

Research Article**A Study on Clinical Profile of Non-Otogenic Otolgia****Dr. Prashant Manohar Dukare****Assistant Professor, Department of ENT, Dr. Ulhas Patil Medical College & Hospital, Jalgaon
Kh****ABSTRACT**

Background: The ear is unique in that no other structure in the body of comparable size has such a unique sensory nerve supply. The sensory nerve supply of the ear is through a combination of four cranial nerves (Cranial nerves V, VII, IX, and X) and two superior cervical plexus nerves (C2 and C3). Presumably, this complex innervation is an excellent survival tool, and any pain perceived in that area causes a heightened sense of alarm. The differential diagnosis is specifically related to the sensory innervations of the ear, and therefore it is required that otolaryngologists have relevant knowledge of the complex neuroanatomic innervation of the external and middle ear. Among all causes of referred otalgia, dental pathology, which transmits referred pain via a branch of the trigeminal nerve, is the most common source of non-otogenic pain. Ear pain can be because of pathologies in the ear or in the surrounding head and neck region. As there is no single, simple algorithm for determining the cause of otalgia and due to complex innervation of the ear frequently the clinicians get puzzled and the cause remains obscure.

Aim: The aim of the study was to identify the various possible non-otogenic causes and their various characteristics which will help in better evaluation of patients.

Material and Method: This study was a cross-sectional study on patients who had presented to the ENT department. All patients over five years of age, both male and female and presenting with earache were selected. Patients having aural pathology to account for pain, having previous surgery on the affected ear, or having hemifacial pain or vague sensation of aural fullness were excluded from the analysis. A total of 150 patients presenting with earache were examined. The procedure was explained and Informed consent was obtained. Detailed history taking & clinical examination ensued. Laboratory investigations, imaging studies, and endoscopic examinations were carried out as and when indicated.

Results: There were 150 patients who presented with earache in our ENT OPD. Out of 150 patients, 70 patients had definite otogenic causes of otalgia and they were excluded from the study. The remaining 80 patients were evaluated and were included in the present study. The most common non-otogenic cause was found to be temporomandibular dysfunction (41.2%). Other causes in descending order of frequency were cervical spondylosis (20.0%), dental caries (10.0%), pharyngitis (6.25%), Eagle's syndrome (6.25%), impacted molar (5.0%), carcinoma oropharynx (5.0%), acute tonsillitis, peri tonsillitis, glossopharyngeal neuralgia, Bell's palsy psychogenic (1.25%). There were 2 cases of malignancies, one along the posterior third tongue other along the tonsil lingual sulcus which was biopsied and confirmed histologically.

Conclusion: In this study, the most common cause is odontogenic followed by cervical lesions. Sometimes an isolated ear pain could be the telltale sign of an early upper aero digestive tract malignancy. Neuralgic pain can also present with ear pain. So, in patients presenting with ear pain examination of dentition and cervical spine should be done in addition to the routine ear, nose, throat, and neck examination. Most cases are presented in the 5th and 6th decade. There are no many differences in presentation in both sexes and laterality.

Keywords: Referred otalgia, Temporomandibular joint dysfunction, Cervical spondylosis.

Key words: CML, GIST, PDGFRA

Introduction:

Otalgia is a complaint of all ages, which is due to complex neuroanatomic innervation of the ear, head, and neck and can be classified as otogenic (primary) or non-otogenic (referred).^{1,2} External and middle ear infections are associated with primary otogenic pain, which most otolaryngologists and primary care physicians are trained to diagnose.³ Common disease processes resulting in primary otalgia include otomastoiditis, cholesteatoma, and foreign bodies lodged within the ear canal. In close to 50% of cases, however, the source of the pain does not reside within the ear. Rather, originates from sources distant from the ear, so-called "referred otalgia".⁴ Otalgia is a very common presentation in the ENT outpatient department. Most of the time the underlying cause is from the ear which is referred to as primary otalgia. There are quite a number of causes that can present as otalgia and the ear looks normal in otoscopy. These are called secondary or referred otalgia.^{1,2} This is because of complex nerve innervations of the ear.

The pain is caused by nerve compression or irritation. In referred pain, the sensation of pain will generally be felt in the somatic dermatome even though the stimuli are from the visceral tissue.⁵ The ear is supplied by four cranial nerves (CN V, VII, IX, and X) and two superior cervical plexus nerves (C2 and C3).⁶ These nerves also supply a lot of structures in the head and neck. This rich innervation of the ear explains the central misinterpretation of the origin of pain arising from the head and neck and is the basis for referred otalgia. In otalgia with negative findings on otoscopic and CT examination of the temporal bone, it is imperative that distant sources within the head & neck be evaluated. This may include inspection with or without the use of flexible endoscopes and palpation of the accessible areas. CT and MR imaging are excellent tools to investigate further regions of the head and neck that are not easily accessible to physical examination.⁶

The origins of referred otalgia may include the cranial cavity to thorax but dental diseases, tonsillitis, temporomandibular joint, and cervical spine disorders are the commonest sites.⁷ Referred otalgia may be the first symptom of a head and neck malignancy. Patients with risk factors for upper aerodigestive tract malignancies and normal ear examination require further evaluation.^{8,9}

The differential diagnosis is specifically related to the sensory innervations of the ear, and therefore it is required that otolaryngologists have the relevant knowledge of the complex neuroanatomic innervation of the external and middle ear. Among all causes of referred otalgia, dental pathology, which transmits referred pain via a branch of the trigeminal nerve, is the most common source of non-otogenic pain.¹⁰ Furthermore, the cause of referred otalgia can also be referred pain from the mouth, teeth, larynx, or thyroid gland; neural, vascular, or lymphatic structures of the neck; or the esophagus.^{11,12} However, with the aging population, physicians must also consider cervical spine degenerative disease (CSDD) as an increasingly common cause of referred otalgia involving the upper cervical plexus (greater auricular and lesser occipital nerve). Cervical spine degenerative disease (CSDD) includes osteoarthritis, cervical facet syndrome, spondylosis, disc herniation, and stenosis. According to several authors, otogenic otalgia is more frequent than reflex otalgia and also appears to be more frequent in children than in adults.^{13,14} Their etiology does not seem to differ according to gender.¹³ As there is no single, simple algorithm for determining the cause of otalgia and due to complex innervation of the ear the clinicians get puzzled and the cause remains obscure. The late diagnosis in some cases e.g., malignancy, can lead to life-threatening conditions. Sometimes it could be psychogenic or malingering where no organic lesions could be made out. The purpose of the study is to identify the various possible non-otogenic causes and their various characteristics which will help in better evaluation of patients.

Material and Methods

This study was a cross-sectional study on patients who had presented to the ENT department. All patients over five years of age, both male and female and presenting with earache were selected. Patients having aural pathology to account for pain, having previous surgery on the affected ear, or having hemifacial pain or vague sensation of aural fullness were excluded from the analysis. A total of 150 patients presenting with earache were examined. The procedure was explained and Informed consent was obtained. Detailed history taking & clinical examination ensued. Laboratory investigations, imaging studies, and endoscopic examinations were carried out as and when indicated. The final diagnosis of referred otalgia was made on positive findings in the other head & neck sites that share sensory innervation with the ear in conjunction with a "Normal on Examination" affected ear. Only 80 patients were diagnosed as having referred otalgia. The distribution of various etiologies in these patients was categorized and tabulated. The data regarding the age, sex distribution, laterality, and the nerves responsible in relation to various etiologies were collected and tabulated.

A thorough ear, nose, and throat examination was carried out on every patient. Apart from routine ENT examinations the temporomandibular joint and dentition were also done. In relevant cases, the opinion of dental, and skin consultants was also taken. Radiological investigations, fiber optic laryngoscopy, diagnostic nasal endoscopy, and biopsy were also done to reach the diagnosis. All patients were interviewed to characterize their otalgia and to get their medical history. An ENT and general physical examination were performed on all patients. Otoscopy was systematically performed using an ENT microscope (Leica brand), which examined the auricle, the mastoid region, the external auditory canal, and the tympanic membrane. Acoumetry was performed using a 512 Hz tuning fork for a brief assessment of hearing. The rest of the examination included the oral

and oropharyngeal cavity, anterior rhinoscopy with a headlamp, examination of the skin of the neck and face, and examination of the cranial nerves. Tympanometry completed the examination in the case of tympanic membrane remodeling without perforation. Audiometry was indicated in case of suspected hearing loss.

The patients underwent thorough examinations covering the ear, the teeth, the temporomandibular joint, the nose, the sinuses, and head and neck areas, and when necessary, examinations included direct and indirect laryngoscopy and biopsy. Patients without an ear, nose, and throat disease were referred to other departments for a definitive diagnosis. Those participants included in the study who had presented with complaints of ear pain due to any underlying cause while those who were excluded who either didn't give consent had a language barrier or had concurrent illnesses that can affect the quality of life like severe heart or lung disease, etc. The questions included were about ear pain, its duration, involving the site, vertigo, the extent of hearing loss, the patient's functional status, and behavioral changes. The distributions of all diagnosed etiologic factors for referred otalgia with their respective nerve pathways were categorized. The data thus collected was tabulated and statistically analyzed.

Statistical Analysis

The data was recorded on a proforma and the descriptive statistics were analyzed to determine frequencies for variables like gender, age, and type of otalgia. Percentages were calculated for the number of times a distant site was implicated in referred otalgia in relation to various age groups and genders. The chi-square test was applied to determine the significance of the involvement of distant sites with respect to the variables; age groups and gender of patients. The analysis was carried out using SPSS 16 for Windows.

Result: -

There were 150 patients who presented with earache in our ENT OPD. Out of 150 patients, 70 patients had definite otogenic causes of

otalgia and they were excluded from the study. The remaining 80 patients were evaluated and

were included in the present study.

Table 1: Causes of non-otogenic otalgia.

Etiology	No. of cases (n)	Percentage (%)
Temporomandibular joint dysfunction	33	41.2
Dental caries	8	10.0
Impacted molar	4	5.0
Cervical spondylosis	16	20.0
Pharyngitis	5	6.25
Acute tonsillitis	1	1.25
Peri-tonsillitis	1	1.25
Eagle's syndrome	5	6.25
Glossopharyngeal neuralgia	1	1.25
Bell's palsy	1	1.25
Carcinoma oropharynx	4	5.0
Psychogenic	1	1.25

The most common non-otogenic cause was found to be temporomandibular dysfunction (41.2%). Other causes in descending order of frequency were cervical spondylosis (20.0%), dental caries (10.0%), pharyngitis (6.25%), Eagle's syndrome (6.25%), impacted molar (5.0%), carcinoma oropharynx (5.0%), acute

tonsillitis, peri tonsillitis, glossopharyngeal neuralgia, Bell's palsy psychogenic (1.25%). There were 2 cases of malignancies, one along the posterior third tongue other along the tonsil lingual sulcus which was biopsied and confirmed histologically. Both cases had presented with ear pain.

Table 2: Distribution of non-otogenic otalgia in both sexes.

Etiology	Males	%	Females	%
Temporomandibular joint dysfunction	21	46.6	15	42.85
Dental caries	6	13.3	4	11.4
Impacted molar	0	0	2	5.7
Cervical spondylosis	10	22.2	7	20
Pharyngitis	2	4.4	2	5.7
Acute tonsillitis	2	4.4	0	0
Peri tonsillitis	0		1	2.85
Eagle's syndrome	4	8.8	1	2.85
Glossopharyngeal neuralgia	0	0	1	2.85
Bell's palsy	0	0	1	2.85
Carcinoma oropharynx	4	8.8	0	0
Psychogenic	0	0	1	2.85
Total	45	0	35	

There were no sex differences in etiology. In both sexes, the most common cause was TMJ dysfunction, followed by cervical lesions.

Table 3: Distribution of non-otogenic otalgia in terms of laterality

Etiology	Bilateral	Unilateral-	Unilateral-
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		right	left
Temporomandibular joint dysfunction	9	10	12
Dental caries	0	4	6
Impacted molar	0	2	1
Cervical spondylosis	4	7	6
Pharyngitis	2	3	0
Acute tonsillitis	0	0	1
Peri tonsillitis	0	0	1
Eagle's syndrome	2	3	0
Glossopharyngeal neuralgia	0	0	1
Bell's palsy	0	0	1
Carcinoma oropharynx	0	2	1
Psychogenic	2	0	0
Total	19	31	30

Most patients had unilateral ear pain. There was no predilection for any side. Only 12 patients had pain in both the ears.

Discussion

Primary ear pain is relatively simple to diagnose. However, the ear is not always the source of earache. It becomes a diagnostic dilemma to identify the source of pain when the ear is found normal on examination. A number of nerves that supply other head and neck structures also supply the ear. A variety of pathological conditions ranging from inflammation to neoplasia can cause irritation of these sensory nerves and cause pain radiating to the ear by virtue of their shared sensory innervation. The national literature on this particular subject is scarce and the majority of the studies are focused on the etiology of primary otogenic pain. The negative impact of the symptomatology on the quality of life (sleep disorders, headaches, poor emotional balance) often compels patients to seek help. In this study, the length of the medical consultation time is quite long because of widespread self-medication in our environment such as using ear drops and the inaccessibility of patients to specialized health structures.¹⁵

Chen et al.2009⁶ and **Vedasalam et al.2010¹⁶** Higher incidence in females, in our study, could be due to their hectic lifestyle. Most

females in the local population around the hospital are hardcore workers, working as laborers and construction workers. Maybe the cervical spine of females isn't meant to handle this kind of stress. In most studies, elderly age groups have had an increased incidence of cervical spine degenerative disease, but in our study, less than 50 years of age group was also equally affected. Yet again, this may be because of the lifestyle of the local population, amongst whom; many are laborers (construction workers) and farmers. The analysis of the aetiologies of otalgia according to gender in our study showed no significant difference between males and females. On the contrary, **Kim et al.2015¹⁴** revealed in their study that otogenic otalgia was more frequent in men, while reflex otalgia was more frequent in women. Their argument was that males may be more likely than females to bite off their ears, which exposes them to more ear diseases, which is not observed in our daily practice. The main symptoms associated with otalgia in this study were auricular (deafness, ear fullness, otorrhoea, auricular pruritus, and tinnitus), which may explain the high frequency of otodynia.

Referred otalgia has been a well-documented phenomenon in the ear, nose, and throat (ENT) and neurosurgical literature. An understanding of the various sensorineural pathways that dually innervate the ear and other sites and

ongoing dialogue with our clinical colleagues will ensure that patients with referred otalgia will receive the best care. Cervical spine degenerative disease referred to as otalgia is propagated through disease of the cervical vertebrae and therefore is postural. This ear pain can be ameliorated with physical therapy, and most studies support conservative treatment, such as cervicothoracic stabilization programs, combined with aerobic conditioning in treating cervical spine disorders.¹⁷ Referred otalgia due to cervical spine disease usually is described as retro-auricular or infra-auricular pain, which is constant and often related to changes in neck position. In this current study, we have shown that cervical spine degenerative diseases, diagnosed either through radiographic or clinical evidence, are an important contributor to referred otalgia. Cervical spine degenerative disease referred to as otalgia represented 26.6% of referred otalgia cases. In the study by **Han DG et al.2010**¹⁸ **Kiakojoori et al.2002**¹⁹ and **Kim et al.2015**¹⁴ toothaches were the most common cause of the referred otalgia. In the **Behnoud et al.2000**²⁰ study, the most frequent etiology was reported to be the temporomandibular joint dysfunction. In the study by **Mulwafu et al.2006**²¹ on 17 patients suffering from carcinoma of the base of the tongue, 33% of the cases suffered from referred otalgia. In **Kiakojoori et al.2002**¹⁹ study, 6% of the etiologies of referred otalgia were reported as pharyngeal carcinoma.

In the US, **Jaber et al.2008**²² studied 123 patients presenting with referred otalgia. The mean age of the patients was 63.5 years. They found that 84% of their patients with referred ear pain had some form of cervical spine pathology. They concluded that cervical spine degenerative disorders constitute important causes of referred otalgia in the elderly. In a Korean study of 294 patients with otalgia, the frequency of primary otalgia was found to be higher in children than in adults and in men than in women. Similarly, otalgia of “unknown origin” was found to be more common in females than in males.¹⁴ The high index of suspicion should be borne in mind to diagnose referred otalgia due to cervical spine

degenerative disease in younger to middle age groups, without which, many such cases could be misdiagnosed or undiagnosed. It now seems that occupational and lifestyle factors have an equally important role to play in comparison to aging factors, in determining the occurrence of cervical spine degenerative disease. Therefore, more research needs to be done to analyze the significance of these factors. Earache is a frequent reason for medical consultation at any age and for any gender. However, ear infections are more frequent in children. Reflex otalgia predominates in adults due to pharyngeal infections, trigeminal neuralgia and oral cavity, and pharyngeal and laryngeal cancers. The causes of otalgia do not differ by gender. Certain practices, such as the use of cotton swabs, seem to favor ear infections.

Conclusion:

In this study, the most common cause is odontogenic followed by cervical lesions. Sometimes an isolated ear pain could be the telltale sign of an early upper aero digestive tract malignancy. Neuralgic pain can also present with ear pain. So, in patients presenting with ear pain examination of dentition and cervical spine should be done in addition to the routine ear, nose, throat, and neck examination. Most cases were presented in the 5th and 6th decade. There are no many differences in presentation in both sexes and laterality. For the patient of otalgia in the absence of primary pathology, one should exercise a thorough examination of the oral cavity, oropharynx, nose, and throat. In young adults, the referred otalgia is mainly due to infective causes but whenever evaluating an older patient one should not miss any occult malignancy of the aerodigestive tract which often has a tendency to present as an isolated otalgia.

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