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Contents

Editorial

- Challenges and Considerations in Medical Journal Publication 5-6
Dr. Md. Matiur Rahman, Md. Kaoser Bin Siddique

Original Article

- Health Problems and Growth Status of Primary School Children in an Upazilla of Bangladesh 7-12
Dr. Md. Azfarul Habib, Dr. Tahsina Nasreen, Dr. Kutbul Zannat, Dr. S.M. Mejba Ul Hoque
Dr. Abdullah Al Mahfuz Ansari, Dr. Hafiza Arzuman
- Platelet count: A Possible Marker for Severity of Community Acquired Pneumonia 13-19
Dr. Md. Shahidul Islam, Dr. Md. Tarikul Islam, Dr. Umme Kusum
- MRI Evaluation of Suspected Cases of Trigeminal Neuralgia 21-27
Dr. Md. Mofazzal Sharif, Dr. Swapna Rani Mondal, Dr. Rezaul Ameen Ferdousy, Dr. Md. Azizul Haque
Dr. Md. Mahbubur Rahman, Dr. Md. Fazlul Haque
- The Pattern of Skin Diseases in Patients Attending OPD of Dermatology and Venereology at TMSS Medical College and Rafatullah Community Hospital, Bogura, Bangladesh 29-35
Dr. Md. Siddiqur Rahman, Dr. Selina Sultana, Dr. Renu Gupta Joyaa, Dr. Sukanta Kumar Ghose
Dr. Shafwanur Rahman

Case Report

- A Case Report on Eagle's Syndrome: A Rare Cause of Oro-facial Pain 37-40
Dr. Md Amirul Islam, Dr. Dipshikha Maharjan
- Gastrointestinal Stromal Tumours in Stomach - A Cause of Occult Anaemia 41-44
Dr. ATM Mostafizur Raman, Dr. Md. Abdul Mazid



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Case Report

A Case Report on Eagle's Syndrome: A Rare Cause of Oro-facial Pain

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Abstract

Eagle's Syndrome, sometimes called as Styloid or Stylohyoid Syndrome, is a symptom complex associated with the elongation of the styloid process or ossification of stylohyoid ligament, clinically characterized by dull and nagging pain in the throat and neck that becomes worse during deglutition and can be reproduced by palpation of the tonsillar fossa. Eagle's Syndrome represents clinical characteristics as non-specific orofacial pain, unilateral, not responsive to painkillers and mostly in adult women. The diagnosis must be made through a detailed history, clinical examination and various imaging modalities (computed tomography with multiplane reconstruction and 3D volumetric reconstructions). This paper discussed a case of lower left facial pain for about 6 months, in a 40 years old, female patient who was diagnosed as unilateral elongated styloid process.

Keywords: Eagle's Syndrome, Elongation of Styloid process, Facial pain.

Introduction

The styloid process of the temporal bone is elongated, conical projection that lies anterior to the mastoid process of temporal bone in the neck between the internal and external carotid arteries and is lateral to the tonsillar fossa.¹ Watt Weems Eagle, 1937, an American Otolaryngologist, was the first person to describe Eagle's syndrome (ES) or Stylohyoid Syndrome as an abnormally long styloid process or styloid chain ossification producing cluster of symptoms, characterized by craniofacial or cervical pain.^{2,3,4,5} The normal styloid process is approximately 2.5-3.0cm in length.⁶ An elongated styloid process is incidental in about 4% of the general population, but of these, only about 4% present with symptoms that are attributable to elongation of styloid; therefore, the true incidence is about 0.16%, with a female-to-male predominance of 3:1. Patients are usually greater than 30 years of age, and this is usually a bilateral process (although unilateral cases are also seen).⁷ The syndrome is usually reported in adults after third decade of life with a female to male ratio of 3:1.⁸ Patient with ES may present with wide variety of symptoms including dull, aching pain on either side of the throat, difficult in swallowing, foreign body sensation in the throat, pain in facial region, cervical pain, otalgia, cervico-facial pain, recurrent headache,

vertigo.⁹ However, the characteristic dull and nagging pain that becomes worsen during deglutition and can be reproduced by palpation of the tonsillar fossa is the hallmark.¹¹ Since these symptoms mimic many maxillofacial and oropharyngeal disorders and neuralgias, a thorough clinical history, examination, and radiological assessment are necessary for attaining a diagnosis. Here, we present one such case of Eagle's syndrome in a female patient, explain the diagnosis and mentioned plan of management.

Case Presentation

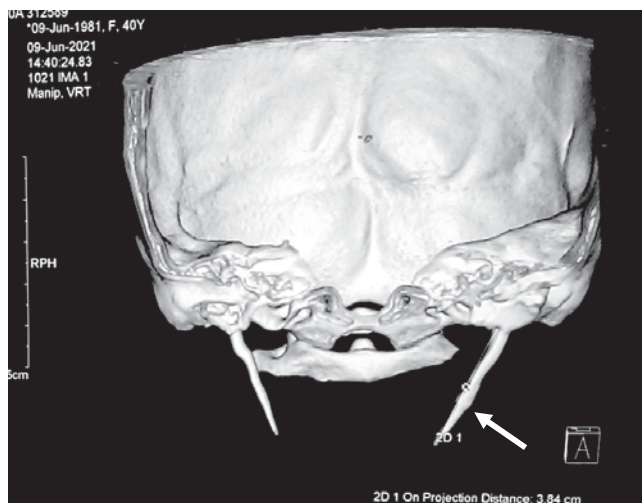
A 40 years old female patient reported to Oral and Maxillofacial Surgery Department of Rafatullah Community Hospital, complained of pain in the left side of the lower jaw for more than 6 months. The nature of pain was dull to moderate intensity, was spontaneous and intermittent in nature. The pain aggravates on opening of the mouth, chewing during swallowing, movement of head and neck from side to side. Her medical history revealed Asthma and Arthritis on her spine under medication.

On extra-oral examination, facial symmetry was symmetrical. There was tenderness present over the left angle of the face with palpable elongated left styloid process. There was no tenderness over the muscles of

mastication, no tenderness was elicited in the temporomandibular joints during mandibular movement. On intra-oral examination, impacted third molar on lower left jaw was present, elevated elongated styloid process was palpated at tonsillar fossa with tenderness present on palpation.

Based on clinical examination, a provisional diagnosis of impacted mandibular third molar on left side with inflammatory changes was made and differential diagnosis of styloid process was kept. After proper history taking and clinical examination, radiological investigation, three dimensional CT scan of the left mid and lower face with 3D reconstruction with special attention to the left styloid process was given for analysis.

CT scan of Maxillofacial region revealed enlarged styloid process measuring 3.84 cm on left side. Treatment plan is decided as reduction of the left styloid process by extra-oral approach and surgical extraction of impacted lower left third molar.



CT scan of Maxillofacial region with 3D reconstruction revealing enlarged styloid process measuring 3.84cm on left side (red arrow).

Discussion

Eagle's Syndrome, sometimes called styloid or stylohyoid syndrome, is defined as the symptomatic elongation of the styloid process or mineralization (ossification or calcification) of the stylohyoid ligament complex.^{11,5} Over a twenty years' period,

Eagle reported over 200 cases and explained that the normal styloid process is approximately 2.5–3.0 cm in length. He observed that even slight medial deviation of the styloid process, could result in severe symptoms of atypical facial pain.^{12,6} Eagle classified two forms of the syndrome, namely, classic type and carotid artery type. In the classic type, i.e. after tonsillectomy where the pain may occur when scar tissue underneath the tonsillar fossa compress the V, VII, IX and X cranial nerves, causing pharyngeal pain localized in the tonsillar fossa that sometimes radiate to the hyoid bone, discomfort and difficulty in swallowing and a feeling of having a foreign object in the throat. The second type, carotid artery type which is symptomatically characterized by persistent pain caused by impingement of styloid process on the carotid territory, headache and nerve problem due to the inflammation of the sympathetic nerve plexus.^{13, 14}

The characteristic dull and nagging pain of an elongated styloid process that becomes worse during deglutition and can be reproduced by palpation of the tonsillar fossa is the hallmark. Duration of patients' symptoms ranged from 8 months to 37 months (mean 14 months), no significant sex predilection in the occurrence of mineralization of the styloid process. However, symptoms are more common in females and patients are usually older than 30 years and rarely younger.¹⁵ The pathogenesis is still in debate but several theories have been purposed which includes embryology, hereditary, genetically determined partial or complete calcification of styloid ligament, growth of osseous tissue at the insertion of stylohyoid ligament, granulation tissue proliferation following trauma, degenerative alterations, metaplasia, nerve compression, and early onset of the menopause.^{16,17} The cause of onset of pain in patients previously free of symptoms is unknown, but several mechanisms have been proposed that include rheumatic styloiditis caused by pharyngeal infection trauma, tonsillectomy and evolutionary changes associated with aging (example: degenerative cervical discopathy, which may shorten the cervical spine and alter the direction of the styloid process). The pathophysiological mechanism for the pain of ES include:

- (1) Compression of the neural elements, the glossopharyngeal nerve, lower branch of the trigeminal nerve, and/ or the chorda tympani by the elongated styloid process
- (2) Fracture of the ossified stylohyoid ligament, followed by proliferation of granulation tissue that causes pressure on surrounding structures and results in pain
- (3) Impingement on the carotid vessels by the styloid process, producing irritation of the sympathetic nerves in the arterial sheath;
- (4) Degenerative and inflammatory changes in the tendinous portion of the stylohyoid insertion, a condition called insertion tendinosis;
- (5) Irritation of the pharyngeal mucosa by direct compression by the styloid process;
- (6) Stretching and fibrosis involving the fifth, seventh, ninth, and tenth cranial nerves in the post-tonsillectomy period.¹⁸

The diagnosis of Eagle's syndrome requires a high degree of suspicion. Diagnosis is based on complete medical history and a thorough clinical examination of the head and neck, to rule out other differential diagnosis including pain related to third molar, otitis media, trigeminal neuralgia, idiopathic glossopharyngeal neuralgia, temporomandibular joint pain, myofascial pain dysfunction syndrome (MPDS), temporal arteritis, cervical arteritis, migraine, cluster headache, psychosomatic, inflammatory and neoplastic diseases of the orofacial region.^{19,20} However, correct indication to radiological investigation, dedicated technology and experience are needed in order to reach a diagnosis of Eagle's syndrome. And the definitive step to confirm the diagnosis is through various imaging modalities which includes Orthopantomogram, Town's projection, lateral Cephalogram, lateral oblique view of the mandible, anteroposterior skull radiographs, CT scan, cone beam computed tomography. Among which three dimensional CT scan, is the gold standard radiological investigation for diagnosis as they precisely measure the length, angulation and calcification of the styloid process.²¹

Conservative medical management is the first line of management and usually precedes any interventional or surgical therapy. Conservative treatment includes Non-steroidal anti-inflammatory drugs (NSAIDs), Opioids, Trans-pharyngeal injection of corticosteroid and lignocaine, anti-convulsant such as carbamazepine, gabapentin, the application of heat.

Surgical management i.e. surgical shortening of the styloid process by extra-oral or intra-oral (trans-pharyngeal) route, is the most satisfactory and effective treatment for Eagle's syndrome. Surgery to shorten the styloid process is the primary treatment for Eagle syndrome is called a styloidectomy. Surgery through the mouth requires removal of the tonsils, and it can be more difficult for the surgeon to access the styloid process. There is also a slightly increased risk of damage to surrounding blood vessels. Surgery through the neck offers better access to the styloid process but will produce a scar. It can also damage surrounding parts of the body and nerves of the face. A 2017 study Trusted Source that examined this process found that 107 of 133 people who had the surgery experienced complete relief of symptoms, with an additional 20 getting partial relief. A total of 122 were satisfied with the appearance of the small cut afterward.²²

These findings suggest that this surgery may be a good option for reducing symptoms. In this presented case, diagnosis was made by complete history taking, thorough physical examination and three dimensional CT scan. Patient was suggested for surgical shortening of the styloid process for maximum clinical response.

Conclusion

Eagle's syndrome is usually rare occurrence cause of craniofacial pain. However, idiopathic unilateral pain, especially in adult women, pain exacerbation by swallowing, chewing, should always include Eagle's syndrome in the differential diagnosis of such case, to help managing the case precisely. An awareness about existence and incidence of pain related to the syndrome is important to all health practitioner (dental surgeon, otolaryngologist and neurologist) involved in diagnosis and treatment of head and neck pain to rationalized the management and get the ultimate clinical outcome.

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