA). OSA is associated with mean heart rate (HR) variations in wakefulness and in sleep.¹ Nasal obstruction is caused by various pathologies like deviated nasal septum, hypertrophied turbinates, vasomotor phenomenon, sinonasal polyps, sinusitis, allergy and chronic granulomatous nasal conditions.

Nasal obstruction compels mouth breathing and during sleep, due to greater negative pressure which develops in the oropharynx area, pliable structures like soft palate, uvula, vibrate leading to snoring. The Starling resistor

 1 - S.S. Institute of Medical Sciences & Research Centre, Davangere, Karnataka
 Corresponding author: Dr Venkatesha Belur Keshavamurthy email: bk.venkatesha@gmail.com model of upper airway collapsibility describes how the upper airway behaves like a Starling resistor, in that obstruction at the inlet (i.e. the nasal airway) produces collapsing forces, that manifest downstream in the collapsible segment, the pharynx.²

Nasal obstruction results in nocturnal mouth breathing, sleep fragmentation³ and snoring, leading to sequele of disturbed sleep^{4,5} like excessive daytime sleepiness. Definite role of nasal obstruction in complex etiopathogenesis of Obstructive sleep apnea (OSA) has been proven in the literature.^{4,6}

Association between snoring / OSA and adverse cardiovascular event is well documented. Time-domain analysis of heart rate (HR) variability, used as the only criterion, could represent an efficient tool in OSAS diagnosis with a sensitivity of 90%.⁷ Heart rate variability is an important parameter measured for the assessment of

autonomic nervous system activity indirectly measuring the adverse cardiovascular outcome. Furthermore elevated Mean HR was reported to be a risk factor for cardiovascular mortality in normotensive patients with end stage renal disease.⁸ HR represents one of the most important independent predictors of cardiovascular, noncardiovascular, and overall mortality.⁹

HR can easily be monitored in various clinical setting. Few literature reviews are available implicating experimentally induced nasal obstruction as an independent factor involved in increase of HR.¹⁰ But role of spontaneous nasal obstruction as an independent factor in increasing the HR has not been proved in the literature. This study is intended to analyze the relationship of severity of nasal obstruction to mean HR, thus indirectly predicting adverse cardiac as well as overall mortality.

Materials and Methods

A cross-sectional study involving 55 patients, who underwent septoplasty and or turbinate reduction between November 2014 to April 2016 for various indications like nasal obstruction, Sluders neuralgia, epistaxis, and as an approach to DCR and FESS. Subjects with known cardiovascular diseases, on medications with beta blockers, hyperthyroidism and OSA (Epworth Sleepiness Scale score >8) were excluded from the study. Subjects were given NOSE (Nasal obstruction and septoplasty evaluation) scale¹¹ and depending on the subjective grading of the severity of the obstruction, they were divided into 4 groups: No obstruction group, mild obstruction group, moderate obstruction group and severe obstruction group.

On the preoperative day, overnight monitoring heart rate was done for minimum of 6 hours. Clinical parameters like Mean Heart Rate (MHR) Minimum Heart Rate (Min HR) Maximum Heart rate (Max HR) and Difference of Maximum and Minimum Heart rate (Max-Min HR) were recorded using 24 hour pulseoximetry.

Statistical tests: Continuous data were expressed as mean and SD. Spearman's Correlation coefficient test was used to determine the relationship between degree of nasal obstruction and Mean HR, Max HR, Min HR, Max-Min HR, Intergroup variability and significance was determined by one way ANOVA test. Linear regression analysis was performed to determine the relation of severity of nasal obstruction to HR. A p value of <0.05 was considered statistically significant.

Results

Clinical characteristics of the patients are shown as in Table I. Group IV had higher age group distribution compared to the similar age distribution in other groups.

PATIENT	"GROUP I (N=10)"	"GROUP II (N=25)"	"GROUP III (N=11)"	"GROUP IV (N=09)"	P VALUE
Age	"16-28yrs Mean 20.2 yrs"	"16-45 yrs Mean 28.2 yrs"	"16-44 yrs Mean 27.64 yrs"	"24-50yrs Mean=36.78 yrs"	"F=5.811 p=0.0017"
Male Gender	9 (90%)	17(68%)	10(90.9%)	6(66.67%)	"F=1.247 p=0.30"
Septal Deviation with concomitant Turbinate pathology	1(10%)	3(12%)	3(27.3%)	2(22.2%)	"F=0.5838 p=0.63"

Table I: Patient Characteristics

126 Main Article

All groups had predominant Male gender population and septal deviation was the predominant nasal pathology in all groups.

Overnight Heart Rate measurement revealed Mean HR of 74.6, 75.24, 74.91 and 71.78, Max HR was 105.6, 103.28, 106.54 and 102.33; Max-Min HR was 44.2, 42.64, 45.54, 43.89 respectively in Group I-IV as shown in Table II.

The association between the two variables would not be considered statistically significant and are negatively weakly correlated. For HR fluctuation (Max-Min HR) the value of correlation coefficient value was 0.02839 and the two-tailed value of P was 0.837 indicating very weak positive relationship for Max-Min HR. These results clearly indicate there is no statistically significant relationship between the severity of Nasal Obstruction

Tuble III Overlinght II					
HEART RATE	"GROUP I	"GROUP II	"GROUP III	"GROUP IV	P VALUE
PARAMETER	(N=10)"	(N=25)"	(N=11)"	(N=09)"	
Mean HR	"74.6	"75.24	"74.91	"71.78	"F=1.759
	(70-78)"	(67-80)"	(70-80)"	(57-80)"	p=0.17"
Max HR	"105.6	"103.28	"106.54	"102.33	"F=1.253
	(100-110)"	(96-114)"	(90-115)"	(96-112)"	p=0.30"
Min HR	"61.6	"60.64	"61	"57.33	"F=2.831
	(58-66)"	(56-66)"	(56-66)"	(49-68)"	p=0.047"
Max-Min HR	"44.2	"42.64	"45.54	"43.89	"F=0.6906
	(40-50)"	(36-54)"	(34-61)"	(36-53)"	p=0.56"

Table II: Overnight Heart Rate monitoring findings

Spearman's correlation coefficient value was -0.15 and the two tailed p value was 0.27583, which indicates there is very weak negative monotonic relationship between Mean HR and severity of the nasal obstruction and as p value is more than 0.05, results were statistically insignificant.

Similarly, Spearman's correlation test was done to find the relationship between Max HR, Min HR, Max-Min HR and Min HR. For Max HR the correlation coefficient value was -0.15316 and the two-tailed value of P were 0.26427 indicating very weak negative relationship without statistical significance. For Min HR, The value of correlation coefficient was -0.25683 and the two-tailed value of P is 0.05838. and HR parameters.

A simple linear regression was calculated to predict HR based on severity of nasal obstruction. A Non significant regression equation was found, R2=1.901752424.10-1, Y=3.138205319.10-2 , X=2.246768581, considering participants' predicted increased HR was equal to or more than 80.8 When severity of nasal obstruction was measured as no obstruction, mild, moderate, severe nasal obstruction, HR did not increase for each degree of nasal obstruction. A scatter plot diagram shows the linear regression as shown in Fig.1. An Analysis of Variance showed that the effect of degree of nasal obstruction was not significant F=1.756, p=0.167.

Discussion

Role of Nasal obstruction in OSA: Nasal obstruction is caused by various pathologies like deviated nasal septum, hypertrophied turbinates, vasomotor phenomenon, sinonasal polyps, sinusitis, allergy and chronic granulomatous nasal conditions. Significant role of nasal obstruction in the etiopathogenesis of OSA is debatable. Mc Nicholas describes the starling resistor model for the development of OSA. Nose being the inlet of the respiratory tube, obstruction of the inlet snoring in a high percentage of cases. Numerous studies examining the effects of experimentally induced nasal obstruction (ie nasal packing) on OSA have indicated a positive association. In predisposed individuals, treatment of allergy has proven a vital role in reducing obstructive sleep apnea.⁶

Even though NO plays a minor role in the complex pathogenesis of OSA, but has relevance in the aetiology of snoring.¹² The switchover from nasal breathing to mouth breathing is crucial in determining the pathophysiology of OSA.² Nasal and oral resistance,



Fig. 1. Showing scattered plot depicting the relationship between increased HR and nasal obstruction.

leading to the development of negative pressure in the oropharynx leading to collapse of compliant pharyngeal structures.² It was observed that fixed nasal obstruction as seen in deviated nasal septum, polyps, hypertrophied turbinates, contributes less when compared to variable nasal obstruction like reversible nasal congestion and allergy.² Since breathing through the nose appears to be the preferred route during sleep, nasal obstruction frequently leads to nocturnal mouth breathing, snoring, and ultimately to OSA.

It has also been demonstrated that nasal stimulation or obstruction determines an increase in the lung airways resistance. In normal subjects, nasal obstruction partial or total, due to various causes (septal deviation, turbinate hypertrophy and other nasal abnormalities) provokes tone of the muscles, architecture of the upper airway, posture of the individual, age of the individual, obesity and medications influences OSA.

Clearing nasal obstruction relieves snoring in few, but some continue to snore indicating multifactorial nature of OSA.^{2,12}

Use of external nasal devices,¹³ surgery for reducing the nasal obstruction like turbinoplasties, correction of nasal septum, removing the polyps have successfully reduced nasal resistance and severity of snoring, but failed to improve AHI, RDI, Arousal Index, total sleep time, sleep architecture, sleep fragmentation, including snoring loudness.³

Furthermore, surgical intervention for OSA was

clearly successful in children and adults where a long segment of upper airway is corrected like removing tonsils, reducing the tongue base, osteotomies and combination procedures, than short segment surgery like isolated correction of soft palate UPPP, septoplasty, isolated turbinate correction which has met with more failure rates.⁴

HR as predictor of Cardiovascular Morbidity: It's a general agreement that heart rate represents one of the most important independent predictor of cardiovascular, non-cardiovascular and overall mortality.⁹ Mean HR was reported to be a risk factor for cardiovascular mortality.¹ In a study by Fulvia Seccareccia, involving 2533 men aged between 40-69 with other variable, calculated the risk of mortality against increase in the HR.⁹

It was found when other risk factors being equal, death risks increase about 50% for each 20- beatper-minute increment, and relative risks between extreme HR levels are more than 2-fold.⁹ Heart rate exhibits great inter individual variability: is influenced by stress, anxiety,⁹ physical activity, psychological makeup, hormonal status, gender etc. It also shows intra individual variation, influenced by circadian rhythm, stages of sleep. Resting heart rate i.e. HR measured during sleep is of more clinical importance than heart rate measured during wakefulness.

Factors like nature of the surgery, less effective pain control, sleep deprivation following surgery, type of nasal packing influences the basal heart rate measurements in the post operative period. Hence preoperative measurement of HR gives near physiological values than post operative measurement. HR monitoring can be easily done with the use of pulse oximetry in resource poor setting.

The differences between healthy and OSA subjects were much smaller between sleep stages indicating similar mechanism for heart rate control in both healthy and OSA patients.¹⁴ The severity of OSA was independently associated with elevated mean HRs during 24 hours, wakefulness, and sleep in patients with OSA and six month treatment with nCPAP reduced the values in those patients. The prognostic significance of elevated mean HRs during 24 hours, wakefulness and sleep is necessary to be clarified in patients with OSA.

Le Heuzey et al described greater visual enlargement of the RR trend and a significantly higher difference between minimal and maximal HR during the night in OSAS patients than in control subjects.⁷ In our current study, higher difference between minimal and maximal HR during night monitoring did not signify any correlation with severity of nasal obstruction.

In a pilot study by John Hart, it was observed that a higher Resting Pulse Rate (RPR) at baseline was associated with increased RPR change, whereas a lower baseline RPR was associated with a stable or reduced RPR change.¹⁵ In a Study by Michael S. Benninger, on athletes to detect the effect of nasal obstruction, on workload, respiratory rate, Mean Heart rate, oxygen saturation, systolic BP, did not find any statistically significant difference in these parameters. Our results matches with this study, with reference to the Mean heart rate.¹⁰

Conclusion

Mean heart rate, Maximum Heart Rate and difference between Max and min HR is not affected by severity of nasal obstruction.

Limitations of the study: Comparison between two objective values (like rhinomanometry, flow, resistance values) may provide a better correlation than assessing subjective scores (NOSE score) with HR. Measurement of HR with reference to various stages of sleep would indicate the effect of nasal obstruction on autonomic control mechanisms of CVS. Concomitant measurement of ECG, with various time domain parameters of HR variability would have given more predictability in terms of adverse cardiovascular events.

References

- Kawano Y, Tamura A, Wantabe T, Kadota J, Influence of the severity of obstructive sleep apnea on heart rate Journal of Cardiology:2010: 56; 27-34
- 2. Mc Nicholas, The Nose and OSA: Variable nasal obstruction may be more important in pathophysiology than fixed obstruction. Eur Respir J. 2008; 32: 3-8
- Migueisa D.P, Thuler L.C.S, Lemesc NA, Moreira C.S.S, Joffily L, Araujo-Melo MH. Systematic review: the influence of nasal obstruction on sleep apnea: Braz J Otorhinolaryngol.

Does Nasal Obstruction Increase Heart Rate?

2016; 82(2):223-31.

- Scharf MB, Cohen AP. Diagnostic and treatment implications of nasal obstruction in snoring and obstructive sleep apnea. Ann Allergy Asthma Immunol. 1998 Oct; 81(4):279-87.
- Prota R, Dalmasso F. Snoring: analysis, measurement, clinical implications and application. Eur Respir J. 1996, 9: 146-59.
- 6. Pevernagie DA, De Meyer MM, Claeys S. Sleep, breathing and the nose: Sleep Medicine Reviews 2005: 9: 437-51.
- Roche R, Gaspoz JM, Court-Fortune I, Minini P, Pichot V, Duverney D, et al. Screening of Obstructive Sleep Apnea Syndrome by Heart Rate Variability Analysis: Circulation 1999: 100:1411-5.
- Cice G, Di Benedetto A, Isa S, De Gregorio P, Marcelli D, Gatti E, Calabrò R.. Heart rate as independent prognostic factor for mortality in normotensive hemodialysed patients. J Nephrol. 2008; 21(5):704-12.
- Seccareccia F, Pannozzo F, Dima F, Minoprio A, Mendittlo A, Noce CL, et al. Heart Rate as a Predictor of Mortality: The MATISS Project: American Journal of Public Health 2001: 91(8): 1258-63.

- Benninger MS, Sarpa JR, Ansari Tariq, Ward J. Nasal patency, aerobic capacity, and athletic performance. Otolaryngology-Head and Neck Surgery 1992:107(2):101-4
- Stewart MG, Witsell DL, Smith TL, Weaver EM, Yueh B, Hannley MT. Development and validation of the Nasal Obstruction Symptom Evaluation (NOSE) scale. Otolaryngol Head Neck Surg. 2004;130(2):157-63
- Kohler M, Bloch K.E, Stradling JR. The role of nose in the pathogenesis of obstructive sleep apnea and snoring. Eur Respir J. 2007; 30: 1208-15
- 13. Pevernagie, D, Hamans E, Cauwenberge V, Pauwels R. External nasal dilation reduces snoring in chronic rhinitis patients: a randomized controlled trial. Eur Respir J. 2000; 15: 996-1000
- Penzel T, Kantelhardt JW, Lo CC, Voigt K, Vogelmeier C. Dynamics of Heart Rate and Sleep Stages in Normals and Patients with Sleep Apnea. Neuropsychopharmacology 2003; 28;48-53
- 15. Hart J, Testing an association between baseline resting pulse rate averages and short-term changes in resting pulse rates: A pilot study J Can Chiropr Assoc. 2015; 59(2):165-72.

Evaluation of Diagnostic Criteria for AFRS: A Hospital Based Study

Krishna V Chaitanya,¹ Lakshmi C Kalavathi²

ABSTRACT

Introduction

Bent and Kuhn criteria are the most commonly accepted diagnostic criteria for diagnosis of Allergic Fungal Rhinosinusitis. Other diagnostic criteria for the diagnosis of Allergic Fungal Rhinosinusitis include unilateral nasal disease, Charcot Leyden crystals, bony erosions which form the minor criteria in the diagnosis of Allergic Fungal Rhinosinusitis. Clinical and Laboratory features in Allergic fungal rhinosinusitis are variable. These variations in the diagnostic criteria in the diagnosis of Allergic Fungal Rhinosinusitis have been analysed in the present study.

Materials and Methods

Prospective study was performed on group of 46 patients of Allergic fungal Rhinosinusitis presenting in the Otorhinolaryngology OPD with symptoms of Allergic fungal rhinosinusitis as diagnosed and persisting for more than 3 months during September 2009 to August 2010.

<u>Results</u>

Absolute eosinophil count was elevated in 80.43%, total serum IgE elevated in 69.67%, skin prick test was positive in 63.05% of patients. CT scan revealed that most common paranasal sinus involved is ethmoid sinus as seen in 73.91% cases. Sphenoid sinus was least involved as seen in 17.40% cases. More than one paranasal sinus were involved in 65.21% of the cases., Complete opacification of all sinuses with calcified deposits were seen in 4.76% cases. Mucosal thickening was seen bilaterally in 73.91% of the patients and bony erosion was noted in 6.52% of patients. Histopathology of nasal smears revealed Eosinophilia in 80.43% of patients. Inflammatory Charcot Leyden crystals were found in 15.21% of the patients45.65% showed goblet cell hyperplasia. Other types of inflammatory cells were seen in 56.52% of the study population and 23.91% patients showed positive fungal hyphae.

Discussion

The significance of absolute eosinophil count, skin prick test, histopathology, CT Scan features and nasal smear cytology have been discussed along with review of literature.

<u>Conclusion</u>

Although management of Allergic Fungal Rhinosinusitis has advanced tremendously with better understanding of underlying pathogenesis, diagnostic strategies are still far from clear and are still emerging. Lot of research work has to be carried out regarding relevant diagnostic criteria for the disease.

<u>Keywords:</u>

Rhinitis, Allergic; Sinusitis; Eosinophils; Immunoglobulin E; Skin Tests, Mucins

llergic fungal rhinosinusitis is observed to be a non-invasive disease involving the nose and paranasal sinuses and it is believed to be due to hyper-reactivity to fungal antigens. Patients of allergic

1 - Department of ENT, Narayana Medical College and Hospital, Chinthareddypalem, Nellore, Andhra Pradesh 2 - Department of Pathology, ACSR Govt. Medical College, Nellore

Corresponding author: Dr Krishna V Chaitanya

email: drvkc17@gmail.com

fungal rhinosinusitis suffer from allergic rhinitis, and most of these patients have increased blood levels of immunoglobulin E (IgE).¹ The exact pathophysiology of the disease spectrum is not clearly understood. However it is believed that fungal allergens elicit immunoglobulin E mediated allergic and possibly type III immune complex mediated mucosal inflammation in the absence of invasion in an immunocompetent individual.²

Most commonly accepted diagnostic criteria for diagnosis of Allergic Fungal Rhinosinusitis are Type I hypersensitivity, nasal polyposis, eosinophilic mucin,

CT findings, and fungal staining which form the basis of Bent and Kuhn criteria. Other diagnostic criteria for the diagnosis of Allergic Fungal Rhinosinusitis include unilateral nasal disease, Charcot Leyden crystals, bony erosions which form the minor criteria in the diagnosis of Allergic Fungal Rhinosinusitis.³

Investigations for diagnosis of Allergic Fungal Rhinosinusitis in India are grouped as in-vivo and in-vitro diagnostic techniques. In-vivo methods of investigation of the disease process include skin-prick test, intradermal test, bronchial challenge test and nasal challenge test. In-vitro investigations include radioallergosorbent test (RAST), enzyme linked immunosorbent assay (ELISA), microarray and immunocap are applied to arrive at diagnosis.⁴

Radioallergosorbent test (RAST) is an in-vitro test for diagnosing IgE in serum for environmental allergens. ELISA gives the amount of IgE circulating in the blood during sensitization by an allergen.⁵ It was observed by the research work performed in this area that the in-vitro tests are more sensitive than in-vivo tests. Though RAST or ELISA is preferred to be more precise investigations in diagnosis of allergic diseases, they are expensive and hence are not commonly preferred in the diagnosis of Allergic Fungal Rhinosinusitis. Also the role of these tests are believed to be limited in India for diagnosis of allergic diseases as purified proteins and antibodies for these tests are not available for characterization based on major allergen content in India.⁶

The investigative criteria for diagnosis of Allergic Fungal Rhinosinusitis have undergone numerous revisions.⁷ Apart from haematological investigations used for detecting underlying pathology in allergic fungal rhinosinusitis, most authors agree on non-contrast CT scan in rhinosinusitis demonstrating hyperattenuating allergic mucin within the lumen of the paranasal sinus and the presence of non-invasive fungal elements within that mucin, detectable on staining or culture as diagnostic criteria in allergic fungal rhinosinusitis.⁸

Although the management of allergic fungal rhinosinusitis has advanced tremendously with better understanding of the underlying pathogenesis, the diagnostic strategies are still far from clear and are still emerging. The purpose of this study is to identify the sufficiency of the present investigations in the diagnosis of Allergic Fungal Rhinosinusitis by observing the values of absolute eosinophil count, total serum IgE and skin prick test in patients of allergic fungal rhinosinusitis while correlating the clinical diagnosis of allergic fungal rhinosinusitis with histological confirmation by nasal smear cytology.

Materials and Methods

A prospective study was performed on a study group of 46 patients of Allergic Rhinosinusitis presenting in OPD, Department of Otorhinolaryngology with symptoms of the disease persisting for more than 3 months during September 2009 to August 2010 to observe the values of absolute eosinophil count, total serum IgE and skin prick test in patients of allergic fungal rhinosinusitis and also to study the variation in clinical diagnosis of Allergic Fungal Rhinosinusitis with histological diagnosis and nasal smear cytology.

Patients in the age group of 20 to 60 years with no sex predilection with a clinical history of Allergic Rhinosinusitis with symptoms of nose block, headache, nasal itching, sneezing, rhinorrhoea for more than 3 months who were not responsive to medications were included in the study group.

Patients having symptoms of Allergic Rhinosinusitis for less than 3 months were excluded from the study, as also the patients having bronchial asthma, atopic dermatitis and immunosuppressive diseases. Allergic Fungal Rhinosinusitis is mainly believed to be due to Type I hypersensitivity and Type III reaction and formation of immune complexes in the disease spectrum is doubtful thus excluding the involvement of systemic diseases from the study. These minor criteria of Bent and Kuhn in diagnosis of Allergic fungal Rhinosinusitis always need not be present in all the patients with allergic fungal rhinosinusitis.

After obtaining a written informed consent, the patients were evaluated with a clinical questionnaire for clinical symptoms of headache, nasal obstruction, nasal itching, sneezes and rhinorrhoea. Diagnostic nasal endoscopy was performed in all the patients to observe for anatomical abnormalities, shiny allergic mucin and Main Article

any space occupying sino-nasal polyposis in the nasal cavity.

The patients were investigated with absolute eosinophil count, total serum IgE, and skin prick test. CT scan of paranasal sinuses Coronal cuts 2 mm thickness with axial reconstruction was performed in all these patients. Nasal lavages were collected from the patients to study nasal smear cytology and histopathological examination.

These specimens were examined using light microscopy after digestion with 10% potassium hydroxide (KOH). Haematoxylin and Eosin stains were used for all sections in histopathological examination. Other special stains were used as and where required. Histopathological findings of allergic mucin consisting of degenerating eosinophils, cellular debris, Charcot Leyden crystals inflammation, and presence of fungal hyphae were recorded. Samples were cultured onto Sabouraud's dextrose agar and incubated for 4 weeks as and where required.

Results

In the present study laboratory investigations of absolute eosinophil count revealed elevated levels in 80.43% of patients. Total serum IgE was elevated in 69.67% of patients. Skin prick test performed with CREDISOL® reagents on the forearm of the patients for allergens based on local climatic conditions with positive control of histamine and negative control of normal saline was positive in 63.05% of patients.

CT scan findings in the present study revealed that the most common paranasal sinus involved being the ethmoid sinus in 73.91% of patients while sphenoid sinus was the least commonly involved sinus in 17.40% of patients. Involvement of more than one paranasal sinus was the common finding in this study and it was observed in 65.21% of patients.

Complete opacification of sinuses with calcified deposits was observed in 4.76% patients in the present study. Mucosal thickening was observed bilaterally in 73.91% of patients. Bony erosion was observed in 6.52% of patients. These results were tabulated in table I.

Histopathology of the specimens of nasal smears revealed that Eosinophilia was the most common finding in 80.43% of patients. Predominance of eosinophils is usually observed in patients of Allergic fungal

Table I: CT scan findings in Allergic Fungal Rhinosinusitis (N=46)

	CT SCAN FINDINGS	NO OF CASES N=46	%
	Ethmoid	34	73.91
	Frontal	24	52.17
Sinus involved	Maxillary	32	69.57
	Sphenoid	8	17.4
	Multiple sinus involvement	30	65.21
	Partial	18	39.13
Sinus opacification	Complete	28	60.87
· · · · · · · ·	Complete with calcification	2	4.76
Mucosal	Unilateral	12	26.08
thickening	Bilateral	34	73.91
Bony erosion	Present	3	6.52

rhinosinusitis.⁹ For quantification of Eosinophils cells in the nasal smears we propose Grading of AFRS based on number of Eosinophils observed per high power field.

NO. OF EOSINOPHILS	GRADING OF EOSINOPHILIA
1-10 per HPF	1+
10-25 per HPF	2+
25-50 per HPF	3+
>50 per HPF	4+

Table II: Grading of Eosinophilia

In the present study the patients having elevated levels of Eosinophils were observed in the 3+, 4+ grading of eosinophils and hence were classified as Eosinophil rich. The method of quantification of grading of Eosinophils is mentioned below in Table II.

Inflammatory Charcot Leyden crystals were observed in 15.21% of patients, Goblet cell hyperplasia was observed in 45.65% of patients. Other types of inflammatory cells were observed in 56.52% of patients. It was observed that only 11 (23.91%) patients of the present study showed positive fungal hyphae microscopically. These results were tabulated in Table III.

In the present study when the investigations of total serum IgE, AEC, Skin prick test was compared with nasal smear cytology there was a variation in diagnosis in 4.34% of the patients. These results were documented in Table IV.

Discussion

In this study, clinically 46 cases were diagnosed to have Allergic Fungal Rhinosinusitis based on the clinical findings. The mean age of our cases was 29.4 years with a range of 20–60 years. A slightly higher incidence of the disease was observed in the females with the male: female ratio of 1: 1.19. The findings of our study were similar to the study by Kaur et al.¹⁰

, ,	,	
HISTOPATHOLOGY	NO OF CASES N=46	%
Allergic mucin	36	78.26
Charcot laden crystals	7	15.21
Fungal hyphae	11	23.91
Goblet cell Hyperplasia	21	45.65
Eosinophilia	37	80.43
Other cells increase	26	56.52

Table III: Histopathology of nasal smear in Allergic Fungal Rhinosinusitis (N=46)

Table IV: Diagnosis of Allergic Fungal Sinusitis (N=46)

	NO OF PATIENTS N=46	%
Clinical diagnosis of Allergic Fungal Sinusitis	24	52.17
Histopathological diagnosis of Allergic Fungal Sinusitis	22	47.83
Change in diagnosis	2	4.34

In the patients diagnosed as suffering with allergic fungal rhinosinusitis clinically, laboratory investigations of absolute eosinophil count revealed elevated in 80.43% of patients. Total serum IgE was elevated in 69.67% of patients. Eosinophilic count higher than 500 cells per ml was considered as serum eosinophilia while IgE levels were considered to be raised when the counts were $>100U/ml.^{11}$ Eosinophilia is observed to be one of the minor criteria useful for diagnosis of Allergic fungal

rhinosinusitis.12

Skin prick test was positive in 63.05% of patients. Skin test reactivity depends on at least three separate factors: (1) an intact immune system; (2) the presence of IgE sensitized mast cells that release mediators when exposed to antigen; (3) and skin that can respond to histamine with the development of inflammatory response including erythema and induration.¹³ However a negative skin prick test in patients of allergic fungal rhinosinusitis is presumed as a probability of vasomotor aetiology

The presence of allergic mucin in histopathology specimens forms an important diagnostic criterion in addition to the demonstration of fungal elements in diagnosis of Allergic fungal rhinosinusitis.¹⁴ In the present study Allergic mucin was observed in 78.26% of patients and cheesy debris was observed in 48.70% in the nostrils of the patients. The lower percentage of allergic mucin in patients of allergic fungal rhinosinusitis can be attributed to the usage of indiscriminate use of corticosteroids which might have probably disturbed the mucin blanket.

CT scan findings form one of the important diagnostic criteria in the diagnosis of allergic fungal rhinosinusitis. The usual radiological findings of the disease process include pan sinus disease with expansion and smooth thinning of the affected sinuses with thin peripheral enhancement with no enhancement noted in the central sinus contents. There is usually involvement of multiple sinuses.¹⁵ In the present day scenario there is a need for a specific radiological classification for Allergic Fungal Rhinosinusitis where there is a scope of lot of research in this area.

Nasal smears cytology revealed that Eosinophilia was the most common finding in 80.43% of patients. Inflammatory Charcot Leyden crystals were observed in 15.21% of patients, Goblet cell hyperplasia was observed in 45.65% of patients. Other types of inflammatory cells were observed in 56.52% of patients. These other types of cells observed were attributed to neutrophil predominance which may be due to an acute bacterial Rhinitis. It was observed that 11 (23.91%) patients of the present study showed positive fungal hyphae microscopically.

It can be considered to evaluate Allergic Fungal Rhinosinusitis based on Antigen specific IgE, microscopic evaluation of allergic mucin obtained for evaluation of degranulation products and proteins obtained after degranulation and serological testing of precipitating antibodies.¹⁶ However in developing countries like India owing to the cost effectiveness of the above investigations these are not routinely available for the diagnosis of the disease spectrum of Allergic Fungal Rhinosisnusitis.

Conclusion

In the present day clinical scenario despite the newly emerging clinical investigations and efforts, allergic fungal rhinosinusitis remained an entity which is difficult to diagnose. With the description of newer categories like eosinophilic fungal rhinosinusitis and eosinophilic mucin rhinosinusitis, it has become more difficult to establish criteria for diagnosis.

At the present time it is likely that initiation of the inflammatory cascade leading to allergic fungal rhinosinusitis is a multifactorial event, requiring the simultaneous occurrence of such things as IgE-mediated sensitivity, specific T-cell HLA receptor expression, exposure to specific fungi, and aberration of local mucosal defence mechanisms.¹⁷

There exists a controversy regarding the documented reports of histologic invasion in possible cases of allergic fungal rhinosinusitis. Thus it can be considered that apart from clinical diagnosis, type I hypersensitivity, Eosinophilia, Charcot-Leyden crystal may also be considered important criteria to define this entity.

Although the management of allergic fungal rhinosinusitis has advanced tremendously with better understanding of the underlying pathogenesis, the diagnostic strategies are still far from clear and still emerging. As clinical evidence of allergic fungal rhinosinusitis emerged, controversy regarding its diagnosis remained naive still and despite past and current efforts, many of these controversies remain incompletely resolved. Continuing clinical study has illuminated some aspects of the disease but still lot of research work has to be carried out regarding the relevant diagnostic criteria for the disease. The possibility of inclusion and usefulness, efficacy of Antigen specific IgE, microscopic evaluation of allergic mucin obtained for evaluation of degranulation products and proteins obtained after degranulation and serological testing of precipitating antibodies in the diagnostic criteria for Allergic Fungal Rhinosinusitis can be considered.

Acknowledgement

We express our sincere and heartfelt gratitude to Dr. Col. C. G. Wilson, Principal, Kamineni Institute of medical sciences, Dr. Amrithlal Professor & Head, Department of ENT, Dr. Sailaja, Department of Pathology, and the staff, management of Kamineni Institute of Medical sciences, Narketpally for their support during this study.

References

- Bousquet J, Khaltaev N, Cruz AA, Denburg J, Fokkens WJ, Togias A, Zuberbier T, Baena Cagnani CE, Canonica GW, Van Weel C, Agache I. Allergic rhinitis and its impact on asthma (ARIA) 2008. Allergy 2008; 63(s86):8-160
- Meltzer EO, Hamilos DL, Hadley JA, Lanza DC, Marple BF, Nicklas RA, Bachert C, Baraniuk J, Baroody FM, Benninger MS, Brook I. Rhinosinusitis: establishing definitions for clinical research and patient care. Journal of Allergy and Clinical Immunology 2004; 114(6):155-212
- Bent III JP, Kuhn FA. Diagnosis of allergic fungal sinusitis. Otolaryngology—Head and Neck Surgery 1994;111(5):580-8
- Corey JP, Gungor A, Karnell M. Allergy for the laryngologist. Otolaryngologic Clinics of North America 1998; 31(1):189-205
- 5. Hadley JA. Evaluation and management of allergic rhinitis. Medical Clinics of North America 1999; 83(1):13-25

- Gupta RC, Sidharth RD, Gupta N, Mukesh M. Asthma and Applied Immunology, Jaipur Indian J Allergy Asthma Immunol. 2005; 19(2):99-124
- Corey JP, Gungor A, Karnell M. Allergy for the laryngologist. Otolaryngologic Clinics of North America 1998; 31(1):189-205
- Aribandi M, McCoy VA, Bazan III C. Imaging Features of Invasive and Noninvasive Fungal Sinusitis: A Review. Radiographics 2007; 27(5):1283-96
- 9. Bent III JP, Kuhn FA. Diagnosis of allergic fungal sinusitis. Otolaryngology-Head and Neck Surgery 1994; 111(5):580-8
- Kaur R, Lavanya S, Khurana N, Gulati A, Dhakad MS. Allergic Fungal Rhinosinusitis: A Study in a Tertiary Care Hospital in India. Journal of Allergy 2016; 2016:1-6
- 11. Haase G, Brakhage AA. Function of Melanin as a Factor in Pathogenesis. Human Fungal Pathogens. 2003; 12:67
- Benninger MS, Ferguson BJ, Hadley JA, Hamilos DL, Jacobs M, Kennedy DW, Lanza DC, Marple BF, Osguthorpe JD, Stankiewicz JA, Anon J. Adult chronic rhinosinusitis: definitions, diagnosis, epidemiology, and pathophysiology. Otolaryngology-Head and Neck Surgery 2003; 129(3):S1-32
- Rasool R, Shera IA, Nissar S, Shah ZA, Nayak N, Siddiqi MA, Sameer AS. Role of skin prick test in allergic disorders: a prospective study in Kashmiri population in light of review. Indian J Dermatology 2013; 58(1):12
- Yamashita K, Nabe T, Tomioka H, Kohno S. Repeated antigen inhalations alter chemical mediators that cause asthmatic obstruction in guinea pigs. The Japanese J Pharmacology 1999; 81(1):48-55
- 15. Ahmed A. Imaging of the paediatric paranasal sinuses: review article. SA J Radiology 2013; 17(3):91-7
- Bush, Robert K. "Fungal allergy as yet unsolved." Allergy frontiers: Clinical manifestations Springer Japan, 2009. 471-485
- 17. Haase G, Brakhage AA. Melanized Fungi Infecting Humans: Function of Melanin as. Human Fungal Pathogens. 2013;12:67.

Main Article

Comparison of Various Graft Materials in the Reconstruction of Ossicular Chain in Patients with Chronic Otitis Media: A Prospective Hospital Based Study

Niaz Fakhruddin,¹ Chethan Kumar Gangaiah,¹ Gopakumar KP,¹ Geogin George Thottan,¹ Mon Noufal¹

ABSTRACT

Ossicular chain reconstruction is the surgical procedure used to correct the hearing problems in patients with chronic otitis media (COM). In this era, where a large variety of innovative artificial prosthetic materials are being used to replace and reconstruct the ossicular chain, autografts still play a significant role.

Materials and Methods

Introduction

The present study included 40 patients. Study population was selected based on inclusion and exclusion criteria. The ossicular reconstructive procedure was done under anesthesia and according to the status of the ossicular chain. Temporalis fascia was used to close the perforation. Mainly two procedures were performed: one is intact canal wall and the other is canal wall down. **Results**

Most of the study population was middle aged and males were more compared to females. Thirty five patients had air bone gap above 30dB. In 38 patients, the incus had undergone necrosis. Most of the patients underwent short columella reconstruction. A closure of air bone gap with in 20 dB was achieved in 72.2% in patients with malleus stapes assembly. In short columella 22.2% of patients had closure of air bone gap within 20 dB. Incus remnant grafts gave better hearing gain. **Discussion**

The published literature on the result of use of sculptured ossicle and cartilage in tympanoplasty have been reviewed *Conclusion*

In the present study, cases with COM showed good hearing results patients implanted with autogenous cartilage and bone. *Keywords:*

Otitis Media, Suppurative; Tympanoplasty; Incus; Necrosis; Cartilage

The ear surgeons of today have at their disposal, a wide range of surgical procedures for the treatment of chronic otitis media (COM), with and without cholesteatoma.¹ The middle ear includes the tympanic cavity, the mastoid air cell system and the auditory tube. The parts to be considered are, in order; the tympanic membrane, the tympanic cavity and its contents, the mastoid anturm and mastoid air cells, and

1 - Department of ENT, Sree Mookambika Institute of Medical Sciences, Padanilam, Tamil Nadu

<u>Corresponding author:</u> Dr Chethan Kumar Gangaiah email: meetchethu2007@gmail.com the auditory tube.² Middle ear reconstruction can be done after successful removal of the disease. However, the primary aim of any surgical procedure is the complete removal of bone destroying disease. This could be either by canal wall up or canal wall down mastoidectomy.^{3,4}

For a successful ossicular reconstruction an air-filled middle ear and a functioning eustachian tube are very important prerequisites. The tympanic membrane must be intact, healthy and mobile. The ossicular reconstruction must be secure and stable. Grafts and biomaterials chosen for use in middle ear reconstruction ideally should not induce a sustained foreign body reaction, extrude or biodegrade.^{2,5} Most ear surgeons prefer to use healthy, fresh, autologous tissues whenever possible and the success rate with these materials is high. The second

choice has been preserved allogenic tissues. The deep external auditory meatus and middle ear are sites where immune rejection responses to a tissue allograft across major histocompatibility barrier are somewhat muted. Current preoperative otologic allograft preservation techniques also appear to make these tissues less susceptible to rejection after grafting, by altering, to a greater or lesser extent, the molecular configuration of antigenic determinants of transplanted antigens. This appears to diminish the ability of the graft to immunize the recipient, but does not alter their specificity.⁶ The present study was conducted to compare various graft materials in the reconstruction of ossicular chain in patients with COM.

Materials and Methods

This prospective study was conducted in a medical college in the state of Tamil Nadu. The study period was two years. This study was cleared from Institutional Research Committee and Institutional Human Ethics Committee. 40 patients were selected for this study. Informed consent was taken from all patients before initiation of study protocol. Demographic data (age, gender) and clinical data (perforated ear, type of symptom, type of perforation, air bone gap) of all patients and ear involved were noted in the case sheet. The procedures were explained to the study population.

Patients of both mucosal and squamous type of chronic otitis media with good cochlear reserve and good Eustachian tube function were selected. The Inclusion criteria were COM and willingness to be included in our study. Exclusion criteria were previous ear surgery, congenital ear abnormalities, sensorineural hearing loss, pregnancy and lactation and ototoxic medication (Aminoglycosides, diuretics).

Both intact canal wall and canal wall down procedures were included. The operations were performed under local or general anesthesia. Post aural or endaural incision were used. After clearing of disease from the middle ear and mastoid as necessary, the status of ossicular chain was assessed. Ossicular reconstructive procedure was planned according to the status of the ossicular chain. Temporalis fascia was used to close the perforation. In this study, we have included only the cases where autogenous cartilage (conchal) or autogenous bone (incus remnant), homograft septal cartilage was used between: (i) Malleus and head of stapes (malleus-stapes assembly), (ii) Malleus and foot plate (malleus footplate assembly), (iii) Stapes head and newly constructed tympanic membrane (short columella) and (iv) Footplate and newly constructed tympanic membrane (long columella).

All patients underwent audiometric assessment before surgery and three months and six months after surgery. Pure tone averages (500Hz, 1000Hz & 2000Hz) were compared between pre-operative and postoperative audiograms. Statistical analysis of the data was expressed in number and percentage. Statistical Package for Social Sciences (SPSS ver 16.0) was used for analysis. Chi square test was applied to find the statistical significance. p value less than 0.05 (p<0.05) was considered statically significant at 95% confidence interval.

Results

A total of 40 patients were included in the study. 30 patients were in the age group of 21-30 years. Males (28) were more compared to females (12) (Table I). 21 patients had disease in left ear, 12 in right ear and 7 in both ears. Hearing loss and otorrhoea were the most common complaint in the study population. Maximum number of patients were suffering from ear problem from last 3 years in the study population.

Central perforation is more common in the patients

PROCEDURE	NUMBER	PERCENTAGE (%)
Intact canal wall technique	32	80.00
Canal wall down	8	20.00
Total	40	100.00

Table I: Comparison of type of procedure among studypopulation

138

AIR BONE GAP (DR)	INTACT (CANAL WALL	CANAL W	ALL DOWN
	NUMBER	PERCENTAGE (%)	NUMBER	PERCENTAGE (%)
1-10	0	0	0	0
11-20	0	0	1	12.5
21-30	3	9.37	1	12.5
Above 30	29	90.63	6	75
Total	32	100	8	100

Table II: Pre-operative hearing loss in patients with type of surgery

compared to posterior and attic. 35 patients had an air bone gap more than 30 dB (Table II).

32 patients underwent intact canal well technique

 Table III: Comparison of intra operative ossicular status

OSSICULAR STATUS	NUMBER	PERCENTAGE (%)
Necrosed malleus	11	27.5
Necrosed incus	38	95.0
Absent stapes suprastructure	9	22.5

and others underwent canal wall down procedure. 29 patients in intact canal wall group and 6 in canal wall down group had air bone gap above 30 dB. 38 patients had necrosed incus, 11 had necrosed malleus and 9 had absent stapes superstructure (Table III).

The results varied according to the type of reconstruction. A closure of air bone gap with in 20 dB was achieved in 72.2% in patients where malleus stapes assembly was done. In short columella 22.2% of patients had closure of air bone gap within 20 dB. All the patients with the above described techniques had closure of air bone gap within 30 dB (Fig. 1). In patients with long columella, air bone gap less than 20 dB wasseen in 25%. As the results were assessed according to the type of

graft material used, it was found that the air bone gap closure less than 20dB in patients using incus remnants was 6.2%. In canal wall down technique, closure within 20 dB using septal cartilage was 12.5% (Tables IV, V and VI).

Discussion

This study was conducted to know the commonest ossicular pathology in COM and the various materials used for ossicular reconstruction and to compare the results with pre-operative and post-operative airbone gap hearing thresholds. This study included 40 patients clinically diagnosed as COM either mucosal or squamous type.

There were 28 males and 12 females. Left ear (52.5%) was more commonly involved than right ear (30%). 17.5% patients had bilateral ear disease. Homograft prosthesis was used exclusively in ossicular reconstruction from 1972 to 1986.^{7,8} Guildford et.al and others recommended transposing the residual autograft incus onto its side so that it lies on the stapes capitulum and beneath the manubrium.⁹ Zollnner et.al described the benefits of sculpturing the autologous incus in order to obtain a better assembly and reduce subsequent ankylosis.¹⁰ Wehrs et.al and others refined this technique and advocated the use of homograft ossicles.¹¹

The use of self-stabilizing pre-sculptured cartilage homografts for middle ear reconstruction has an established track record with long term results equivalent to those of reconstruction methods using alloplastic

	CLOSURE WITHIN 20DB		CLOSURE V	WITHIN 30 DB
RECONSTRUCTION	NUMBER PERCENTAGE (%)		NUMBER	PERCENTAGE (%)
Malleus stapes assembly	2	6.2	4	12.5
Malleus footplate assembly	-	-	2	6.2
Short columella	16	50	7	21.8
Long columella	-	-	1	3.1

Table IV: Distribution of patients based on type of reconstruction in intact canal wall technique

Table V: Distribution of patients according to the type of graft material (ICW technique;A: Auto graft, H: Homo graft)

TVPE OF GRAFT	CLOSURE	WITHIN 20DB	CLOSURE V	WITHIN 30 DB
MATERIAL	NUMBER	PERCENTAGE (%)	NUMBER	PERCENTAGE (%)
Incus remnant	2	6.2	25	84.4
Conchal cartilage (A)	1	3.1	-	<u> </u>
Septal cartilage (H)	-	-	4	

Table VI: Distribution of patients according to the type of graft material (CWD technique; A: Auto graft, H: Homo graft)

TYPE OF GRAFT	CLOSURE V	VITHIN 20DB	CLOSURE W	ITHIN 30 DB
MATERIAL	NUMBER	PERCENTAGE (%)	NUMBER	PERCENTAGE (%)
Incus remnant (A)	3	37.5	1	12.5
Septal cartilage (H)	1	12.5	3	37.5

materials.^{12,13} Austin et.al in their study reported good stability of hearing results with autografts.¹⁴ Black et al compared the results of malleus stapes assembly with malleus footplate assembly and achieved the closure of air bone gap to within 20dB in 86% of patients in the former and 80% in the latter.¹⁵ McGee and Hough reported excellent results of hearing i.e. air bone gap closure within 10dB with sculptured ossicles.

They observed that the type of ossicular defect influenced the success of the operation. The results of malleus stapes assembly (85% patients with AB gap

closure within 20dB) were better than malleus footplate assembly (73% patients with AB gap closure within 20 dB).¹⁶ Bauer et.al., analyzed his 34 years of experience of autogenous incus and cortical bone to form a collumella between stapes head and tympanic membrane. In their study 85% showed an air bone gap closure < 20 dB and 43% showed closure < 10 dB when the tympanic membrane was normal.¹⁷

We analyzed our results according to the type of reconstruction and found that malleus stapes assembly gave best results i.e., 72.72% within 20 dB and 100%



Fig. 1. Distribution of patients based on reconstruction in canal wall down procedure

within 30 dB and followed by short collumella with 22.22% within 20 dB and 100% within 30 dB in intact canal wall technique. Kartush et al found that the results of incus remnants and cortical bone were similar.¹⁸ They also found that the autogenous, bone provides better sound transmission than cartilage. In the present study too, we achieved better results with autogenous bone as compared to homograft cartilage. In this era, where a large variety of innovative artificial prosthetic materials are being used to replace and reconstruct the ossicular chain, autografts still play a significant role.

Conclusion

In the present study, in patients with chronic otitis media, we have found fairly good hearing results in patients implanted with autogenous cartilage and bone. These are easily available and cost effective. Moreover, they are stable and are easily accepted by the body and rarely extruded.

Limitations of study: The major limitation of this study is small sample size.

References

- 1. Muhammad MB, Muhammad A, Ifra S, Syeda F. Prevalence of cholesteatoma and its complications in patients of chronic suppurative otitis media. JRMC 2011;15(1):16-7
- Pinilla M, Garcia BJR, Ramirez C, Bujan J, Jorge HE. Biomaterials in the reconstruction of the human middle ear. JMSMM 1995; 6(12):745-8
- Pramila Padmini M, Narasinga Rao B. Morphological variations in human fetal ear ossicles- A study. Int J Anat Res. 2013; 1(2):40-2
- Hisham SK, Paul CWT. Canal wall down mastoidectomy: A long term commitment to the outpatients. BMC Ear Nose Throat Disord. 2003; 3:1
- 5. Ricardo FB, Anna CDOF. A brief history of mastoidectomy. Int Arch Otorhinolaryngol. 2013; 17(2):168-78
- Siva SRP, Srinivasa V, Elangovan S, Jarvis R. Ossicular chain abnormalities in chronic suppurative oitis media and its management. JEMDS 2013; 2(38):7263-6
- Shrinivas SC, Prateek VJ, Jeevan NV, Dharmendra KR, Himayat K. Ossiculoplasty: A Prospective study of 80 cases. Iran J Otorhinolaryngol. 2014; 26(76):143-50
- 8. Wehrs RF. Homograft ossicles in tympanoplasty. Laryngoscope 1982; 92:540-6
- Guildford F. Repositioning of the incus. Laryngoscope 1965; 75:236

- Zollner F. Die Schalleitung's plastiken. Arch Otol. (Stockh) 1955; 45:168
- 11. Wehr's R. Results of homografts in middle ear surgery. Laryngoscope 1978; 88:808-15
- 12. Chole RA. Ossicular replacement with self-stabilizing presculptured homologous cartilage. Arch Otolaryngol Head Neck surgery 1982; 108:560-2
- 13. Chole RA. Use of presculptured, banked cartilage from plants in ossicular reconstruction. Arch Otolaryngol. 1987; 113:145-8
- 14. Austin DF. Ossicular reconstruction. Otolaryngology clinics of

North America 1972; 5:145-60

- Black B. Spanner malleus/footplate assembly. Laryngoscope 1994; 104:775-8
- Mc Gee, M, Hough J.U.D. Ossiculoplasty. Otolaryngology clinics of North America 1999; 32(3):471-87
- 17. Bauer M. Ossiculoplasty: Autogenous bone grafts, 34 years experience. Clinical otolaryngology 2000; 25:257-63
- Kartush JM. Ossicular chain recorstruction. Otolaryngology clinics of North America 1999; 27:689-715.

Does Anterior Tunnelling Really Help in Underlay Type I Tympanoplasty?

Sohag Kundu,¹ Bhaskar Ghosh,¹ Bijan Kumar Adhikary,¹ Mainak Dutta¹

ABSTRACT

Introduction:

Stabilizing the graft can be difficult with the conventional method of underlay tympanoplasty when the tympanic membrane perforation is subtotal, large or anteriorly placed with thin anterior rim. Tympanoplasty with anterior tunnelling has been tried to overcome this problem.

Materials and Methods

A prospective study over two-year period was carried out with follow up for three months on 59 patients under two groups- the underlay tympanoplasty with anterior tunnelling and the conventional tympanoplasty with anterior tucking for comparison in terms of pre-and post-operative anatomical correction and physiological improvements

<u>Results</u>

Follow up at 6 weeks and 12 weeks post operatively gives statistically comparable graft take up, hearing result and residual perforations.

Discussion

Among various techniques of dealing with these types of perforations, statistical comparability of the two groups brings in an acceptability to this simple but satisfying procedure of the underlay tympanoplasty with anterior tunnelling.

<u>Conclusion</u>

Underlay tympanoplasty technique (type-I) for subtotal, large or anteriorly placed perforations with thin anterior rim, can be managed by combining with anterior tunnelling which provides at least comparable results (if not more security against graft medialization) in respect of anatomical closure of perforations and hearing outcomes. <u>Keywords:</u>

Tympanoplasty; Anterior Tunnelling

Il of us are aware of the uncertainty related to the postoperative results of tympanoplasty especially in large or subtotal perforations, which are prone to graft medialization, residual perforations or reperforations and lack of significant improvements in hearing.

This article provides a comparison between the anterior tunnelling technique of underlay tympanoplasty and the anterior tucking technique of conventional ones i.e. the underlay tympanoplasty with anterior tunnelling (ut+T) and the conventional tympanoplasty group with anterior tucking (ut+at), with regards to the graft uptake

1 - Department of ENT, Medical College, Kolkata

<u>Corresponding author:</u> Dr Bhaskar Ghosh email: docgvas@gmail.com rates and physiological restoration of hearing.

Underlay tympanoplasty with anterior tunnelling differs from conventional ones with anterior tucking in that the fascial flange is taken out through an anteriorly created tunnel over the antero-superior portion of the deep meatus adjacent to the drum annulus thereby providing additional support; whereas in conventional underlay tympanoplasty with anterior tucking the anterior end of the fascia is blindly placed beneath the anterior drum remnant supposedly over the deep meatal bony canal with gel foam support beneath medially.¹

Materials and Methods

Fifty-nine patients with inactive (for more than 3 weeks) large to subtotal or anteriorly placed central perforations (involving antero superior part of drum) with very minimal

anterior drum remnant were included in this prospective study where every alternate patient was distributed between the two cohorts under comparison. The patients had undergone underlay Type I tympanoplasty with anterior tunnelling or the conventional Type I tympanoplasty with anterior tucking, depending upon the group they belonged to.

The study population of 59 patients was further statistically compared in terms of demography, pre-and post-operative anatomical correction and physiological improvements. The patients were recruited during the two-year period from May 2014 to April 2016 and each patient was subsequently followed up for 3 months in the post-operative period.

Chronic otitis media with squamous disease, retraction pocket, cholesteatoma or marginal perforation or ears needing atticotomy/ mastoid exploration were excluded from the study, as also the cases of ossiculoplasty. Patient below twelve years of age or patients with bilateral perforations² were also excluded from the study.

Preoperative evaluation - Patients in the OPD presenting with COM were followed with proper elicitation of history, examination with otoscope and microscope, estimation of hearing with tuning-fork tests, pure tone audiometry at 0.5, 1, 2, and 3 kHz and assessment of nose and throat with rhinoscopy and indirect laryngoscopy accompanied by vestibular function tests and cranial nerve examinations, if needed. Lateral oblique x-ray of the mastoids and haematological investigations for pre-anaesthetic check-up were done next. High-resolution computed tomography of the temporal bones, tympanometry and occipito-mental view of paranasal sinuses were obtained in selected cases. Those fulfilling the criteria were put up for the study.

Surgical technique - After infiltration with 2% lidocaine with 1:200,000 adrenaline, all patients were operated through a post aural approach (Wilde's incision) by the same set of surgeons to gain surgical access after elevating a prior vascular strip end aurally.³ After elevation of tympanomeatal flap and freshening of the margin, ossicular mobility and continuity were checked. In cases of an intact and mobile ossicular assembly and no evidence of squamous disease, patients

were subjected to randomization to either the underlay tympanoplasty with anterior tunnelling (ut+T) or the conventional tympanoplasty group with anterior tucking (ut+at) by allocating sequentially.

Temporalis fascia is harvested through the same incision line and wet temporalis fascia graft trimmed to approximately double the area of the perforation⁴ was placed in an over-underlay fashion by placing the graft over the malleus and under the annulus.⁵

For the anterior tunnel group, a small horizontal incision about 3 mm in size (enough for passage of the tip of an alligator forceps and approximate the size of the diameter of a round knife) is placed, about one millimetre lateral to the annulus, in the antero superior portion of deep meatus and a suitable tunnel is created with the help of a round knife (Fig. 1) by medially elevating a small cuff of deep meatal skin along with the annulus along the anterior bony wall (starting at about one o'clock for the right ear and eleven o'clock for the left). The completion of the tunnel is judged by passing a curved needle through the tunnel into the anterior mesotympanum (Fig. 2). Through this tunnel, the tip of temporalis graft is pulled out with the aid of tip of micro suction cannula (Fig. 3) or an alligator forceps and the projection of graft is made to rest in between the skin cuff and meatal bony wall- (Fig. 4) and (Fig. 5 schematic).

For the conventional underlay tympanoplasty (type



Fig. 1. Incision for the tunnel- left ear



Fig. 2. Boring the tunnel and checking it with curved needle and suction tip

I) anterior tucking is done by dislodging the anterior aspect of the annulus from the tympanic sulcus with a Cawthorne's knife and dragging it along with the anterior drum remnant laterally by scraping over the anterior deep bony meatal wall, thereby raising a miniscule flap for an amount deemed sufficient for tucking the anterior end of the fascial graft with the help of a flap repositor from the medial aspect of the graft, blindly. Middle ear and canal packing was done next with gel foams. Postural wound was closed in layers.

Follow up - Peri and postoperative cephalosporins were injected for 2 days; the aural pack was impregnated



Fig. 4. Tucking the rest after closure of the tunnel



Fig. 3. Taking the fascial flange out through the tunnel

with framycetin. Removal of mastoid bandage, aural pack and stitches were done on the seventh postoperative day. Oral antibiotic coverage was continued for three weeks following operation. Gel foams were removed manually after two weeks and any remnant, with ofloxacin eardrop instillation for the next one week (canal toileting). Thus, patients were followed up weekly for the first three weeks. Primary surgical success was assessed by closure of anatomical gap. The patients were reassessed at sixth and twelfth weeks for proper epithelization and graft uptake, and hearing status respectively.

Results

Initially 66 patients were selected for the study- 32 in the ut+at and 34 in ut+ tunnelling groups. (Table I) In the ut+at group 3 patients were lost to follow up at twelfth week. The attrition for the latter group is 4. Thus, the study was completed with 59 patients- 29 in ut+at, 30 in ut+T.

The ut+at group was made up of 12 males and 17 females, aged 20 to 50 years (mean: 30), and the ut+T comprised of 14 males and 16 females, aged 18 to 55 years (mean: 37). The two groups were thus more or less alike in terms of preoperative demographic variables.

At 6 weeks, residual or re-perforations were observed in 4 of the remaining 29 patients in the ut+at group and in 1 of the 30 in the ut+T group. Thus, the anatomic



Fig. 5. Schematic diagram showing the essential steps of tympanoplasty with anterior tunnelling.

success in the ut+at group was 86.2%, and that of the ut+T group, the success was found to be 96.67%. These results were not found to be statistically significant (p value 0.329 with Yate's correction, df=1, Chi square value=4.96).

No anterior canal blunting, canal wall sagging or granulation tissue were found in the two groups.

The average air bone gap closure within 30 dB at 6 weeks and 12 weeks show a rise of 10 % and 3% of effective study population respectively for the ut+T and ut+at group. The values, individually at 6 and 12 weeks, for these two groups are not statistically significant. (See Table I - p value 0.71 at 6 weeks, 0.365 at 12 weeks)

At 3 months, no significant differences in terms of pure tone average and gain were found between the two groups. No morphological changes were observed in the form of delayed graft failures, or retraction or thinning of neo tympanic membranes.

The failures with anterosuperior quadrant residual perforation in the ut+at group totalling 3, were caused by medial graft displacement. They were treated surgically after a sufficient span of 6 months and on inability of Valsalva's manoeuvre to repose the graft. The one patient in the ut+T group, who developed a tiny perforation at 6 weeks, in the middle of the neotympanum, was managed with conservative TCA cautery.

Discussion

The large or subtotal or anteriorly placed medium sized perforation usually pose a problem in that we can never be sure of the ultimate fate of graft uptake because of the precarious anatomical thin anterior margin which prevents a proper overlap of raw residual tympanic remnant with fascial graft as well as access to the bony deep meatal shelf margin in case where a large tympano meatal flap has to be elevated due to an anteriorly located perforation. Also, this prevents a proper support for the anterior most part of the graft which is often laid to rest against the lateral aspect of eustachian tube with medially placed gel foams in the protympanum. Medialisation of graft most often at antero-superior portion of perforation is not uncommon. 146

Table I. Summary of the characteristics of the two groups of study populations pre-operatively, at 6 weeks and at 12 weeks

PROCEDURES	UT+AT (UNDERLAY TYMPANOPLASTY TYPE-1 + ANTERIOR TUCKING)	UT+T (underlay tympanoplasty type-1 + tunnelling)
N (total 66)	32	34
Attrition	3	4
Effective study population	29	30
Male: Female	12:17	14:16
Age range (Yrs)	20-50	18-55
Average Age (Yrs)	30	37
Av preop PTA value (dB)	33	35
Av post op PTA 6 weeks (dB)	30	29
Av post op PTA 12 weeks (dB)	26	27.5
Average ABg closure within 30 dB at 6 weeks	16 (55.17%)	18 (60%)
Average ABg closure within 30 dB at 12 weeks	17 (58.62%)	21 (70%)
Residual perforation at 6 weeks	4 (13.8%)	1 (3.33%)
Morphological changes in the neograft at 3 months	0	0

Nativitial population styleaverlagen Algy iAird Boyingapt. PTA- Pure Tone Average solve this problem. For anteriorly located small perforations a collar button suture⁶ can be done by placing a small incision over the anterior drum remnant through which the tip of graft can be taken out like a collar button stud providing anchorage. But this is not possible in our scenario where the anteriorly located medium to large perforation bears a very minimal tympanic rim.

Over-Underlay technique, where graft is placed over handle of malleus, as has been carried out in this study, provides additional support to a large fascia in the subtotal perforations if done with finesse and preventing too much manipulation of handle of malleus.⁷

Circumferential sub annular grafting of Mokhtarinejad et al⁸ where the annulus is elevated from sulcus tympanicus in the anterior segment for placement of the graft in between anterior annulus and anterior canal wall, which well-nigh mimics our conventional ut+at group, is another option if done with care to prevent anterior blunting. However, our ut+T group does away with this problem altogether as it requires very minimal raising of the annulus in the limited portion of the tunnel.

The superiorly based flap for anterior or subtotal perforations as described by Lee et al⁹ is procedurally challenging with respect to ut+T.

In our study, placement of the graft, under vision, in the anterior canal wall, by drawing the anterior tip of the tongue shaped temporalis fascia through the tunnel created in the modified Underlay tympanoplasty type-1 with tunnelling, not only ensures graft anchorage in the anterior bony wall but also obviates the need of gel foam placement in the middle ear if the eustachian function is good.

The anatomic success as evinced by the graft take up rate at 3 months and the hearing at 3 months were statistically comparable for both ut+T group and ut+at group ; this is not only comparable to other studies of Ganguly et al, Gupta et al^{10, 11} but the statistical comparability of the two groups brings in a new dimension to the problem of graft failure in large to subtotal perforations by shedding the inhibitions in accepting this simple but satisfying procedure of the underlay tympanoplasty with anterior tunnelling (ut+T).

Conclusion

The Underlay tympanoplasty type-I with tunnelling is at least as effective if not better than the conventional underlay tympanoplasty type-I with anterior tucking, both in terms of anatomical as well as physiological dispositions as evinced from our present study. A larger sample size and a longer duration would have given a better idea but is left for futurity.

References

- Athanasiadis-Sismanis A. Tympanoplasty: tympanic membrane repair, p475-477 in: Gulya AJ, Minor LB, Poe D. Glasscock-Shambaugh Surgery of the Ear (6th Edition). Shelton, Ct.: People's Medical Publishing House-USA; 2010
- Saha AK, Munsi DM, Ghosh SN. Evaluation of improvement of hearing in type I tympanoplasty & its influencing factors. IJLO. 2006;58(3):253-257
- Vadiya SI, Shah SK, Chaudhary M. Comparison of canal wall incisions for tympanoplasty for large central perforations. Indian J Otol. 2015; 21:186-9

- 4. Merchant SN, Rosowski JJ, Shelton C. Reconstruction of the middle ear, p240 in: Ed Snow JB, Wackym PA. Ballenger's Otolaryngology Head and Neck Surgery (17th Edition). Shelton: PMPH USA, Ltd.; 2014
- 5. Kartush JM, Michaelides EM, Becvarovski Z, LaRouere MJ. Over under tympanoplasty. Laryngoscope 2002; 112:802-7
- Poe SD. Perforations of the tympanic membrane. In: Ed Nadol JB, McKenna MJ. Surgery of the Ear and Temporal Bone (2nd edition). Philadelphia: Lippincott Williams and Wilkins; 2004
- 7. F. Fiorino, F Barbieri. 'Over-underlay' myringoplasty with umbo-anchor graft. J Laryngol Otol. 2008:122(8); 854-7
- Mokhtarinejad F, Okhovat SA, Barzegar F. Surgical and hearing results of the circumferential sub annular grafting technique in tympanoplasty: A randomized clinical study. Am J Otolaryngol. 2012; 33:75-9
- Lee HY, Auo HJ, Kang JM. Loop overlay tympanoplasty for anterior or subtotal perforations. Auris Nasus Larynx 2010; 37:162-6
- Ganguly SN et al. Underlay tympanoplasty with tunnelling and placement of graft in anterior canal wall. Journal of College of Medical Sciences-Nepal 2011;7(4):44-47
- 11. Gupta S, Kalsotra P. Hearing gain in different types of tympanoplasties. Indian J Otol. 2013(19) Issue 4:186-193.

Role of Polidocanol as Sclerosant in Treatment of Hemangiomas of Head and Neck Region

Rupanjita Sangma,¹ Mukul Patar¹

Introduction

ABSTRACT

Haemangiomas are common presentation in head and neck, prevalence being 60% followed by 25% and 15% respectively in trunk and limbs. This report studies the efficacy of Polidocanol as sclerosant in the treatment of heamangiomas in head and neck.

Materials and Methods

The two year prospective study conducted from January 2015 to December 2016 with Polidocanol as sclerosant on 55 patients attending the department of ENT. Intralesional injections of 3 % polidocanol were given at 2 week intervals.

<u>Results</u>

Out of 55 patients 15 cases did not follow up after the first dose, so results were calculated out of 40 patients. 12 patients showed complete regression & 15 showed regression to half the size. Thus 67.5 % patients showed acceptable results. There were no side effects except hyperpigmentation in 2 patients. There were no cases of recurrence during our study period. <u>Conclusion</u>

Sclerotherapy is a promising method of treatment for haemangiomas of head and neck that may obviate the need for surgical intervention.

<u>Keywords</u>

Sclerotherapy ; Hemangioma ; Polidocanol

Hemangioma is benign developmental abnormality of blood vessels due to proliferation of endothelial lining of blood vessels. Hemangioma is usually not seen at birth. A majority of hemangiomas appear during the first 6 weeks of life. These lesions occur more commonly in females, with a ratio of 3:1 (F:M). This tumor is also more frequent in whites than in blacks. Clinically, hemangioma is characterized by a rapid postnatal growth (the proliferative phase) for the first 8 to 12 months, followed by a slow regression over 5 to 8 years (involutive phase). Hemangioma in its proliferating phase is composed of rapidly dividing endothelial cells forming syncytial masses with or without lumens.

1 - Department of ENT, Jorhat Medical College, Jorhat, Assam

Corresponding author: Dr Mukul Patar email: patarmukul@gmail.com During the involutive phase, endothelial cell activity diminishes and the cellular parenchyma is replaced by fibrofatty tissue. The first sign of a hemangioma is a macular patch that blanches on pressure or a localized area of telangiectasia surrounded by a halo. Rarely, a fully-grown hemangioma is present at birth. Eighty percent of hemangiomas occur as an isolated lesion, whereas 20% are multiple hemangiomas.

The most common site is the head and neck region (60%), followed by the trunk (25%), and then the limbs (15%). Cutaneous hemangiomas are superficial (capillary) in approximately 60% of cases and deep (cavernous) in 15% of cases.

Most hemangiomas in the head-and-neck region grow as small tumors and invariably regress, leaving inconsequential skin changes. Clinical studies confirm that complete resolution of hemangiomas occurs in over 50% of children by age 5 years and in over 70% by the age of 7 years, with continued improvement in

the remaining children until ages 10 to 12. Typically, the skin after involution exhibits mild atrophy, or it may have a wrinkled quality, or a few telangiectatic vessels. The skin may be slightly pale than normal skin. Treatment is indicated for those lesions that do not regress. Besides surgical excision, various approaches like Laser, photocoagulation therapy, cryotherapy, thermocauterization, corticosteroids, interferon alpha 2a and sclerotherapy have been in use.^{1,2,3}

Intralesional injection of sclerosants is one of treatment modalities for haemangiomas, which causes damage of blood vessels followed by their obliteration. Polidocanol has been used for years in the treatment of haemangiomas and varicose veins (as 3%, 1% or 0.5%)^{3,4,5,6} but the gold standard treatment for small circumscribed lesions or peripheral hemangiomas is surgical excision.⁷ Sclerotherapy is used because of its effectiveness, ease to application, inexpensive nature and ability to conserve the surrounding tissues⁸ with the aesthetic benefit, where surgery could leave unpleasant scarring.^{1,5,9} This prospective clinical study was undertaken to evaluate the efficacy of sclerotherapy with polidocanol (3%) injection in treatment of hemangiomas of head and neck region.

Materials and Methods

The study was conducted in the department of ENT. Study period was from January 2015 to December 2016. Total no of cases studied is 55. Fifteen cases were lost in follow up after the first dose, so results were calculated out of 40 patients.

Polidocanol 3% used as the sclerosing agent for treatment. Polidocanol has sclerosant and local anaesthetic effect too, hence the reason for almost painless sclerotherapy. Effect is directed mostly at the vein intima. It effects the intima, which causes fibrosis of the vessel and obliteration. Polidocanol is a safe drug as has been observed in our study. It causes thrombophlebitis of the vessels, thus leading to pain, tenderness and oedema at the site of the lesion. This subsides by the 2nd day. There is a remote possibility of the thrombus being dislodged which may lead to embolisation. So all the patients were hospitalized and kept under observation for 2 days after injection. In our study there was no incidence of embolism.

Diagnosis was done on basis of history, clinical examination and FNA. Ultrasonography was done for hemangiomas in external cheek and neck. Routine blood examination including TLC, DLC, ESR, Hb%, BT & CT was done for all patients.

Procedure: All patients of hemangioma were prepared for injection with 3% Polidocanol in ENT OT. Most of the patients did not require any anaesthesia. 10% lignocaine as surface anaesthesia was administered for lesions in oral cavity. General anaesthesia was required for 2 uncooperative paediatric patients. Intralesional injections of 3 % polidocanol were given at 2 week intervals. Patients on follow up were observed for regression of swelling and subsequently called for further doses. They were also observed for any side effects like hyperpigmentation, periphlebitis, necrosis, discoloration of skin etc.

Results

Twenty five (45.45%) patients were under the age of 15 years, 19 patients (34.54%) were between 15 to 30 years and 11 patients (20%) were above 30 years of age. Thirty six patients in this series were females (65.5%) and 19 were males (34.5%).

The sites of the haemangiomas are are noted in Table I.

The number patients treated with single dose therapy were 27, whereas 12 patients received 2 doses, 9 patients received 3 doses, 2 patients received 4 doses and 5 patients needed 5 doses.

Out of 40 cases who were followed up, 12 patients (30%) showed complete regression of swelling. (Figs. 1&2) (Table II).

Two patients with past history of surgery came presented with recurrence of swelling. One of them had recurrence of haemangioma of left external auditory canal two months after surgery. He was given 1 dose of sclerosant and swelling completely regressed in size. Another patient had haemangioma of preauricular region on right side for 14 years. She had past history of surgery few years back. She was given two doses of sclerosant at 2 weeks interval. Swelling size reduced to 150

#	SITES OF HEMANGIOMAS	NO. OF CASES
1.	Buccal mucosa	10 cases
2.	Cheek (external surface)	8 cases
3.	Tongue	17 cases
4.	Lip	6 cases
5.	Hard palate	3 cases
6.	Misc. (neck, post auricular region, pre auricular region, pinna , external audito-ry canal etc)	11 cases

Table I: Site wise distribution of haemangiomas (N=55)

less than 1 cm and no further increase in size noted at subsequent follow up. (Fig. 3)

15 patients (37.5%) showed regression of swelling to half the original size (Fig. 4) and 11 patients (32.5%) showed <50% reduction in size of the lesion.

There were no side effects except hyperpigmentation in 2 patients. These two patients had haemangioma of right pinna and post auricular region respectively.

Each of them received 1 dose, showed regression of

swelling, but with blackish discoloration of skin at the site of pedicle. These two patients were clubbed in the group showing less than 50% regression. There were no cases of recurrence during our study period.

Discussion

Most of the cutaneous vascular anomalies are haemangiomas, being more common in females. They are mainly superficial and present mostly on head and neck area followed by trunk.

Histologically, haemangiomasshow plumpendothelial cells with multilaminated basement membranes and numerous mast cells; immunohistochemistry demonstrates increased vitronectin, perlecan. The use of the immunohistochemical marker GLUT-1 to accurately distinguish haemangiomas (GLUT-1 positive) from vascular malformations has been advocated.

Up to 93% of haemangiomas are easily diagnosed without additional diagnostic tests. An ultrasound in experienced hands is a portable and available tool that can easily confirm a suspected haemangioma without additional testing. Doppler colour flow imaging is notable for its ability to distinguish between high-flow and low-flow lesions.

In our study majority of the lesions were between 5-8 cm and mostly uncomplicated, however ulceration and infections were observed in some cases especially those in oral cavity. The present study was conducted on 55 patients and only 40 patients had completed the



Fig. 1. Hemangioma of tongue (left lateral surface) received 5 doses and on follow up after 3 months showed complete regression (A)Before intervention (B) After 3 doses (C) After 5 doses

NO. OF PATIENTS	SITE	NO OF DOSES	FOLLOW UP
1	Left external auditory canal(POC)	1	2 months
1	Lower lip	5	3 months
2	Cheek	3	2 months
2	Cheek	4	3 months
2	Right cheek	5	3 months
1	Tongue(lateral surface)	5	3 months
1	Tongue(undersurface)	5	3 months
1	Neck	1	2 months
1	Right preauricular region(POC)	2	2 months

Table II: Complete regression of swelling

treatment and turned up for follow up. Out of 40 cases 12 (30%) cases showed complete regression of lesion, in 15 (37.5%) cases size of lesions regressed to half of original size and in 13 (32.5%) cases there was only slight regression in size of lesion. Agarwal in 2012 conducted one study in 20 cases of oral hemangioma and demonstrated high success rate by sclerotherapy with total regression of lesion in 19 cases and partial regression only in one case.³ Bhadoria et al have reported a case where they injected Polidocanol after diluting it with normal saline and observed regression with no complications.¹⁰ In our study we did not dilute the drug.

The study of Patel et al in 2015 on 10 cases of oral cavity haemangioma, shows satisfactory results with no

severe complications, that is comparable to our study.¹¹ Singh et al in 2012 reported a case of haemangioma tongue who was injected with polidocanol diluted with distilled water in the ratio 1:3. The patient experienced pain which was dealt with by giving oral analgesics. There was regression of swelling with no recurrence.¹²

The surgical treatment has its own risks and advantages, similar to other treatment modalities.¹³ Advantage of surgical treatment is that, it allows for a complete surgical excision of the lesion and microscopical diagnosis but with the risks of excessive bleeding, functional impairment of vital functions such as swallowing, speech and airway.¹³ Recurrences are fairly common if complete excision is not done and



Fig. 2. Hemangioma of tongue (undersurface) received 5 doses and on follow up after 3 months showed complete regression (A) before intervention (B) after 3 doses (C) after 5 doses

hence the need for non surgical modalities.

Another treatment option for treatment of hemangioma is the laser therapy and Crisan et al (2010) demonstrated laser therapy as a more effective treatment of vascular lesion than sclerotherapy procedure¹⁴ but Witman et al (2006) demonstrated the different complications from laser treatment of hemangiomas, including pain, ulceration, scarring or hyperpigmentation, skin atrophy and even life threatening bleeding.¹⁵

Sclerotherapy with polidocanol is a minimally invasive modality of treatment with negligible side effects. Patient compliance is high with very little or no



Fig. 3. (A) Recurrent haemangioma of right pre auricular region (B) received 2 doses and showed complete regression on follow up after 2 months

morbidity. No anaesthesia is required for most patients and hospital stay is also reduced. Also with sclerotherapy there are no risks of scarring when we compare with surgical management.

In our study, there were variations in number of injections according to the type and size of lesions, single dose therapy was given to 27 cases and maximum five doses given to five cases. Resmije A.A. et al (2016) mentioned two sessions of injections for the treatment of superficial hemangioma.¹³

Winter et al in 2000 published their experience with 132 patients with cavernous hemangiomas treated by



Fig. 4. Hemangioma of lip (A) after receiving 3 doses showed reduction of swelling to half the original size (B)

polidocanol and demonstrated a satisfactory response and requiring only one to three injections.⁶ The quantity of the drug and number of applications (doses) during the scerotherapy treatment depend on the size and location and involvement of adjacent structures and results should be evaluated before the next dose.^{1,4,5}

Conclusion

It is apparent that sclerotherapy is a valuable treatment option in the management of head and neck hemangiomas. As experience grows, its use will become more commonplace. Sclerotherapy with 3% polidocanol is a safe, effective and inexpensive method. It is a valuable and promising treatment of hemangiomas and may obviate the need for any surgical treatment.

References

- Rodrigues Johann ACB, Ferreira Aguiar MC, Vieira Do Carmo MA, Gomez RS, Castro WH, Mesquita RA. Sclerotherapy of benign oral vascular lesion with ethanolamine oleate: an open clinical trial with 30 lesions. Oral surgery, Oral Medicine, Oral Pathology, Oral Radiology and Endodontology 2005; 100(5):579-84
- Waner M, Suen JY, Dinehart S. Treatment of hemangiomas of the head and neck. Laryngoscope 1992; 102(10):1123-32
- Agarwal S. Treatment of oral hemangioma with 3% sodium tetradecyl sulphate: a study of 20 cases. Indian Journal of Otolaryngology and Head and Neck Surgery 2012; 64(3):205-7
- Minkow B, Loufer D, Gutman D. Treatment of oral hemangiomas with local sclerosing agents. International Journal of Oral Surgery 1979; 8(1):18-21
- 5. Selim H, Selim A, Khachemoune A, Metwally SAFA. Use

of sclerosing agent in the management of oral and perioral hemangiomas: review and case reports. Medical Science Monitor 2007; 13(9): CS114-9

- Winter H, Drager E, Sterry W. Slcerotherapy for treatment of hemangiomas. Dermatologic Surgery 2000; 26(2):105-8
- Cardoso CL, Da Silva LMP, Fernandes R, Rocha JF, Goncales ES, Ferreira O Junior, De Assis Taveira LA. Surgical approach of intraoral hemangioma. Odontologia Clinico-Cienttifica 2010; 9(2):177-80
- C. Bonet-Coloma, I. Minguez-Martinez, C. Palma-Carrio, S. Galan-Gil, M. Penarrocha-Diago, J. M. Minguez-Sanz. Clinical characteristics, treatment and outcome of 28 oral hemangiomas in pediatric patients. Medicina Oral, Patologia Oral Cirugia Bucal 2011; 16(1):E19-E22
- 9. Zanettini I, Zanettini RN, Gollo G. Sclerotherapy as an alternative treatment of oral vascular lesions. Revista de Clinica e Pesquisa Odontologica 2005; 2:119-26
- S.Bhadoria S, R. Saxena R, A.Lavania A. Management of haemangioma neck using sclerosing agent- a case report. Journal of College of Medical Sciences-Nepal 2012; 8(1):56-9
- Patel N, Prajapati BJ, Palas A, Shah HR, Meel AM, Dharajia D. Sclerotherapy for Haemangioma of Oral Cavity. Indian Journal of Applied Research 2015; 5(6):649
- Singh R, Agarwal S, Sinha ON, Gaur S: A case report- Inj. Polidocanol in Haemangioma of Tongue. Scholars Journal of Applied Medical Sciences (SJAMS) 2016; 4(7):2433-5
- Ademi Abdyli R, Abdyli Y, Perjuci F, Gashi A, Agani Z, Ahmedi J. Sclerotherapy of Intraoral Superficial Hemangioma. Case Reports in Dentistry. 2016;2016:4320102. doi:10.1155/2016/4320102.
- Crisan BV, Baciut M, Baciut G, Campian RS, Crisan L. Laser Treatment in Oral and Maxillofacial Hemangioma and Vascular Malformations. Timisoara Medical Journal 2010; 60(1):34-8
- Witman PM, Wagner AM, Scherer K, Waner M, Frieden IJ. Complications following pulsed dye laser treatment of superficial hemangiomas. Lasers in Surgery and Medicine 2006; 38(2):116-23.

Midline Nasal Dermoid - A Series of Thirteen Cases and Review of Literature

Vedula Padmini Saha,¹ Debangshu Ghosh,² Santanu Dutta,³ Somnath Saha,⁴ Sumit Kumar Basu²

ABSTRACT

Introduction:

Congenital midline nasal masses include nasal dermoid sinus cysts, encephaloceles and gliomas. They are rare malformations, of which dermoid cyst is a relatively common congenital abnormality. A systematic review of the clinical feature and management outcome of congenital midline nasal dermoids would help in predicting the outcome of such cases.

Materials and Methods

A prospective observation was made on a series of patients with nasal dermoids with or without sinus tract for a period of three years. Altogether thirteen patients were evaluated. Age of the patients ranged from two to twenty seven years. There were seven males and six females. Intracranial extension was ruled out radiologically. Excision of the dermoid cyst with or without sinus tract was undertaken under general anesthesia and followed up for one year.

Conclusion

Surgery is the gold standard treatment for nasal dermoids and complete excision and cosmetically good repair is mandatory for good results.

<u>Keywords:</u>

Dermoid Cyst; Nose

I solated congenital anomalies of the nose are very rare despite its complicated embryological development and intimate association with development of the face and brain.¹ Many fetuses die in utero due to severe structural abnormalities in this region.² Developmental anomalies of the nose encompass a diverse group of conditions. Embryologic developmental anomalies of the nose consist of congenital midline nasal masses (CMNMs) that include nasal dermoid sinus cysts (NDSC), gliomas and encephaloceles. They are rare malformations of which dermoid cyst is a relatively common congenital abnormality.^{1,3,4,5} Others are

1 - Department of Plastic and Reconstructive Surgery, RG Kar Medical College, Kolkata
2 - Department of ENT, Medical College, RG Kar Medical College, Kolkata
3 - Department of ENT, Chinsurah District Hospital, Hooghly

4 - Department of ENT, NRS Medical College, Kolkata

<u>Corresponding author:</u> Dr Debangshu Ghosh email: ghoshdr.d777@ymail.com nasal clefts, proboscis lateralis, arhinia, polyrrhinia, supernumerary nostrils, nasopharyngeal teratoma, and epignathus.^{4,6} They may also be a part of Beckwith-Wiedemann syndrome and other associated anomalies.^{5,7} Congenital nasal deformities were classified into four categories.⁸

Type I, hypoplasia and atrophy, represents paucity, atrophy, or underdevelopment of skin, subcutaneous tissue, muscle, cartilage, and/or bone. Type II, hyperplasia and duplications, representing anomalies of excess tissue, ranging from duplications of parts to complete multiples, are categorized here. In the type III category, clefts where the comprehensive and widely utilized Tessier classification of craniofacial clefts is applied. Type IV deformities consist of neoplasms and vascular anomalies.

Management of congenital nasal defect requires surgery and a team work encompassing skills from different specialities like Radiology, Maxillofacial surgery, Neurosurgery, Otolaryngology, Plastic surgery etc. Because of rarity of incidence and cosmetic challenge, these patients present a great challenge to the practising Otolaryngologist. Nasal dermoids are the

most common congenital nasal anomaly encountered in ENT clinics.^{3,4}

In this article we share, discuss and review the cases of midline nasal dermoids and their management at a tertiary care hospital to compare their pre and postoperative results and surgical complications.

Materials and Methods

The study was conducted from June 2009 to May 2012. Patients who presented at ENT OPD with midline nasal mass causing cosmetic deformity with or without sinus or abscess and discharge thereof were taken up for the study. A total of thirteen such patients were observed whose age ranges from two to twenty seven years. Among the patients, seven were males and six females and male:female ratio being almost equal (1.2:1). Computed tomography (CT) scan was performed in all patients to evaluate the disease and to follow its extension if any. All of them were found to have soft tissue shadow in between the two nasal bones at different levels due to the cyst, sinus or soft tissue mass in the scan.

All the patients and/or parents were counselled for surgery and informed consent was taken for surgery as well as for medical photography (both preoperative, intraoperative and postoperative) on special forms. All the patients were followed up for a period of one year postoperatively for any complications or recurrence and these, if any were noted at the end of one year postoperatively. Some cases deserve special mention.

One twenty seven year old male patient came for the treatment of recurrence of the nasal dorsal mass after a surgery done elsewhere two years back (Fig.1).

Another seven year old girl presented with infected cyst like swelling over dorsum of nose, who was diagnosed clinically as infected midline nasal dermoid with abscess formation and underwent incision and drainage followed by antibiotic treatment. After control of infection she was evaluated radiologically and prepared for surgery. One two year old boy presented with a cyst and a sinus simultaneously over the dorsum of nose. Among others three patients presented with cyst (Fig.2) and the rest with sinus tract over the dorsum of nose (Fig.3).

One patient presented with recurrence and was advised to do a sinogram preoperatively which showed persistence of sinus tract and soft tissue mass over dorsum of nose without having any intracranial connection (Fig.4). All the patients were dignosed clinically as having congenital nasal dermoids and evaluated radiologically by CT scan of nose and paranasal sinuses (PNS) (Fig. 5). Nobody was found to have any intracranial extension of the sinus tract.

Operative Procedure: We selected a uniform protocol

Fig.1. Dermoid cyst of nose with superadded skin inflammation





Fig.3. Midline nasal dermoid cyst with sinus(arrow) in a young female



Fig.4. Sinogram to delineate the sinus tract in a dermoid sinus with cyst

for management of the cases with nasal dermoids. After treatment with antibiotics and incision-drainage wherever necessary, a CT scan of nose and PNS was done in every patient. After preparing the patients who were declared fit for GA, proper counselling and informed consent obtained and patients were taken up for definitive surgical excision of the dermoid cyst with sinus tract where exists.

Careful preoperative examination was done to plan excision. Probing of the sinus tract and injection of dye (methylene blue) just before incision helped to trace the tract during excision. Majority of the cases were dealt with an elliptical incision around the cyst or sinus tract (Fig. 6), two separate horizontal incisions were given along the Langer's line in cases where cyst and the tract coexisted. Incision was so fashioned that not to lose excess skin over the dorsum of nose, so that during repair adequate skin was present and finally the scar would not be under tension.

After giving incision, soft tissue dissection was carried out gently to separate the cyst or tract from surrounding tissue. In two cases we got multiple branches of the sinus tract, one ended in lacrimal sac and other ended at the root of nose while rest others with blind sacs. In cases of cysts, we incised the periosteum over nasal bones to dissect it en-bloc with the cyst. External rhinoplasty approach was not used in any of these patients nor any medial or lateral rhinotomy or attempt to fracture nasal



Fig.5. Axial CT scan of a dermoid showing splaying of nasal bones with a subcutaneous mass under nasal dorsum

bones to reunite them again were made as because all the patients except one were below 18 years of age which is generally believed to be the minimum age limit for the maturity of facial skeleton.

After total excision of the cyst or tract, soft tissue gaping were sutured by 4-0 synthetic absorbable (vicryl®) suture and the skin was sutured by 4-0 monofilament (ethilon®) suture without tension.Zplasty closure was fashioned in one case to have best cosmetic result. Putting a drain is usually not needed



Fig.6. Excision of cyst becomes easy after delineation of sinus tract by injection of methylene blue dye

	FOLLO	r 18 m	15 m	20 m	12 m	16 m
	COMPLN	Ugly Scar	Nil	Post-op infection	Nil	Post-op infection
	SX.	+	+	+	+	+
	INITIAL TT.		ı	I/D+ Antibiotics	Antibiotics	I/D+ Antibiotics
ids	NASAL BONE	Present	Present	Present	Present	Present
ith midline nasal dermoi	INTRACRANIAL EXTENSION	NIL	NIL	NIL	NIL	NIL
data of the patients w	CLINICAL PRESENTATION.	Cyst+sinus	Sinus tract	Infected Cyst	Cyst	Cyst+Sinus (Infected)
I: Clinical	"AGE (YRS)/ SEX"	2/M	4/M	5/F	3/F	8/F
able	#	1	2	3	4	S

	V OUT COME	No	Do	Do	Do	Do	Do	Do	Do	Do	Do	Do	Do	Do
	FOLLOW	18 m	15 m	20 m	12 m	16 m	14 m	12 m	13 m	15 m	12 m	12 m	12 m	16 m
	COMPLN.	Ugly Scar	Nil	Post-op infection	Nil	Post-op infection	Nil	Nil	Nil	Post-op infection	Post-op infection	NIL	NIL	NIL
	SX.	+	+	+	+	+	+	+	+	+	+	+	+	+
	INITIAL TT.			I/D+ Antibiotics	Antibiotics	I/D+ Antibiotics		'		I/D+ Antibiotics	ı			
	BONE	Present	Present	Present	Present	Present	Present	Present	Present	Present	Present	Present	Present	Present
	INTRACRANIAL EXTENSION	NIL	NIL	NIL	NIL	NIL	NIL	NIL	NIL	NIL	NIL	NIL	NIL	NIL
in commend and to man	CLINICAL PRESENTATION.	Cyst+sinus	Sinus tract	Infected Cyst	Cyst	Cyst+Sinus (Infected)	Sinus tract	Cyst	Sinus tract	Infected Cyst	Cyst(Recurrent)	Cyst	Cyst	Cyst with sinus
I. CIIIIVAI	"AGE (YRS)/ SEX"	2/M	4/M	5/F	3/F	8/F	5/M	10/F	M/T	6/M	27/M	M/9	5/F	24/F
ante	#	1	7	e	4	w	6	Ъ	œ	6	10	11	12	13

if haemostasis is strictly maintained. Early removal of the sutures help to avoid ugly scar and the sutures were removed five days postoperatively. Suture removal was done under sedation specially in small children. Despite our honest effort one patient developed a bad scar postoperatively and an opinion from plastic surgeon was sought for scar revision which was planned as a second look surgery.

Results

In our study among 13 patients, seven were males and six were females. Seven patients presented with cysts, three patients presented with sinus and the rest three presented with both cyst and sinus. One patient presented to us with recurrence of the cyst, probably due to incomplete removal, two years after the previous surgery done elsewhere. Three patients required initial management with antibiotics and incision-drainage for infected nasal dermoid cysts which was followed by definitive surgery. No immediate complications such as major bleeding related to the surgery were observed. Minor complications such as postoperative infections were recorded in two lesions, and they were treated by antibiotics without any more sequelae. One patient developed a bad scar on nose postoperatively. All the results in this study were recorded in a proforma after at least one year follow up (Table I).

Discussion

As far as development of nose is concerned, it occurs through three distinct phases, (1) preskeletal phase- development of mesenchymal swelling surrounding external nasal placodes, (2) chondrocranial phase-development of cartilaginous framework and (3) ossification phase-influx of cellular elements and fusion of nasal skeletal elements.9 At about third week of fetal life, paired thickenings (the olfactory or nasal placodes) appear in the cranial ectoderm near the embryonic anterior neuropore. Invagination of these placodes results in formation of nasal pits, which as they deepen serve to delineate medial and lateral prominences of the frontonasal process.

Nasal dermoids are epithelial lined cavities or sinus tracts with variable number of skin appendages including hair follicles, sebaceous glands and eccrine glands that arise from clustures of epithelium trapped during ectodermal process. A true dermoid cyst may occur alone beneath the skin without a cutaneous opening or sinus superficial to nasal bones, appearing as slowly enlarging mass. The dermoid sinus with or without a cyst, on the other hand is an extensive lesion extending into nasal cartilage and bones. If skin remains attached to the fibrous tissues of the nasal capsule in the pre-nasal space or the ponticulus, a tract is formed between developing bones, many a times attached with dura, creating a tract between the nasal skin and the dura (Foramen caecum).

The sinus may also extend through foramen caecum between the frontal bones superiorly and the nasal bones inferiorly (the so-called ponticulus nasofrontalis). Post et al described an unusual presentation of extension of a NDSC into the frontal sinus itself requiring osteoplastic flap approach with a local midline incision.¹⁰ Nasal dermoids account for 3.7-12% of all dermoids of the head and neck and 1.1% of all the body dermoids.¹¹ These are typically midline lesions, manifesting anywhere from the columellar base to the glabella. Clinical presentation may be single or multiple lesions in the form of a nasal pit, mass or a fusiform tract, often infected (presenting as an abscess) with or without hair and sebaceous materials. These are slightly more prevalent in males than in females and are usually visible at birth. In one study, four out of twenty-five (16%) patients had intracranial extensions and 4% patients presented with recurrence within three years of surgical excision.¹¹ Re et al reported two cases of NDSCs with intracranial extensions which they managed with endoscopic endonasal procedures and, in their opinion, frequency of intracranial extensions of such cysts varies from 5% - 45%.³ In our series we could not get any such extensions.

Dermoids may extend intracranially and should be differentiated from encephaloceles both clinically and radiologically. A CT or magnetic resonance (MR) imaging aids in diagnosis as well as helps to determine the extent of intracranial involvement, if any. Radiological findings include fusiform swelling within the nasal septum, widening of the nasal vault and foramen caecum, bifid septum and crista galli, glabellar destruction, bony proliferation above cyst level, large ethmoidal cystic spaces etc.

Complications include abscess formation following infection (commonest), orbital/ periorbital cellulitis, osteomyelitis, meningitis and rarely brain abscess. There may also be sequlae of mass effect like new onset seizures.⁹

Craniofacial approach is required if intracranial extention is present. An incision and drainage with antibiotic treatment sometimes needed if secondary infection with abscess formation occurs, followed by excision of the tract. Various skin incisions like elliptical, Y, H type with or without medial nasal osteotomies may be needed depending upon the case variety.

According to a study, very large lesions those in adults in whom underlying bone and cartilage have been damaged by prior surgery or erosion and in patients with known intracranial extension, a longitudinal zig-zag rhinotomy is used for wide exposure.¹¹ The incisions are designed with limbs extending superiorly at angles greater than forty degrees but less than ninety degrees. Any fistulous opening is excised by fusiform excision. Scar prognosis is better than with a straight incision because the zig-zag are at less than ninety degrees to the relaxed skin tension lines running horizontally across the nose.

According to several studies^{1,5,9} open rhinoplasty approach with a stair-step columellar incision is one of the routes in many cases for the following advantages: ease of exposure, wide exposure of the nasal dorsum, controlled external osteotomies, ease of dorsal reconstruction, and wide exposure of the upper lateral cartilages and septum.

Timing of the surgery is decided considering the risk of infection or complications. Growth inhibition within the anterior facial skeleton can occur from extensive dissection, resulting in trauma to growth centres. Recurrence is attributed to incomplete excision. We got a single case of such recurrent dermoid cyst.

Conclusion

Although nasal dermoid sinus cysts are uncommon and complex lesions, they can be managed successfully with careful clinical assessment preoperative CT scans or MRI and appropriate surgery.

A minority of these patients will have intracranial extension, and the importance of preoperative CT scan is highlighted in our study to confirm the anatomy prior to surgery. Tailored definitive surgery including Z-plasty technique during skin closure and addressing defined anatomy and pathology, allows successful treatment with a low recurrence rate and fewer complications.

References

- Morgan DW, Evans JN, Developmental nasal anomalies. J Laryngol Otol.1990; 104(5): 394-403
- Chiu HH, Hsu WC, Shih JC, Tsao PN, Hsieh WS, Chou HC. The EXIT (ex utero intrapartum treatment) Procedure. J Formos Med. Assoc. 2008; 107(9):745-8
- Re M, Tarclini P, Macri G, Pasquini E. Endonasal endoscopic approach for intracranial nasal dermoid sinus cyst in children; Int J Pediatr Otorhinolaryngol. 2012, 76(8):1217-22
- Kennard CD, Rasmussen JE, Congenital nasal masses; diagnosis and management. J Dermatol Surg Oncol.1990, 16(11):1025-36
- Wardinsky TD, Pagon RA, Kropp RJ, et al. Nasal dermoid sinus cysts; association with intracranial extention and multiple malformations. Cleft Palate Craniofac. J. 1991; 28(1):87-95
- Hallak A, Jamjoom H, Hosseinzadeh T, Supernumerary nostrils; a case report & review. Asthetic Plast Surg. 2001; 25(3):241-3
- Broekmann ML, Hoving EW, Kho KH, et al. Nasal encephalocele in a child with Beckwith-Widemann Syndrome. J Neurosurg Pediatrics, 2008; 1(6):485-7
- Loosee JE, Kirschner RE, Whitaker LA et al. Congenital nasal anomalies: a classification scheme. Plast. Reconstr. Surg. 2004; 113(2):676-89
- Adil E, Huntley C, Choudhary A, Carr M. Congenital Nasal Obstruction: clinical and radiologic review. Eur J Pediatr, 2012; 17(4):641-50
- Post G, McMains KC, Kountakis SE. Adult nasal dermoid sinus cyst. Am J Otolaryngol. 2005; 26(6):403-5
- Blake WE, Chow CW, Holmes AD, Meara JG. Nasal dermoid sinus cysts-A retrospective review and discussion of investigation and management. Annals of Plast.surgery 2006; 57(5):535-40.

Congenital Midline Nasal Mass: Four Cases with Review of Literature

Sambhaji Govind Chintale,¹ Sonali Prafull Jatale,¹ Vilas Rambhahu Kirdak,¹ Kaleem Azeem Shaikh¹

ABSTRACT

Introduction

Congenital midline nasal masses include nasal dermoids, gliomas, encephaloceles. Although rare, these disorders are clinically important because of their potential for connection to the central nervous system. Preoperative knowledge of an intracranial connection is a necessity to allow for neurosurgical consultation and possible planning for craniotomy. This study discusses the clinical presentation of congenital midline nasal mass and the role of imaging modalities like CT scan and MRI in diagnosis and the surgical management.

Materials and Methods

This prospective study is carried from March 2014 to March 2016, during which 4 cases presented to the Otorhinolaryngology department. Pre-operative evaluation of the patients included endoscopic evaluation along with haematological investigations, CT Scan and MRI. The masses were removed with nasal endoscopic sinus surgery or by external approaches and neurosurgical intervention.

<u>Result</u>

The age of the patients ranged from 3 years to 25 years. Three of them were male and one female. There was one case of nasoethmoidal encephalocele and the other three were dermoids (intranasal dermoid cyst, nasal dermoid cyst and nasal dermoid sinus cyst).

Conclusion

Congenital midline nasal masses are rare. These disorders are clinically important because of their intracranial connection which require proper evaluation with radiological imaging like CT scan and/or MRI before FNAC and any surgical intervention. <u>Keywords</u>

Dermoid Cyst; Encephalocele; Tomography, X-Ray Computed

ongenital midline nasal masses include nasal dermoids, gliomas, encephaloceles. These are rare congenital anomalies, estimated to occur in 1:20,000 to 1:40,000 births.¹ Although rare, these disorders are clinically important because of their potential for connection to the central nervous system. Biopsy of a lesion with an intracranial connection can lead to meningitis or cerebrospinal fluid leak.

Preoperative knowledge of an intracranial connection is a necessity to allow for neurosurgical consultation and possible planning for craniotomy. Pre-operative evaluation is by CT scan and MRI.CT scan help in extent of lesion with any bony involvement and MRI helps in

1 - Department of ENT, JIIUS IIMSR, Jalna, Maharashtra

<u>Corresponding author:</u> Dr Sambhaji Govind Chintale email: drsamchinto@gmail.com intracranial soft tissue involvement. The differential diagnoses of a midline nasal mass include inflammatory lesions, traumatic deformity, benign neoplasms, malignant neoplasms and congenital masses.

Materials and Methods

This was a prospective study to evaluate the clinical presentation of congenital midline nasal mass and the role of imaging modalities of CT scan and MRI in diagnosis and its surgical management. Duration of study was from March 2014 to March 2016. Total number of patients studied were 4, of which the first case was nasoethmoidal encephalocele, the second was intra-nasal epidermoid cyst, the 3rd was nasal dermoid cyst and the 4th was nasal dermoid sinus cyst. All these cases reported to the Otorhinolaryngology OPD. All the patients were evaluated with haematological





Fig.1. Encephalocele of nasal dorsum with endoscopic picture

investigations, endoscopic examination and radiological imaging like CT scan and MRI and got operated with endoscopic sinus surgery or external approaches and neurosurgical intervention.

Case Reports

Here we present thecases of congenital midline nasal mass, who presented to the Otorhinolaryngology OPD. The number of congenital midlinenasal masses were four. Three cases were male ranging age from 3 years to 18 years and one was female of age 25 years. The need of radiological imagingwith CT scan and MRI in diagnosis of this congenital mass before FNAC and operative intervention are emphasized.

Case 1

An 18 year old male presented to the Otorhinolaryngology OPD with history of swelling over dorsum of nose since early childhood, history of partial nasal obstruction with no history of any visual symptoms or any sign and symptoms of intracranial involvement orevidence of any congenital abnormality on gross general examination.

The swelling present over the dorsum of nose was approximately 3cm x 2.5cm extending from root of nose over dorsum upto supratip of nose. On right side, the swelling extended upto nasolabial fold. On palpation, the swelling was soft to firm. No pulsation was seen on coughing. On anterior rhinoscopy, there was no mass in both the nasal cavities but on endoscopic examination, there was a bulge present at the roof of the right nasal cavity (Fig. 1).

CT scan showed a heterogenous solid cystic lesion involving the premaxillary and nasolabial region on



Fig.2. CT scan coronal view MRI sagittal and coronal view of encephalocele

162

right side causing remodelling along with erosion and irregularity with lytic defect of underlying nasal bone. On right side the lesion extended to right hypoplastic frontal sinus. The lesion passed through the two nasal bones causing expansion of the bridge to continue intracranially through anterior skull base defect. CT scan showed bilateral mild maxillary sinusitis with hypertrophied bilateral inferior and middle turbinates.

On MRI there was herniation of brain parenchymal involving gyrus rectus on right side through foramen caecum in to right ethmoid sinus with CSF intensity content extending in to nasal soft tissue. Gliotic changes noted in the herniated brain parenchymal with defect in the skull base.Gross focal dilatation of temporal horn of right lateral ventricle suggested possibility of colpocephaly.Skull base of the anterior cranial fossa was mildly depressed and anteroinferiorly truncated. Imaging features suggested right nasoethmoidal encephalocele (Fig. 2).

This lesion was exposed by giving paramedian incision vertically over the swelling on the nasal dorsum on right side.After exposure, the stalk was identified cut and ligated. The lesion could be excised completely. Intraoperative CSF leak was managed with the help of bipolar cautery, Surgicel® and fascia lata graft was placed over these. After operation the patient was fine.Anterior skull base defect is closed with nasal endoscopic procedure with the help of neurosurgeon. There was nasal dorsum defect due to loss some part of nasal cartilage nasal bone was intact and later on augmentation rhinoplasty was done to improve



Fig. 4. Intranasal dermoid cyst with depressed nose and endoscopic image

aesthetics of nose (Fig. 3).

Case 2

A 25 year old female patient presented with history of left nasal obstruction, left nasal mucopurulent discharge with depressed nose and left orbital protrusion for last two years.

On examination, nose was depressed at mid dorsum. On anterior rhinoscopy, there was mucopurulent discharge which was intermittent non-foul smelling. On endoscopic examination, there was a bulge in the roof of the left nasal cavity extending from the axilla of the middle turbinate to the lower left alar cartilage laterally and to the septal cartilage medially (Fig. 4).

CT scan showed cystic lesion in left nasal cavity extending to left frontal sinus with splaying of nasal bones suggestive of nasal epidermoid cyst. MRI showed soft cystic lesion in left nasal cavity approximately



Fig. 3. Intraoperative imageof encephalocele with specimen and histological slide image [H&E, 40x]



Fig. 5. Coronal CT scan image, sagittal and coronal MRI images of intranasal dermoid cyst

2 cm x 1cm with no intracranial extension (Fig.5).

This lesion which extended to the left frontal sinus was removed endoscopically. Augmentation rhinoplasty was done later on for cosmetic correction of the depressed nasal bridge (Fig. 6).

Case 3

An eight year old male patient presented with history of swelling over nose since 4 years with no history of nasal obstruction.

There was a swelling over the root of the nose between nasion and medial canthus of right eye approximately 1cm x 1cm (Fig. 7). On endoscopic examination, both nasal cavities were normal.

On CT scan, there was a small cystic lesion over dorsum of the nose, no evidence of erosion of underlying nasal and frontal bone. MRI showed soft cystic lesion having no intracranial connection.

Lesion was exposed through a vertical incision over

the swelling and was excised completely. Post operative follow-up period was uneventful.

Case 4

A 3 year old male patient presented with history of swelling over dorsum of nose, history of discharge from dorsum of the nose since last one year but there was no history of nasal obstruction.

On examination swelling of approx 1 cm x 1cm size was present over dorsum of the nose, extending from the root to mid-dorsum. White discharge could be expressed from a small pin-point sinus opening present over the swelling. (Fig. 8)

Anterior rhinoscopy findings were normal on both the sides. On CT scan, a soft cystic swelling was present over the dorsum of the nose without any underlying bone erosion. MRI findings show soft tissue cystic mass present over dorsum of nose only without intracranial connection of sinus tract, suggestive of nasal dermal



Fig. 6. Intraoperative image after removal of nasal dermoid cyst with frontal sinus exposure and histological image of dermoid cyst [H &E,40x]



Fig. 7. Nasal dermoid cyst



Fig.8 . Nasal dermoid cyst sinus

sinus cyst. A vertical elliptical incision was given around the sinus opening and the sinus tract was excised completely upto frontal bone junction to root of nose by cauterizing the end of the sinus tract. There was no intracranial extension.

Results

The most common presentation was nasal dorsal swelling (3 cases out of 4). The least common presentation was nasal obstruction (1 out of 4 cases). Three patients got operated by external approach with neurosurgical assistanceand one was by endoscopic approach. Post-operative results of all the four cases were excellent, with no post-operative complication. Two patients required augmentation rhinoplasty to improve facial appearance. There was no recurrence in the follow-up period.

Discussion

Encephalocele is anomalous herniation of meninges and brain matter beyond the boundary of the cranium. Globally the incidence of encephalocele is 1:35000 births.Aetiology is failure of fonticulus frontalis to close properly thus culminating in herniation.¹ Hereditary relation are found in family members who have developmental anomaly of central nervous system.²

As the defect is more pertinent to embryological development, an encephalocele entity is much more common in infants, and so may be considered for differential diagnosis of any mass related to nose, maxilla and forehead.³ Encephalocele found in older age group is very rare occurrence as in our case, the patient presented to us at age of 18 years. Encephalocele can be grouped according to the anatomical location in to occipital,sincipital and basal variants. Herniation through cribriform plate and fovea ethmoidalis are example of basal encephalocele.⁴ Similarly, they can also be classified as anterior and posterior. Anterior encephalocele again is divided into fronto-ethmoidal,naso-frontal,naso-orbital and nasoethmoidal.¹

Frontonasal type of frontoethmoidal encephalocele is the subtype that occur more frequently.⁵ CT scan and MRI are required to diagnose these pathology to note the exact location, extent and involvement of other structures.⁶ Turgut et al reported that there is a mortality of 46% if an encephalocele contains brain tissue and hence prompt repair of such a defect is required.⁷ Repair of frontonasal encephalocele in toddler require simple surgical closure for defect even for larger and big lesion.⁸ In comparison to children frontonasal encephalocele in adult is a complicated lesion to repair, as with increase in age there is increase in defect size. Gliotic tissue is also increased as well as the sinus size. So, these type of defects are complicated to close. We had to use bone, cartilage graft or other material to close them.⁹

Nasal dermoid is a developmental anomaly of nose. Unlike other craniofacial anomalies, nasal dermoid can present as a cyst, a sinus, a fistula and may have intracranial extension.¹⁰ Incidence is estimated at 1:20,000 to 1:40,000 births.^{1,11,12} Pathogenesis involve incomplete obliteration of of neuroectoderm frontonasal region.¹³ Progressive in developing enlargement of nasal dermoid cause soft tissue and bony deformity, local infection, meningitis and brain abscess and so require prompt investigation by CT scan and MRI.14,15 Surgical excision by midline vertical incision is common approach.¹⁶ Other approaches includes transverse incision, lateral rhinotomy, external rhinoplasty are other approaches.¹⁷⁻²¹

Conclusion

Congenital midline nasal masses include nasal dermoids,

nasal gliomas, and encephaloceles. These disorders are clinically important because of their potential for connection to the central nervous system. Biopsy of a lesion with an intracranial connection can lead to meningitis or cerebrospinal fluid leak. The treatment of these masses is surgical excision. Preoperative knowledge of an intracranial connection allows for neurosurgical consultation and planning for craniotomy. So we require imaging study of suchcongenital lesion by CT scan and MRI. Surgical strategy depends on the location and extent of the lesion, ranging from local excision via an open or endoscopic approach to a combined transcranial approaches.

References

- 1. Suwanwela C. Geographical distribution of fronto-ethmoidal encephalomeningocele. Br J Prev Soc Med. 1972; 26:193-8
- Kallen K. Maternal smoking, body mass index, and neural tube defects. Am J Epidemiol. 1998; 147:1103-11
- Rahbar R, Resto VA, Robson CD, Perez-Atayde AR, Goumnerova LC, McGill TJ, et al. Nasal glioma and encephalocele: diagnosis and management. Laryngoscope 2003; 113:2069-77
- Celin S. Contemporary diagnosis and management of anterior skull base cephalocele and cerebrospinal fluid leaks. In: Arriaga M D-DJ, editor. Neurosurgical Issues in Otolaryngology. Philadelphia: Lippincott Williams Wilkins 1999
- Mahatumarat C, Rojvachiranonda N, Taecholarn C. Frontoethmoidal encephalomeningocele: surgical correction by the Chula technique. Plast Reconstr Surg. 2003; 111:556-65
- CD P. Neuroradiologic imaging in craniofacial surgery. In: Lin KY OR, Jane JA, editor. Craniofacial Surgery: Science and Surgical Technique: Philadelphia: Saunders 2002; pp 153-60
- Turgut M, Ozcan OE, Benli K, Ozgen T, Gurcay O, Saglam S, et al. Congenital nasal encephalocele: a review of 35 cases. J Craniomaxillofac Surg. 1995; 23:1-5

- Satyarthee GD, Mahapatra AK. Craniofacial surgery for giant frontonasal encephalocele in a neonate. J Clin Neurosci. 2002; 9:593-5
- Agrawal A, Rao KS, Krishnamoorthy B, Shetty RB, Anand M, Jain H. Single stage craniofacial reconstruction for fronto-nasal encephalocele and hypertelorism in an adult. Singapore Med J. 2007; 48:e215-9
- 10. Sessions RB. Nasal dermal sinuses: new concepts and explanations. Laryngoscope 1982; 92(pt 2, suppl 29):1-28
- 11. Pratt LW. Midline cysts of the nasal dorsum: embryologic origin and treatment. Laryngoscope 1965; 75:968-80
- Hughes GB, Sharpino G, Hunt W, Tucker HM. Management of the congenital midline nasal mass: a review. Head Neck Surg. 1980; 2:222-33
- Weiss DD, Robson CD, Mulliken JB. Transnasal endoscopic excision of midline nasal dermoid from the anterior cranial base. Plast Reconstr Surg. 1998; 102:2119-23
- Uglietta JP, Boyko OB, Rippe DJ, Fuller GN, Schiff SJ, Heinz ER. Intracerebral extension of nasal dermoid cyst: CT appearance. J Comput Assist Tomogr. 1989; 13:1061-4
- Fornadley JA, Tami TA. The use of magnetic resonance imaging in the diagnosis of the nasal dermal sinus-cyst. Otolaryngol Head Neck Surg. 1989; 101:397-8
- Brunner H, Harned JW. Dermoid cysts of the dorsum of the nose. Arch Otolaryngol. 1942; 36:86-94
- Posnick JC, Bortoluzzi P, Armstrong DC, Drake JM. Intracranial nasal dermoid sinus cysts: computed tomographic scan findings and surgical results. Plast Reconstr Surg. 1994; 93:745-54; discussion 755-6
- Yavuzer R, Bier U, Jackson IT. Be careful: it might be a nasal dermoid cyst. Plast Reconstr Surg. 1999; 103:2082-3
- Denoyelle F, Ducroz V, Roger G, Garabedian EN. Nasal dermoid sinus cysts in children. Laryngoscope 1997; 107:795-800
- Rohrich RJ, Lowe JB, Schwartz MR. The role of open rhinoplasty in the management of nasal dermoid cysts. Plast Reconstr Surg. 1999; 104:1459-66; quiz 1467; discussion 1468
- Morrissey MS, Bailey CM. External rhinoplasty approach for nasal dermoids in children. Ear Nose Throat J. 1991; 70:445-9.

SkullBaseLangerhansCellHistiocytosiswithDiabetesInsipidusand Panhypopituitarism - A Rare ClinicalEntity

Anirban Ghosh,¹ Mithun Chaudhury,² Abhishek Mukherjee³

ABSTRACT

Introduction

A case of Langerhans cell histiocytosis (LGH) involving extensive area of base of skull resulting in panhypopituitarism and diabetes insipidus (DI) is reported.

Case Report

A 16 year old male presented with diminished vision, bilateral ptosis, left sided lateral rectus palsy, hypoesthesia of trigeminal nerve with nasal obstruction for last 5 months. There was polypoidal, bleeding mass in both nasal cavities. Contrast enhanced CT Scan showed a large homogenous mass arising from sphenoid extending into cavernous sinus and the suprasellar region. Endoscopic nasal biopsy revealed abundant Langerhans cell histiocytes, macrophages, neutrophils. Chemotherapy and radiotherapy were administered. But within 2 months the patient presented with Cushingoid features and further diminution of vision. Detailed work-up revealed Hypogonadotrophic hypogonadism and diabetes insipidus. Debulking of the tumour was done and left optic nerve decompression was done. PET scan was performed and showed large, well defined mass with increased FDG uptake in the skull base with suprasellar extension, reaching upto petrous temporal bone and causing bony erosion of ethmoid and sphenoid sinuses. Patient was then advised adjuvant chemotherapy.

Discussion

Langerhans cell histiocytosis is a rare group of disorders characterised by abnormal clonal proliferation and accumulation of abnormal dendritic cells. Involvement of base of skull is even rarer. Though diabetes insipidus has been reported in Langerhans cell histiocytosis involving pituitary, panhypopituitarism is rare. These combinations of extensive Langerhans cell histiocytosis of base skull with clinical features of Diabetes insipidus and panhypopituitarism makes this case a rare clinical entity. <u>Keywords</u>

Myeloproliferative Disorders; Langerhans Cell Histiocytosis; Hypogonadism; Diabetes Insipidus; Skull Base.

angerhans cell histiocytosis (LCH) is a part of a group of disorders called Histiocytosis characterised by abnormal proliferation of histiocytes. These cells are epidermal dendritic cells derived from bone marrow. LCH is manifested either as single bone disease or as multisystem multi organ disorder with clinical features of fever, lethargy, lytic

- 1 Hope nursing home, Raniganj
- 2 Medica Superspeciality Hospital, Kolkata
- 3 Suraksha Diagnostics, Kolkata

<u>Corresponding author:</u> Dr Anirban Ghosh email: way2anirban@gmail.com bone lesions. It is also known as Eosinophilic granuloma, Hand-Schuller Christian disease, Letterer- Siwe disease.

Case Report

A 16 year old male presented with diminished vision, bilateral ptosis, left sided lateral rectus palsy (Fig. 1), hypoesthesia of trigeminal nerve with nasal obstruction for last 5 months. On examination, the patient showed normal strength of upper and lower limbs, hypotonia, hyperreflexia and dorsiflexion in plantar reflex. There was polypoidal, bleeding mass in bothnasal cavities. Contrast enhanced CT Scan showed a large homogenous mass arising from sphenoid extending into cavernous



Fig. 1. Clinical photograph showing bilateral ptosis and left lateral rectus palsy

sinus and the suprasellar region, superiorly reaching upto optic chaisma, inferiorly extending upto hard palate, anteriorly occupying posterior nasal cavity (Fig. 2). Endoscopic nasal biopsy revealed abundant Langerhans cell histiocytes, macrophages, neutrophils (Fig. 3). Chest X-ray, skeletal survey, abdominal scans were normal.

Chemotherapy and radiotherapy were administered. But within 2 months the patient presented with



Fig.2. Coronal CT Scan showing isodense lesion in sphenoid (left>right) with extension into lateral recess, choana, destruction of lateral wall and involvement of left orbital apex

Cushingoid features and further diminution of vision. Detailed work-up revealed hypogonadotrophic insipidus. CECT hypogonadism and diabetes revealedthat the mass extended into suprasellar region, posteriorly upto clivus, involvement of both optic nerves and erosion of both ethmoids and sphenoids. MRI showed a predominantly T2 hypointense mass in the sphenoid sinus extending to the left orbital apex and compression of the left temporal lobe (Fig. 4) and



Fig.3. Photomicrograph of nasal tissue biopsy showing distension of nodal sinuses by reniform Langerhans cell surrounded by rims of abundant eosinophils (H&E, 40X)



Fig.4. Axial cut MRI showing predominantly T2 hypointense left sided sphenoid mass extending into left orbital apex and causing compression of left temporal lobe

extension into the clivus (Fig. 5).

Visual acuity in the right eye was diminished to finger counting close to the face and on the left side, it was 6/18. Debulking of the tumour was done and left optic nerve decompression was done. There was erosion of cribriform plate and lateral wall of sphenoid; no bone was found between dura and the mass. Patient developed exacerbation of diabetes insipidus that was managed with hydrocortisone and desmopressin.

PET scan was performed and showed large, well defined mass with increased FDG uptake in the skull base with suprasellar extension, reaching upto petrous temporal bone and causing bony erosion of ethmoid and sphenoid sinuses. Patient was then advised adjuvant chemotherapy.

Discussion

LCH may be unisystem (monoostotic or polyostotic) or multisystem multifocal. Unisystem LCH or Eosinophilic granuloma typically presents as slowly progressive lytic lesion of bones, generally involving skull bones, upper extremity long bones, ribs, pelvis, vertebrae.¹ Sphenoid



Fig.5. Sagittal section MRI showing heterogenous mass on T2 involving the sphenoid sinus and clivus

sinus involvement is extremely rare;² involvement of clivus, petrous apex have also been reported.³ Orbit involvement is also reported.⁴ In the present case involvement of sphenoid sinus, extension into cavernous sinus, parasellar, suprasellar region, orbit, petrous apex have been noted.

LCH usually affects patients of 1-10 years of age with peak incidence between 5-10 years and incidence of 1 in 2,00,000, ; even rarer in adults with incidence of 1:5,60,00. It is sporadic and non-hereditary. Multisystem disease usually affects patients aged less than 2 years and bears poorer prognosis.

Debate continues regarding the pathogenesis of LCH, whether reactive or neoplastic process. Comparative favourable prognosis, release of cytokines point towards reactive aetiopathology;⁵ whereas clonal proliferation of BRAF proto-oncogene in 57% of LCH biopsies⁶ classifies LCH as "Myeloproliferative disorder".

Clinically it produces non-specific inflammatory response resulting in fever, lethargy, weight loss. Bone involvement causes painful bone swelling, pathologic fractures due to osteolytic lesion. Skin rash in intertriginous region as scaly erythematous lesion to red papules is seen in 80% patients of LCH. 50% LCH patients present with lymphnode enlargement and 20% to 30% present with hepatosplenomegaly. Most common CNS affection in LCH is hypothalamic- pituitary axis and cerebellum.7 Less commonly meninges and brain parenchyma of temporal and occipital lobes are involved either by direct extension or by spread through epidural space.⁸ Diabetes insipidus is the most common endocrine manifestation of LCH. About 23% of LCH patients will have diabetes insipidus due to lack of Anti-Diuretic Hormone (ADH); more commonly seen in multisystem disorder and especially with skull and orbit involvement.9 Diabetes insipidus is caused by posterior pituitary infiltration by Langerhans cells causing local tissue damage by increased amount of Interleukin-2 (IL-2), Prostaglandin E2 (PGE-2).

Radiologically, both CT Scan and MRI are important tools for identifying and delineating boundaries of the disease process. At an early stage, the lesion and the brain parenchyma remain isodense in appearance on CT Scan, making it difficult to identify the lesion and its extent. MRI shows the mass isointense or hypointense to normal brain tissue with brilliant enhancement with Gadolinium contrast, thus making it investigation of choice.⁸ It also shows thickening of pituitary stalk with granulomatous mass lesion in the region of hypothalamus with absence of posterior pituitary bright spot.

Diagnosis is definitely histological. Presence of Langerhans cells with distinctive margin, pink granular cytoplasm accompanied by varying number of neutrophils, macrophages and multinucleated giant cells best visualised in Papanicolaou stain is the hallmark of diagnosis. A definitive diagnosis can be made by demonstrating Birbeck's granules in electron microscopy and positivity for Cluster Differentiation (CDI) antigen.¹⁰ S-100 protein and MHC-II expression are also noticed in LCH.¹¹ Treatment in solitary lesion is essentially excision and low dose radiotherapy. Mutifocal multisystem LCH requires chemotherapy. Prognosis of LCH is usually favourable, however young age at diagnosis, hepatosplenomegaly, thrombocytopenia, polyosteotic disease have poor prognosis.

References

- Stull MA, Kransdrorf MJ, Devaney KO. Langerhans cell histiocytosis of bone. Radiographics: a review publication of the Radiological Society of North America, Inc 1992 July 12(4):801-23
- Yu G, Huang F, Kong L, Kong X, Zhang L, Xu Q. Langerhans cell histiocytosis of the sphenoid sinus: a case report. Turk J Pediatr. 2010 Sep-Oct; 52(5):548-51
- 3, Krishna H, Behari S, Pal L, Chhabra AK, Banerji D, Chhabra DK, Jain VK. Solitary Langerhans-cell histiocytosis of the clivus and sphenoid sinus with parasellar and petrous extensions: case report and a review of literature. Surg Neurol. 2004 Nov; 62(5):447-54
- 4. Shetty SB, Mehta C. Langerhans cell histiocytosis of the orbit. Ind J Ophthalmology 2001; 49(4):267-8
- 5. Broadbent V, Davies EG, Heaf D, et al. Spontaneous remission of multisystem histiocytosis-X. Lancet 1984; 8371:253-4
- Badalian-Very G, Vergilio JA, Degar BA et al. Recruitment of BRAF mutations in Langerhans cell histiocytosis. Blood 2010; 116(11):1919-23
- 7. Favara BE, Jaffe R. Pathology of Langerhans cell histiocytosis. Hematol Oncol Clin North Am. 1987; 1:75-97
- 8. Graif M, Pennock JM. MR imaging of histiocytosis X in the central nervous system. AJNR Am J Neuroradiol. 1986; 7:21-3
- 9. Dunger DB, Broadbent V, Yeoman E, et al. The frequency and natural history of diabetes insipidus in children with Langerhans cell histiocytosis. N Engl J Med. 1989; 321: 1157-62
- Kramer TR, Noecker RJ, Miller JM, Clark LC. Langerhans cell histiocytosis with orbital involvement. Am J Ophthalmol. 1997; 124:814-24
- 11. Pohar-Marinsek Z, Us-Krasovec M. Cytology of Langrehans cell histiocytosis. Acta Cytol. 1996; 40:1257-64.

Hydatid Cyst of Tongue: A Diagnostic Challenge

Neeraj Aggarwal,¹ Tanaya Panja,¹ Khuku Biswas,¹ Titas Kar¹

ABSTRACT

Introduction

Hydatid disease or human cystic echinococcosis is a parasitic zoonosis, endemic in the cattle and dog rearing region worldwide as well as in some parts of India.Cystic echinococcosis affects mostly the liver and lung (80%), but tongue is one of the very rare sites.

Case Report

A 10 year old boy had presented with an isolated cystic lesion in tongue, which was expelled spontaneously with no residual lesion. On detailed examination, no other site in the body was involved. **Discussion**

Parasitic cyst of the tongue is rare entity. Microbiological and histopathological examination helped clinch the diagnosis Hydatid cyst should be considered as a differential diagnosis in isolated cystic lesion of tongue, especially in the risk group. <u>Keywords</u>

Echinococcosis; Cysts; Tongue

ydatid disease or human echinococcosis is a well-known parasitic zoonosis,endemic in the cattle and dog rearing region worldwide, mostly in the Mediterranean region, South America, Australia, Central Asia and East Africa.¹ The incidence of hydatid cyst in India is 1-200 per 1,00,000 population; mostly in Kashmir, AndhraPradesh, Gujrat and Tamil Nadu.² It occurs due to ingestion of gravid proglottids of Echinococcus granulosus.³

Dogs and other carnivores act as the definitive host where as cattle act as the intermediate host. Humans act as an accidental, intermediate and dead-end host. Hydatid disease is very rarely seen in the head and neck region.⁴ Here, we are presenting a case of hydatid cyst of tongue in a 10 year old child.

Case Report

A 10-year-old boy had presented in the

1 - Department of ENT, Medical College, Kolkata

Corresponding author: Dr Neeraj Aggarwal email: drneeraj5887@gmail.com Otorhinolaryngology Outpatients Department with a tongue swelling, incidentally discovered 1-2 months back and gradually increasing in size, with history of contact with dogs and frequent tonguebite. The swelling was soft, cystic, non-tender, 2 x 2 cm approximately, at the left lateral border of the anterior two-third of tongue with overlying normal tongue mucosa. The tongue movement was normal but a slight change in speech articulation was there. The differential diagnosis of mucous retention cyst, minor salivary gland tumour, epidermoidcyst, lymphangioma were made and Magnetic Resonance Imaging of tongue had been advised, but the patient did not follow-up.

The patient came after 1 month with pain and difficulty in swallowing for 2-3 days. The white cyst wall was exposed through a mucosal breach in the superior and lateral surface of tongue (Fig. 1A) and pulling the tongue laterally while examination caused more bulging. The patient was planned for surgical excision, but before it could be operated there was a sudden increase in pain and the cyst, as a whole, was expelled spontaneously (Fig. 1B). No remnant of the cyst was felt in the tongue mucosa. The cyst was round, 2 cm in diameter, white, with an area of bluish discolouration (Fig.1C).



Fig. 1.A. The white cyst wall visible on protrusion of the tongue. B. The cyst after spontaneous expulsion. C. The close-up view of the cyst.

The watery fluid was aspirated (Fig. 2A), and sent for cytological, biochemical and microbiological analysis. In wet mount, invaginated scolex of Echinococcus was seen in Haematoxylin-Eosin stain (Fig. 2B).

The histopathology of cyst wall, in Periodic Acid Schiff stain, revealed outer hyaline and inner germinal layer. Both microbiological and histopathological reports confirmed the cyst to be a hydatid cyst.

Subsequent ultrasonography of neck and whole abdomen, chest X-ray and computed tomographic scan of brain failed to reveal any residual or disseminated disease. The tongue healed in approximately 14 days and there was no recurrence even after 6 months of follow-up.

Discussion

Hydatid disease, in human, affects mostly the liver and lung (80%), but can be foundrarely in head and neck.⁴ The commonest cyst in oral cavity is mucous extravasation or retention cyst. Parasitic tongue cyst is quite unusual, cysticercosis caused by Taenia solium being the commonest.⁵ Tongue, as a location of hydatid cyst, is very rare.⁶ Only two cases of auto-expulsion of hydatid cyst of tongueare reported till date, both in India, in 1972⁷ and 2007. The present case is only the third reported case of auto-expulsion of tongue hydatid cyst, more unique in its isolated tongue involvement. The present case is only the third reported case of auto-expulsion of tongue hydatid cyst. West Bengal is not an endemic zone in India and the cyst, in location and appearance was no different from tongue mucous retention cyst. For this scenario hydatid cyst, as a differential diagnosis, was not considered initially. The suspicion came from the history of intimate contact of the child with dogs and cattle and its unique spontaneous expulsion. The diagnostic dilemma was overcome by a very simple technique of aspiration from the expelled cyst and the specific evidence of Echinococcus from the microbiological analysis.

The management plan prior to expulsion of the cyst was surgical excision under general anaesthesia. If the cyst ruptured during surgery, fatal anaphylactic reaction could have happened due to spillage of the hydatid cyst fluid over the buccal mucosa, upper respiratory tract and lung.⁸ The spontaneous expulsion itself could have been fatal,had the cyst ruptured during the process.

So, in isolated cystic lesion of tongue, hydatid disease should be considered as a differential diagnosis and during surgery, precautions should be undertaken to avoid rupture or spillage. Also the set-up for urgent general anaesthesia and supportivecare is needed even while performing surgery under local anaesthesia. In all cases of cystic tongue lesions, aspiration of cyst fluid



Fig. 2. A. Fluid being aspirated from the cyst. B. Invaginated scolex of Echinococcus (H&E, 40X)

after surgical excision and its microbiological analysis is routinely recommended along with histopathology of cyst wall to exclude any parasitic cyst.

References

- 1. Rochidi Y, Raji A, Elhattab Y. A rare localization of hydatidosis: A cervical hydatid cyst. Fr ORL 2007; 92:315-7
- Parikh F. Echinococcosis cut to cure but what about control? J Assoc Physicians India 2012; 60:9-10
- Alaparthi RK, Yelamanchili S, Nunsavathu PN, Sode U. Intraoral hydatid cyst: A rare case report. J Indian Acad Oral Med Radiol. 2015; 27;457-60

- Onerci M, Turan E, Ruacan S. Submandibular hydatid cyst (A case report). J Craniomaxillofacial Surg. 1991; 19:359-61
- Khare P, Chauhan N, Dogra R, Kala P, Chand P. Isolated cysticercosis of tongue: A case report. Diagn Cytopathol. 2014; 42(8):716-8
- 6. Kirmani MA, Sajad M, Patigaroo AR, Khan AR. Hydatid cyst of tongue. JK Practitioner 2007; 14(2):107
- Goel VP, Mehrotra TN, Bhatia BP, Gupta SN. Hydatid cyst of tongue (A case report). Journal of Indian Medical Association 1974; 63(1):28-30
- Khurana S, Das A, Malla N. Increasing trends in sero prevalence of human hydatidosis in North India: A hospital-based study. Trop Doct. 2007;37;100-2.

Venous Malformation in the Neck

Bijan Kumar Adhikary,¹ Sohag Kundu,¹ Bhaskar Ghosh,¹ Ramanuj Sinha¹

ABSTRACT

Venous malformation is a benign vascular lesion. Approximately 40% of such cases occur in head and neck. Case Report

Venous malformation in a 19 year old man is reported, which presented as a lateral neck swelling. **Discussion**

The venous malformation in the head and neck region may confuse the surgeon in distinguishing it from its other common congeners e.g. hemangioma, lymphangioma etc. and may also cause dilemma regarding its treatment modality. Treatment options include surgery, laser therapy, sclerotherapy or a combination depending upon the complexity of the lesion. **Keywords**

Vascular Malformations; Angiography, Digital Subtraction; Jugular Veins

enous malformation which comes under the broad heading of vascular malformation (Arterial, venous, lymphatic or combination) is an uncommon entity in the head neck region. They are present since birth but may manifest in later life with or without causing any symptoms.

Case Report

Introduction

A 19 year old male patient presented to the ENT outpatient department with swelling on the leftside of neck for the one year along with mild pain and numbness around the swelling. (Fig. 1) The swelling was around 4 cm x 3 cm in size, ovoid, occupying the lateral aspect of the middle third of the neck with ill-defined margin and apparently superficial to sternocleidomastoid muscle. It was soft, compressible, non-tender, not fixed to adjacent underlying or overlying structures and vertically mobile. There was neither any fluid thrill, pulsation, cystic fluctuation nor was there any change with Valsalva manoeuvre. There was not history of trauma or surgery. The temperature was not raised.

Examination of oral cavity, oropharynx and hypopharynx revealed no abnormality and no neck nodes or glands were palpable and there was no venous engorgement in the neck or around the lesion. Auscultation of the lesion did not produce any hum or bruit. USG Doppler study revealed that it was a venous malformation, but feeding vessel could not be identified.

A lobulated vascular malformation was seen in digital subtraction angiography (Fig. 2). It was opacified on venous phase after injecting the dye in left internal carotid artery.(According to USG Doppler report there was a communication with intracranial venous sinus or upper part of left internal jugular vein.)

The patient was planned for surgical intervention as he was fit for anaesthesia and it was deemed feasible to remove the mass in its entirety surgically. A 5 cm vertical incision was made along the anterior border of the sternocleidomastoid muscle so as to reach the subplatysmal plane and to expose the sternocleidomastoid muscle.

The bluish lobulated mass was seen lying over the sternocleidomastoid muscle and was communicating with the external jugular vein. Exploration medially did not reveal any feeder vessel from common facial or internal jugular vein. Ligation of the communicating external jugular vein vessel was done under the muscle and lesion was excised. Wound was closed in two layers.

1 - Department of ENT, Medical College, Kolkata

<u>Corresponding author:</u> Dr Bijan Kumar Adhikary email: drbadhikary@gmail.com



Fig.1 Clinical photograph at presentation. Arrow showing the venous swelling.

Biopsy revealed muscle fibres and fibroadipose tissue along with variable size of vascular channels lined by flattened epithelium consistent with venous malformation (Fig. 3).

Discussion

Approximately 40% of the venous malformations manifest in the head-neck region.¹ Commonly affected sites are cheek, neck, palate and tongue. Its blood flow is low as it is a post capillary lesion. Majority of the cases are sporadic and commonly occur in the air passage, oral cavity and muscle.¹ Venous malformation may be superficial or deep or it may be single or multiple. Venous malformation causes not only deformity but also other symptoms like pain, paraesthesia, ulcers, bleeding and compression or invasion of the surrounding structures. These may cause impairment of speech, swallowing, respiration or even lead to death due to bleeding or suffocation.²

Pathogenesis of venous malformation is unclear. It



Fig.2. Digital subtraction angiogram

may be due to developmental defect of venous system. TIE2 receptor mutation has been seen in venous malformation syndrome(blue rubber bleb nevus syndrome) and multiple myocutaneous and venous malformation.³

Colour of the skin or mucous membrane may be blue or purple. In our case the lesion was typically lobulated, bluish red on gross morphology and its histopathology was characteristic of venous malformation.

Pain is a common symptom in venous malformation.A significant increase in nerve cells is seen in some venous malformations.⁴ Static venous pool leads to thrombosis and results in phlebitic syndrome. Expression of matrix metaloprotinase-9 was seen in intramuscular venous malformation.⁵ Progesteron receptors are highly expressed in some cases.⁵ In our case there was localized mild pain and numbness around the non tender swelling.

There are several methods of treatment of venous malformation including surgery,laser therapy,sclerotherapy,copper needles,electrocoagulation therapy.⁶⁻⁸ There are several advantages and



Fig.3. Histopathological features (H&E, 10X)

disadvantages in each method. Treatment depends on site, size and others factors. In large venous malformations multimodality treatment is required. In our case as the lesion was of 4 cm x 3 cm in diameter and there was a distinct feeder on DSA we opted for the monomodality curative surgery where proper exposure of the lesion and ligation of the feeder suffices.

References

- 1. Buckmiller LM, Richter GT, Suen JY. Diagnosis and management of hemangiomas and vascular malformations of the head and neck. Oral Dis. 2010; 16:405-18
- Dubois J, Garel L. Imaging and therapeutic approach of hemangiomas and vascular malformations in the pediatric age group. Pediatr Radiol. 1999; 29:879-93
- Boon LM, Mulliken JB, Enjolras O, Vikkula M. Glomuvenous malformation (glomangioma) and venous malformation: distinct clinicopathologic and genetic entities. Arch Dermatol.

2004; 140:971-6

- Meijer-Jorna LB, Breugem CC, de Boer OJ, Ploegmakers JP, van der Horst CM, van der Wal AC. Presence of a distinct neural component in congenital vascular malformations relates to the histological type and location of the lesion. Hum Pathol. 2009; 40:1467-73
- Duyka LJ, Fan CY, Coviello-Malle JM, Buckmiller L, Suen JY. Progesterone receptors identified in vascular malformations of the head and neck. Otolaryngol Head Neck Surg. 2009; 141:491-5
- Lewin JS, Merkle EM, Duerk JL, Tarr RW. Low-flow vascular malformations in the head and neck: safety and feasibility of MR imaging-guided percutaneous sclerotherapy--preliminary experience with 14 procedures in three patients. Radiology 1999; 211:566-70
- Ogawa Y, Inoue K. Electrothrombosis as a treatment angioma in the face and scalp and varicosis of the leg. Plast Reconstr Surg. 1982; 70:310-8
- Li ZP. Therapeutic coagulation induced in cavernous hemangioma by use of percutaneous copper needles. Plast Reconstr Surg. 1992; 89: 613-22.

How We Do It: A DIY Smart Phone based ENT Endoscopy

Shyam Duvvi,¹ P Neelapala,² A S Duvvi,³ B Nirmal Kumar⁴

ABSTRACT

We present a simple method of preparing a smart phone based ENT endoscopy for OPD clinic and for on call ENT to provide emergency ENT care in Accident and Emergency department when required. In our experience, this method is efficient, economical and ensures mobility and flexibility of remote visual inspection and documentation. This aids in better care for patients especially where there is no easy access to traditional ENT treatment room facility. This provides a fast and costeffective solution to simultaneously view capture and save photos and videos with a smartphone and, if required, send these pictures to oncall Consultant. This enables quicker identification, documentation and support regardless of location. <u>Keywords:</u>

Smartphone; Endoscopy; Otolaryngology

S mart phone technology is becoming increasingly integrated into medical care. Recent advances and the widespread availability of smart phones have ushered in a new wave of innovations in healthcare.^{1,2,3,4.} Mobile health⁵ is a term used for the practice of medicine and public health and is supported by mobile devices, such as mobile phones, smart phones, tablet computers, and PDAs (personal digital assistants). Mobile health has emerged as a sub segment of electronic health.

The aim was to transform or use a smart phone as video endoscopy purpose. To make this happen we need to connect the smartphone's camera to the endoscope. The technical part is simple we used a modified commercial telescopic adapter to connect the smartphone.

We describe a simple technique and present our initial experience how a smartphone can be used for

1 - Department of ENT, Tapani Hospital, Rajahmundry Andhra Pradesh

2 - Department of Gyneacology, Tapani Hospital,

Rajahmundry Andhra Pradesh

3 - A Level Student, Tapani Hospital, Rajahmundry Andhra Pradesh

4 - Department of ENT, Wrightington, Wigan and Leigh NHS Trust, Wigan lane, Wigan, UK

<u>Corresponding author:</u> Dr Shyam Duvvi email: sduvvi@rediffmail.com otorhinoscopic image capturing and management, as well as its application in ENT. This method has helped in our unit where there are patients especially without easy access to traditional ENT treatment room facility in providing emergency services.

Materials and Methods

This requires a smart phone, endoscope, portable battery powered light source, a modified telescopic adaptor which is available commercially in the market to attach to smart phone for holding the endoscope (Fig. 1).

Discussion

Our aim is to introduce a convinient method to carry ENT Examination quick, simple even at the bed side without any need for sophisticated traditional equipment. This was very useful in treating patients in patients in Accident and Emergency department and dealing with cross referrals from non-ENT wards in other wards.

A survey on out-of-hours facilities in otolaryngology by Moorthy etal⁶ has shown that not all ENT units have appropriately equipped out-of-hours facilities and there is a need for nationally agreed guidelines stating the minimum equipment and assistance required to provide a safe and adequate service.



Fig. 1. ENT smart phone endoscopy unit: (A)Smart phone telescopic adaptor and its parts. (B) 1. Adaptor 2. Endocope 3. Portable light source 4. Smart phone. (C) Assembly of scope and adaptor. (D) Scope adaptor and phone as one unit. (E - F) usage of the portable Smartphone endoscope.



Fig. 2. Smart phone Oto-endoscopy pictures (A-D) (F) Mira cast/ chrome cast display on monitor

In conjunction with rigid scope the adaptor provides a fast and cost-effective solution with a smart phone in helping capturing saving photos and videos if required send these to a third party for quicker identification and documentation.

The DIY smart phone based endoscopy comprises an endoscope with quick attach-release coupling adaptor to fit for all smart phones. The coupler case acts as an endoscopic holder for the smart phone and establishes an optimal connection between the endoscope and the smart phone camera. (Fig. 1)

The quick-release coupling adaptor is modified from a commercially available telescopic adaptor by placing three diagonally based screws which will hold the endoscope to the smart phone camera surface in conjunction all the standard features offered by modern smart phones can be utilized. These include image and video recordings, image zoom, select focusing, image and data transfer via e-mail, Mira/chrome cast data



Fig. 3. Traditional endoscopy pictures using routine endoscopic camera for comparison

transfer function and much more. Moreover, various new smart phone apps (eg Pics Art, AVS Editor) are available for optimizing camera operation and image and data processing.

To maintain patient confidentiality information from the remote site about the raw data without patient personal details using a unique ID no and about the clinical condition were transmitted through the 3G network to a consultant for a primary diagnosis. With informed consent all the information gathered through this method will pass through Hospital Internet gateway mobile system and hard ware only

No further adjustment is necessary when the smartphone is combined with the endoscope. A modern smartphone with good image quality combined with the high-power and portable battery light source with good illumination will provide meaningful examination recordings. In our experience we use 13 MP (megapixel) camera with 4160 x 3120 pixels in native mode 4:3 with

179

focal length/2.0 aperture this gives reasonable good images.

In order to see the patients who are inaccessible to ENT treatment room we have prepared and used this DIY smart phone based ENT endoscopy for the management of common emergencies eg. Ear pain, Nose Bleed, Throat pain/quinsy. This method of examination is helpful mainly in stable patients rather than acutely ill or uncomfortable or anxious patients.

As per the protocol, after morning ward rounds the respective on call specialist trainee or foundation doctor can discuss with seniors and ensure that the steps taken for the patients seen on the on call were appropriate as this method has better visualization and documentation flexibility. The smart phone ENT Endoscopy Unit along with Mobile is a dedicated set. In order to avoid contamination can be cleaned easily and the endoscope sterilized and maintained in good condition with a daily record. A detailed search of literature and books on ENT surgery found no prior similar setup descriptions of this simple technique.

This method of DIY smart phone set up is very cost effective than the traditionally available adaptor from a standard equipment providers.⁷ The total cost of our unit with mobile and light source accounts to INR 16000 (USD 250 approx) the traditional standalone adaptor 7 unit itself amounts to INR 50000 (USD 780).

Conclusion

In conclusion, we present this as a simple and practical method that improves access to the basic ENT endoscopy which is now becoming a norm for examination in ensuring safe practice. This technology based instrumentation is widely available and used in Asia and Africa with cost effectiveness and helps improving patient centered approach.

Acknowledgments

To Miss Asha Sree Duvvi A level observer student for putting the kit together.

References

- Thomale UW, Knitter T, Schaumann A, Ahmadi SA, Ziegler P, Schulz M, Miethke C. Smartphone-assisted guide for the placement of ventricular catheters. Childs Nerv Syst. 2013 Jan; 29(1):131-9
- Sohn W, Shreim S, Yoon R, Huynh VB, Dash A, Clayman R, Lee HJJ. Endockscope: using mobile technology to create global point of serviceendoscopy. Endourol. 2013 Sep; 27(9):1154-60
- Avidan A, Shaylor R, Levin PD. Smartphone assisted laryngoscopy: a new technique to overcome light failure in a laryngoscope. Anesth Analg. 2013 Nov; 117(5):1262-3
- Lee M, Savage J, Dias M, Bergersen P, Winter M. Box, cable and smartphone: a simple laparoscopic trainer. Clin Teach. 2015 Dec; 12(6):384-8
- Beratarrechea A, Lee AG, Willner JM, Jahangir E, Ciapponi A, Rubinstein A. The Impact of Mobile Health Interventions on Chronic Disease Outcomes in Developing Countries: A Systematic Review. Telemed J E Health. 2014 Jan 1; 20(1): 75–82
- Moorthy R, Magarey M, Joshi A, Jayaraj SM, Clarke PM. A study of out-of-hours facilities in otolaryngology: current provision and problems. J Laryngol Otol. 2005 Mar; 119(3):202-6
- KARL STORZ SMART SCOPE.Optimal connection between endoscopes and smartphones. https://www.karlstorz.com/fj/en/ karl-storz-smart-scope.htm.

Book Review



Title: Endoscopic Ear Surgery - A New Horizon Author: Dr Saurav Sarkar ISBN-10 9385891626 ISBN-13 978-9385891625 First Edition, 2016 Pages: 96 Publisher: Jaypee Brothers Medical Publishers (P) Ltd. New Delhi, India

Major advances in the field of medical science have always come with pioneers trying to do things differently and the recent endeavours of Dr Saurav Sarkar are certainly a step in the right direction. Using the technological advancement of modern endoscopes in the field of otology, in this pioneering book, he introduces new and experienced otologists to the concepts of endoscopic ear surgery. Clear, concise and profusely illustrated, this book will take surgeons interested in this new aspect of ear surgery through the entire process - from the setup of equipment to the gamut of commonly performed otoendoscopic ear surgeries - with aplomb.

The book is divided into convenient sections - a brief explanation about the concept of *Functional Endoscopic Ear Surgery* is followed by detailed instructions for setup of equipment and a vivid description of endoscopic anatomy of the ear. These are followed by a section on otoendoscopic procedures divided for convenience into two subsections comprising office-based procedures and commonly performed major ear surgeries. The book is rounded off with a honest comparison between the endoscopic method and the conventional microscopic method and a small section to aspire other enthusiasts for *'The Road Ahead.'*

The standout features of this book include the very lucid language, the plethora of relevant illustrations and a layout which grabs the otologist's interest from the very first page and keeps him submerged till the last page is turned. The organisation of the book helps in quick revision after the first read for the busy surgeons. This book would certainly be of immense help for anyone willing to venture into this new horizon for otologists.

Reviewed by: Dr Swagatam Banerjee

Bengal Journal of Otolaryngology and Head Neck Surgery

Published by: The Association of Otolaryngologists of India, West Bengal CMC House, 91B Chittaranjan Avenue, Kolkata - 700073