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RESEARCH ARTICLE

CASE REPORT: EAGLE'S SYNDROME-ABNORMALLY ELONGATED BILATERAL STYLOID PROCESS AND CALCIFIED STYLOHYOID LIGAMENT

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Abstract

Eagle's syndrome occurs when an elongated styloid process or calcified stylohyoid ligament (a ligament connecting styloid process to hyoid) causes recurrent throat pain or foreign body sensation, dysphagia, or facial pain.[1] It is a rare condition affecting around 4 to 8 per 10,000 people. [7] It is more common in females than males (2:1 ratio) and in ages greater than 50 years. [8] The symptoms related to this condition can be confused with those attributed to a wide variety of facial neuralgias.[5]

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Introduction:-

Eagle's syndrome occurs when an elongated styloid process or calcified stylohyoid ligament (a ligament connecting styloid process to hyoid) causes recurrent throat pain or foreign body sensation, dysphagia, or facial pain. Additional symptoms may include neck or throat pain with radiation to the ipsilateral ear.[1] It classically presents unilaterally rarely bilaterally.[9] The symptoms related to this condition can be confused with those attributed to a wide variety of facial neuralgias.[5] Differential diagnoses include temporomandibular disorders, tumours, ear pathologies, skeletal neck pain, trigeminal and glossopharyngeal neuralgia.[10] In adults, normal styloid process measures up to 25 mm; clinically, styloid processes are described as elongated at lengths above 30 mm. and its tip is located between the external and internal carotid arteries, just lateral to the tonsillar fossa. It may develop inflammatory changes or impinge on the adjacent arteries or sensory nerve endings, leading to the symptoms described. [2] Diagnosis can usually be made on physical examination by digital palpation of the styloid process in the tonsillar fossa, which exacerbates the pain. Radiographic workup should include anterior-posterior and lateral skull CT films. [3] The treatment of Eagle's syndrome is primarily surgical. The styloid process can be shortened through an intraoral or external approach. We present below a case. [4]

Case Study:-

A 32 year old, Male presented to ENT OPD with chief complaints of pain and difficulty during swallowing since 1 month. He had on and off symptoms of presenting chief complaints since 4 years and the symptoms have aggravated over a period of last 1 month.

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Examination:-

General:-No abnormality detected clinically

Local:-

Ear:- Bilateral Tympanic membrane intact

Nose:-Left sided Deviated nasal Septum with Bilateral hypertrophied inferior turbinates

Throat (Oral cavity and Oropharynx):- Bilateral Grade I Tonsillar hypertrophy, Bilateral styloid process palpated in tonsillar fossa, Posterior Pharyngeal Wall No abnormality detected clinically

Neck:- Bilateral level Ib lymph node palpable

Investigations:-

CT Styloid process with TM joint showed Both styloid processes are elongated (R>L) without obvious fracture or focal lesion.Length of the styloid processes measure 72 mm and 50 mm on the right and left side respectively.The tip of the Right styloid process abuts the right oropharyngeal wall .Bilateral mandibular condyles are bifid separated by a shallow notch.

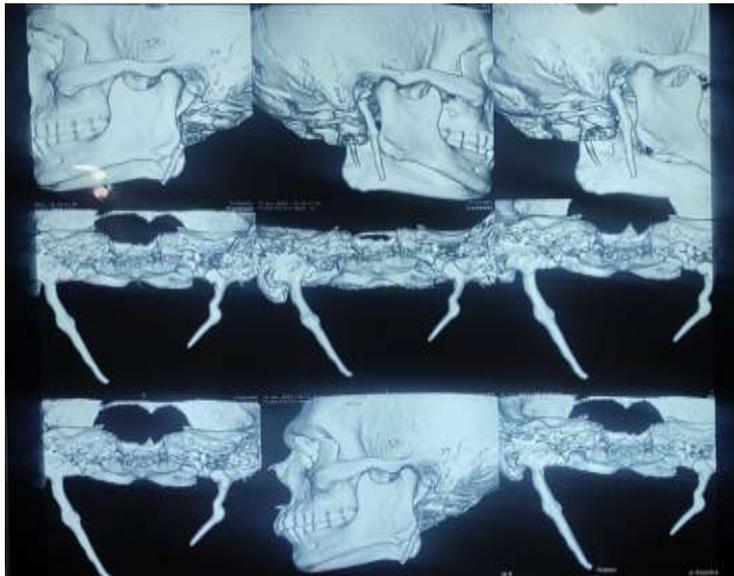


Fig 1. CT Styloid process with TM joint

Routine blood investigations were within normal limits

Treatment:-

Bilateral styloidectomy post Bilateral Tonsillectomy done under General Anaesthesia. No bleeding seen post procedure. Post operative care was taken((Medications:-Antibiotics, Anti-inflammatory, Analgesics, Antiseptic solution gargle, Antiemetics, Proton pump inhibitors, Anti fibrinolytics) with Tonsillectomy diet chart which includes soft ,cold diet for 15 days).The patient was discharged in a hemodynamically stable and improved conditions and was asked for follow-up after 10 days.In follow-up the patient was found to be symptomatically better.

Discussion:-

Eagle's syndrome, first described by Watt Weems Eagle in 1937. [6] It is a rare condition affecting around 4 to 8 per 10,000 people. [7] It is more common in females than males (2:1 ratio) and in ages greater than 50 years. [8] The aetiology of Eagle's syndrome is poorly understood. Approximately 4% of the population have elongated styloid processes, although only 4% of these cases will present clinically with Eagle's syndrome. Incidentally elongated styloid processes are also found at post-mortem. Elongation and calcification of the stylohyoid ligament can co-exist.[11] The stylopharyngeus, stylohyoid and styloglossus muscles attach to the base, middle part and tip of the styloid process, respectively. The spinal accessory and vagus nerves run medial to the styloid process. The facial nerve runs anteromedially, before piercing the substance of the parotid gland. Hence, symptoms may be intra-oral, facial, and in the neck. Examples of symptoms include cerebrovascular insufficiency, dysphagia, sensation of a

foreign body or facial/neck pain. The length of the process does not typically relate to the severity of the pain. [11] The elongated process in Eagle's syndrome can cause interaction with the internal carotid artery. Tears in the vessel and compression can cause cerebrovascular accidents. [12] Horner's syndrome has also been documented in some cases. [13] Rarely, compression of the internal jugular vein can cause increased intracranial pressure. The cause of onset of pain in patients previously free of symptoms is unknown, but several mechanisms have been proposed that include vascular/neurological impingement, rheumatic styloiditis caused by pharyngeal infections, trauma, tonsillectomy and involutional changes associated with aging (e.g. degenerative cervical discopathy, which may shorten the cervical spine and alter the direction of the styloid process). [14] Three-dimensional reconstruction of the CT images is considered to be the gold standard for diagnosing Eagle's syndrome. Eagle's syndrome classifications.

	Langalis classification (1986)- radiological appearance [15]
Type I	Uninterrupted, elongated styloid process
Type II	Styloid process apparently being joined to the stylohyoid ligament by a single pseudoarticulation. This gives the appearance of an articulated elongated styloid process.
Type III	Interrupted segments of mineralized ligament, creating the appearance of multiple pseudoarticulations within the ligament
O'Carroll and Jackson classification - location of ossification of Styloid process [16]	
Type I	Higher than the mandibular foramen (bilateral)
Type II	On the same level as mandibular foramen (bilateral)
Type III	Lower than mandibular foramen (bilateral)
Type IV	Unilateral or having different lengths on either side

Treatment of Eagle's syndrome depends on the nature of the symptoms, functional limitation and biopsychosocial impact; in these cases, of chronic pain. In our case outlined above, the patients had had long periods of time before a diagnosis was made, and is commonly a matter of years. Conservative management, pharmacological management, and surgery are all viable options for patients suffering with Eagle's syndrome. Medications suggested include steroids, anti-inflammatories, local anaesthetics, and carbamazepine. Injection of local anaesthetic ± steroid under ultrasound guidance can be considered, targeting the tonsillar branches of the glossopharyngeal nerve. Stellate ganglion block has also been considered in some cases where patients do not wish for surgical intervention. [7] There is a significant increased risk with these procedures, including arterial injection and damage to closely related structures. Other more unusual treatments [17] include traditional Chinese medicines, and manipulation with manual fracture, although this is reported as rarely of benefit, and has significant associated risks. Surgical resection is the only definite treatment; [18] this may be by an intra-oral or extra-oral approach, depending upon the surgical

assessment. Surgical outcomes vary; however, patients undergoing styloidectomy may achieve a success rate of up to 80%.^[8]

Conclusion:-

Eagle's syndrome is uncommon, and a difficult condition to diagnose, with involvement from multiple specialists. In the case presented, diagnosis was made in ENT OPD after several years of difficulty in swallowing with chronic throat pain. It is clear that a number of specialists need to have awareness of this syndrome (pain, ENT, vascular) to allow for effective diagnosis and management due to the wide range of symptom presentation. Good relations with other specialists should ensure optimal outcome for patients. There are no clearly defined diagnostic criteria for classification of Eagle's syndrome in current use. 3D CT scan is highly beneficial to understanding the anatomical relationship of structures especially when undergoing surgical resection.

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Conflicts of Interest :

None

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