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Case Report

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Referred otalgia secondary to rotator cuff tendon tear and acromioclavicular joint osteoarthritis: A Case Report and Review of Literature

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Abstract

***Background*:** Otalgia is a very common symptom induced by a wide variety of diseases. Depending on the location of the pathology, otalgia is categorized into primary and secondary types. In primary otalgia, the main source of the pain is located within the ear. On the contrary, secondary or referred otalgia is defined as a disorder which the underlying pathology is outside of the ear complex giving rise to the sensation of pain in the ear. Multiple etiologies have been reported to produce secondary otalgia including temporomandibular joint dysfunctions, cervical spine pathologies, laryngitis, pharyngitis and tonsillitis.

Case Presentation: In this study, we present a 57-year-old man with referred otalgia, associated to rotator cuff tendon tear and acromioclavicular joint osteoarthritis, which is uncommon and atypical.

Conclusion: Shoulder pathologies like rotator cuff tendon tear and acromioclavicular joint osteoarthritis can be considered as a possible but rare cause of referred otalgia

***Keywords*:** Acromioclavicular Joint, Otalgia, Rotator Cuff

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Introduction

Ear pain medically termed otalgia is a common complaint in patients referring to both general practice and otorhinolaryngology clinics with approximately 100% lifetime prevalence of all ages, especially among children(1, 2). Ear structures can be divided into three parts including external, middle and inner ear(3). The auricle supplied by intricate sensory innervation system including multiple cranial nerves (cranial nerves V, VII, IX, X) as well as C2 and C3 nerve roots of the cervical plexus(4). In literature two types of otalgia has been defined: primary and secondary otalgia. In primary otalgia, the pathology responsible for producing pain is localized within the ear which is mostly infective and usually this otogenic cause of the ear pain can be diagnosed by accurate physical examination and otoscopy (5, 6). Otitis media, external otitis, cerumen impaction, tympanic membrane inflammation,

eustachian tube dysfunction, cholesteotoma, otomastoiditis, folliculitis, foreign body lodgment and neoplasms are common causes of primary otalgia(7, 8). But in secondary otalgia the source of the lesion is not located within ear anatomical structures. Therefore, otoscopy and physical examination fail to reveal the underlying pathology and warrants cross-sectional imaging like CT-scan and MRI for better investigation; consequently this phenomenon can be considered "referred or non-otogenic otalgia"(9, 10). Up to 50% of cases with otlagia are considered as non-otogenic type (8, 11). In this case report, we observed such uncommon and unorthodox pattern of referred otalgia secondary to rotator cuff tendon tear and acromioclavicular joint (ACJ) osteoarthritis which had not been reported in previous studies.

Case Presentation

A 57-year-old man with a chief complaint of non-traumatic gradual onset left shoulder pain from one year before, was referred to the rehabilitation ward for physiotherapy treatment. He had past medical history of coronary heart disease and blood hyperlipidemia. He was recreational climber. At the beginning of the pain course, the patient reported intermittent motion-induced localized shoulder pain mostly with overhead arm activities. Prior to commencing any physiotherapy intervention, clinical examination of the shoulder complex was done. The examination showed that the patient's pain was provoked at the end range of the active and passive glenohumeral elevation and internal rotation motions. In addition, the elicited positive results from the cross-body adduction test , empty can test and Hawkins-Kennedy test accompanied by the detection of multiple point tenderness at palpation of the acromion process of the scapula and at the lesser and greater tuberosity of the humeral head were discovered in favor of ACJ osteoarthritis and subacromial impingement syndrome. Furthermore, an examination of the cervical spine was done, revealing painless passive and active range of motion tests for cervical flexion, extension, rotation and sideflexion without any significant limitation of motions. Also cervical foraminal compression and distraction tests were found negative, ruling out any cervical radiculopathy. In addition, the deep tendon reflexes of the left upper limb were found normal. Also the patient did not report any feeling of paresthesia, tingling or numbness or any other nerve-related symptoms. Abnormal imaging findings were detected in his shoulder X-ray including ACJ osteophyte formation, decreased acromiohumeral interval and greater tuberosity sclerosis. The patient then underwent physiotherapy treatment protocol, including transcutaneous electrical nerve stimulation, thermotherapy, ultrasound therapy, muscle strengthening and stretching exercises as well as myofascial trigger point dry needling of shoulder complex musculatures during ten sessions. Despite taking NSAID medications and ten-session-physiotherapy intervention, the recorded clinical outcomes were poor and the treatment efficacy was considered unsuccessful in subsequent follow ups. So he was referred back to his orthopedic physician for ordering next plan of care. According to the failure of conservative treatment, his orthopedic physician decided to perform surgical treatment for him. In the last two months prior to his shoulder surgery, the patient's symptoms exacerbated and the pain behavior changed; it becomes such a deep persistent severe shoulder pain felt even at rest especially at night with referral pain pattern to the ipsilateral neck and ear with self-reported numeric pain rate scale of 7/10 in day and 10/10 at night leading to sleep deprivation. At that time, the patient asserted such a severe left side otalgia with a feeling of impaired hearing sensation at some episodes which made him doing home remedies to manage his earache. Then consultation with an expert otorhinolaryngologist was requested for better ear assessment; Focused ear examination, palpation and otoscopy were done but all these ear tests were normal without indicating any definite pathology in the left ear. As a result, he was labeled as referred otalgia. Even though, Betamethasone ear drop was prescribed for probable pain reduction but it was completely irresponsive. His simultaneous shoulder pain and persistent left side earache existed continually until the day of shoulder surgery even felt at

operation room. Remarkably the patient asserted that his otalgia reduced significantly just at the night immediately after undertaking rotator cuff tendon repair and acromioplasty procedures at the hospital. Furthermore, the ear pain removed completely one day after surgery. In the next multiple follow-ups, the patient reported no feeling of otalgia at all without any recurrence.

Discussion

Previous studies showed that the referred otalgia is mainly seen in adult population in contrast to primary otalgia which is highly prevalent among children(12, 13). This premise supports the finding of our study. In order to explore the different causes of the referred otalgia including common and uncommon etiologies, a non-systematic literature review was conducted. The results of our review revealed that a wide spectrum of diseases can lead to non-otogenic ear pain including:

1. Odontogenic factors consist of dental diseases and abscess like caries, pulpitis, periodontitis and impacted third molars can lead to referred otalgia by means of involving auriculotemporal branch of trigeminal nerve(14-16).

2. Temporomandibular joint dysfunction or masticatory muscles disorder accompanied by symptoms like tinnitus and vertigo mostly secondary to malocclusion and bruxism may result in non-otogenic otalgia especially in women by means of involving auriculotemporal branch of trigeminal nerve(14, 17, 18).

3. Various sinonasal factors including rhinosinusitis, malignancies and surgeries of nasal cavity and paranasal sinuses can lead to referred otalgia due to involvement of trigeminal nerve(19, 20).

4. Neuralgia of different nerves like trigeminal, geniculate, vagal, sphenopalatine, glossopharyngeal and occipital neuralgias may present referred otalgia(21-25)

5. Facial nerve pathologies like Bell's palsy and herpes zoster oticus known as Ramsey Hunt Syndrome result in facial muscles paralysis or weakness can lead to non-otogenic ear pain. This is mostly characterized by retro auricular pain secondary to acute viral infections involving geniculate gangelion or posterior auricular branch of facial nerve(19).

6. Upper aerodigestive tract diseases like pharyngitis, laryngitis, tonsillitis, peritonsillar abscess, post- operative tonsillectomy and benign or malignant neoplasms can result in referred otalgia accompanied by symptoms like odynophagia, dysphagia or hoarseness as a result of involving Jacobson's nerve derivative from glossopharyngeal nerve(26-28).

7. Styloid ligament calcification known as Eagle's Syndrome lead to referred otalgia according to elongation of the styloid process by irritating the glossopharyngeal nerve(6, 29).

8. Cervical spine pathologies like zygapophyseal joints degenerative diseases, atlanto-occipital joint dysfunction, radiculopathy, whiplash injury or cervical tumors can present non-otogenic otalgia due to affecting C2 and C3 spinal nerve roots named greater auricular and lesser occipital nerves(30-33).

9. Fibromyalgia, rheumatic diseases, myofascial pain syndromes and psychological disorders may lead to referred otalgia(19, 34, 35).

10. Temporal arteritis with associated symptoms like headache, scalp tenderness and jaw claudication may present non-otogenic otalgia(36).

11. Gastroesophageal reflux disease, thyroiditis or even myocardial ischemia may result in referred otagia according to involvement of Arnold branch of vagus nerve(37-39).

It is essential to mention that sensory and motor innervations of shoulder complex are made from brachial plexus originating from C5-T1 nerve roots which are completely different from anatomical territories covered by cranial nerves and cervical plexus providing sensory innervation of the ear. In addition, the accessory nerve (cranial nerve XI) innervates the trapezius and sternocleidomastoid muscles considered as synergistic shoulder mobilizers which is again distinct from cranial nerves providing sensory innervation of the ear(40). Therefore, the sensory and motor nerves of the shoulder region do not coincidence or overlap with sensory innervation of the ear complex.

Conclusion

There is no scientific finding in the literature considering shoulder pathologies, such as rotator cuff tendon tears or acromioclavicular joint osteoarthritis, as a source of non-otogenic ear pain. Although earache induced by lesions of the shoulder joint and tendon is unusual and unorthodox, it can be considered as a possible source of referred otalgia.

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Ethical statement

The content of this manuscript is in accordance with the declaration of Helsinki for Ethics. No committee approval was required. Oral and written informed consent to participate were taken from the patient.

Consent for Publication

Written informed consent was obtained from the patient for publication of this case report**.**

Data availability

The raw data supporting the conclusions of this article are available from the authors upon reasonable request.

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Authors Contribution:

Milad Bahari: Patient's physiotherapist and author-
Fariba Jafari: author

Conflict of interest

The authors have no conflict of interest in this study.

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