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Eagle Syndrome – the issue of an elongated styloid process. A literature review.

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Abstract:**Introduction and purpose:**

Since orofacial pain poses a common ailment nowadays, it is important to identify properly its source and implement all the necessary measures that are required to mitigate its consequences. Among a wide range of various medical conditions, Eagle Syndrome should be considered during differential diagnosis. The aim of this article is to have a thorough insight into the matter of this particular condition and expand knowledge based on scientific investigations discussed in literature.

The state of knowledge:

Eagle Syndrome is a condition which manifests either by an elongated styloid process or ossification of the stylohyoid ligaments which interfere with adjacent anatomical structures resulting in many different, often nonspecific, symptoms such as, for example, limited ability of jaw opening, increased salivary flow, foreign body sensation in pharynx as well as tinnitus. However, in majority of cases it is asymptomatic condition and may be detected incidentally during clinical examination. In order to ensure accurate diagnosis, thorough medical history and anamnesis are required as well as clinical and radiographic examination. Treatment

methods that involve either conservative or surgical approach are selected depending on the severity of symptoms as well as patient's general condition.

Materials and Methodology:

The review is based on the thorough analysis of the materials obtained from "PubMed" and "Google Scholar" scientific databases that provide verified sources of information. Research was done using the following key words: Eagle Syndrome; facial pain; orofacial pain; styloid process, systematic review. The aforementioned key words were selected based on their relevance to exploring the matter of Eagle Syndrome.

Conclusions:

Eagle Syndrome is a rare clinical condition that requires detailed and precise diagnostics as well as cooperation between healthcare professionals of various specialties in order to undertake proper measures regarding condition's management. Therefore, knowledge in this field needs to be expanded and due to the complexity and nature of the Eagle Syndrome further studies are necessary to explore the matter more broadly.

Keywords: Eagle Syndrome; facial pain, orofacial pain; styloid process, systematic review

Introduction:

Orofacial pain presents a manifestation of pain referring to a wide range of symptoms, all of which are characterized by a common symptom which is a feeling of pain in the area of head and neck. Its intensity may differ depending on the particular case; however its pattern is frequently described as chronic and without remissions (1). Among various conditions that may be associated with orofacial pain, Eagle Syndrome ought to be mentioned. It was discovered and mentioned in 1652 for the first time by Pietro Marchetti, nevertheless it was not until 1937 that the clinical syndrome was assuredly described by Watt Weems Eagle, after whom it was later named. The condition results from an elongated styloid process or stylohyoid chain ossification and may lead to a diverse range of implications and symptoms such as, for example, cervical, orofacial and craniofacial pain (2). Meanwhile, according to study by Palesy et al. (3) examination of the styloid process is unfortunately a rare practice in a group of patients suffering from facial and cervical pain, as well as from mandibular dysfunction. The aim of

this review is to explore the subject of Eagle Syndrome thoroughly with reference to its etiology, pathophysiology, implications and the methods of treatment based on available, valid, scientific research described in literature.

The state of knowledge:

Definition and epidemiology

Eagle Syndrome presents a rare clinical condition defined by a broad spectrum of symptoms caused either by an abnormally long styloid process or the ossification of the stylohyoid ligaments and their interference with the adjacent tissues and anatomical structures. It may lead to, inter alia, severe pain occurring frequently in the anterolateral neck region with referred pain to the ear (4). However, it should be emphasized that the majority of cases involve asymptomatic patients who learn about the condition inadvertently. The prevalence of elongation of the styloid process is observed in approximately 4% of the general population (5). The frequency and occurrence of symptoms differ among various sources. According to Saccomanno et al. (6) they have been estimated by Eagle as 0,16%. In contrast, Sadaksharam et al. (7) reports that among patients diagnosed with the condition, 4-10.3 % suffer from symptoms. The elongation of the styloid process may be not only unilateral, but bilateral quite frequently as well, and what is intriguing, it has been reported that symptomatic cases mainly involve unilateral symptoms (6). These findings are consistent with other studies (2, 8, 9, 10) that also indicate that in the majority of symptomatic cases the symptoms are unilateral even in cases with bilateral elongation, and that bilateral symptoms are not observed frequently. Furthermore, it is worth to notice that age presents a contributing factor to the development of the condition, as the Eagle Syndrome is most frequently observed among adult patients, especially between 30 and 50 years of age with the emphasis that it is three times more likely to affect women than men (11, 12). A study by Woolery (13) confirms that Eagle Syndrome is more frequently associated with females. Moreover, the study indicates an increased prevalence of the anatomic variant of the stylohyoid complex in the population of elderly women, especially between 60 and 79 years of age.

Classification of Eagle Syndrome

The classification of Eagle Syndrome was developed by dr. Eagle in 1948 and 1949 when he distinguished two types of the condition – the classic type and the carotid artery type

(14, 15). The classic type has been commonly observed after a tonsillectomy procedure. It might lead to a variety of implications and symptoms such as, for example, dysphagia, dysphonia, pain referred to the ear, constant sore throat as well as the sensation of foreign body in the pharynx which leads to an increased need of swallowing (15). According to Eagle's assumptions, the scar tissue in the area of tonsillar fossa has the potential to exert compression and stretching forces on several cranial nerves including the trigeminal, facial, glossopharyngeal and vagus nerves around the styloid process which leads to a wide range of implications (16). In contrast, when elongated styloid process or ossified stylohyoid ligaments impinge the artery, the other type of condition distinguished by Eagle, which is the carotid artery type, should be discussed. It ought to be emphasized that the symptoms related to this type of Eagle Syndrome differ depending on the artery involved (15). When the external carotid is affected the main symptoms involve unremitting cervical pain or a pain radiated to the eye area and customary for those cases is that the severity of symptoms increases during movements of the head towards the affected side. In contrast, when internal carotid artery is affected, the main and primary symptoms involve headache and pain from the occipital area to the ophthalmic region as a result of irritation of the periarterial sympathetic nerve plexus and what it worth noting such condition is named "carotidynia" or "carotid artery syndrome" (15). Furthermore, a study by Farhat et al. (17) which, in this aspect, remains consistent to a study by Cano et al. (18) indicates that direct, near proximity to carotid artery may result in several systemic complications such as carotid dissection and transient ischemic attacks during the movement of the head towards the affected side. The classification introduced by Eagle is still valid and used nowadays, however it should be underscored that it is not universally effective and cannot be always applied as there have been cases described of symptomatic patients without any cervicopharyngeal trauma or tonsillectomy record (19). In addition, there have been many asymptomatic patients reported despite the ossification of stylohyoid complex with no correlation detected between the extent of ossification and the prevalence and severity of pain (15).

Variations of the styloid process

Styloid process is a thin, cylindrical anatomic structure. It provides an attachment site for two ligaments stylomandibular and stylohyoid, as well as three muscles – styloglossus, stylohyoid and stylopharyngeal (6). Its proper length exhibits a range of variability oscillating between 1,52 to 4,77 cm according to Mortellaro et al. (20). However, the literature is not

consistent regarding this issue, as according to Raina et al. (21) normal length of styloid process is estimated between 2,5- 3 cm and if it exceeds this limit it is considered to be elongated. Thot et al. (22) has presented an interesting thesis regarding the length of the styloid process indicating that it is not, in and of itself, a risk factor in contrast to the situation when an abnormal length co-occurs with variations of anatomic shape of the styloid process, as both anteriorly and medially oriented variations make an elongated process a sole cause of the Eagle Syndrome. However, as Murtagh et al.(23) states, since a wide range of patients with radiographically found elongated styloid process are asymptomatic, an extended length as a unique finding cannot be considered as a pathognomonic for Eagle Syndrome. What is interesting regarding the issue of the abnormal length of the styloid process is that it may be a predictor of peak bone mass according to research by Sisman et al. (24).

The implications of certain deviations of the styloid process

The issue of various deviations in the structure of the styloid process and their implications has been brought to attention by Piagkou et al. (2) who has conducted a study which has indicated that a posterior deviation may ensnare internal carotid and jugular vein as well as the glossopharyngeal (IX), vagus (X), accessory (XI), and hypoglossal (XII) cranial nerves, a lateral deviation may be the cause of the impingement of the external carotid artery at its bifurcation into the superficial temporal and maxillary arteries, whereas a medial direction fosters the impingement on the tonsillar fossa. In contrast, anterior angulation may be the reason of the irritation of the mucosa or other structures situated in the tonsillar fossa.

Radiographic appearance of the styloid process

Another interesting classification has been described with regards to the radiographic appearance of the styloid process in terms of its structure, length and angulation as well as on the pattern of ossification measured by calcification degree. It involves several types, among which Type I – an elongated process- is the most common. Type II includes a pseudo-articulated process, whereas for Type III a segmented process is characteristic. Considering the angulation of the styloid process, three types have been distinguished - a narrow type when the angulation is less than 65°, a normal type in cases when the angulation oscillates between 65° and 75° and a wide if the angulation exceeds 75°. Taking another important factor into consideration, which is the calcification degree the most frequent is Type A in case of which, a calcified outline is observed. Type B includes a partially calcified styloid process, whereas for Type C nodular

calcification is characteristic and Type D is fully calcified (2). As Ilgüy et al. states (25), the most commonly observed type of elongated styloid process is Type I calcified either partially or completely. Another intriguing correlation has been observed between stylohyoid complex alterations and the presence of osteophytes in the cervical spine in the study conducted by Guo et al. (26) The study indicated that not only elongated styloid process, but also enlargement and different variations of ossification of the certain parts of stylohyoid ligament were found to be associated with anteroposterior and transverse dimensions of ligamentous ossification or osteophytes of the cervical spine.

Theories of styloid process ossification

Styloid process is a bone projection situated in an anteromedial position with respect to the mastoid process of temporal bone (6). It begins to ossify at the end of pregnancy and undergoes a process of calcification during the first eight years of life. Its etiopathogenesis may be explained by various theories explained in literature, for example the ones described by Steinmann et al. (27) who states that the first one involves the theory of Reactive Hyperplasia, the essence of which is the activation of the remnants of the original fibrocartilaginous and connective cells initiated by a trauma. Another one is the Theory of Reactive Metaplasia which is characteristic for the cases where trauma is followed by an abnormal healing which initiates the ossification of the stylohyoid ligament. In contrast, the third theory, the Theory of Anatomic Variance is not associated with any trauma. Furthermore, another interesting theory has been described by Camarda et al. (19) and it involves a theory of Aging Developmental Anomaly characterized by the lack of evident radiographic ossification. The authors have stated that the loss of elasticity of soft tissues presents a contributing factor for the inflammation localized to the junction of the stylomandibular ligament to the lesser cornu of the hyoid bone which has been named as Pseudostylohyoid Syndrome. It should be emphasized that the ossification of the styloid process is a complex process which is also considered to be related to a wide range of systemic conditions involving, for instance, end-stage renal disease and other conditions proceeding with the alternation of calcium and phosphorus metabolism (28).

Etiology of the Eagle Syndrome

The issue of explaining the etiology of the condition has been brought to attention by Murtagh et al. (23) who has proposed three different theories in order to clarify the matter. The

first theory – the theory of congenital elongation of the styloid process results from the perseverance of the cartilaginous precursor. According to the second theory an indefinite, unknown process leads to the calcification of stylohyoid ligament, whereas the last theory indicates that the osseous tissue grows at the area of stylohyoid ligament ultimately resulting in the development of the condition.

Diagnosis and symptoms

Diagnosis of Eagle Syndrome may not always be straightforward and made without hesitation as a wide range of various symptoms have been observed. They are frequently nonspecific (26), however Sowmya et al. (12) indicates that the exemplary Eagle Syndrome involves increased salivation, limited ability to open the mouth, pain and a sensation of a foreign body in pharynx, as well as tinnitus. Nonetheless, the author also highlights that asymptomatic cases are frequently observed. Due to the aforementioned reasons, arriving at a proper diagnosis may be difficult and the condition may be inadvertently detected or confused with other ailment or disorder such as, for instance, facial neuralgias, temporomandibular joint disorders or dental diseases (29). Patients suffering from persistent symptoms frequently seek help among various specialists such as, for example, dentist, neurologist, otolaryngologist, family physician, neurosurgeon, maxillofacial or oral surgeon as well as psychiatrist (2, 8).

Diagnosis criteria

Diagnosis is usually based on the clinical examination that involves palpation of the styloid process, as well as on collecting a proper medical history, as it must be underscored that fine, detailed anamnesis is of the utmost importance (29). Among other diagnostic tools mentioned in literature (2), local injections of 2% lidocaine into tonsillar fossa also should be distinguished. Using radiographs such as panoramic radiograph or three-dimensional CT scan which is considered to be the criterion standard in radiological diagnosis is also essential (2, 8, 9, 29). Among other radiographic projections that are quite commonly used in order to confirm clinical diagnosis antero-posterior head radiograph, lateral head and neck radiograph and orthopantomogram radiograph should be also distinguished. Nonetheless, CT scan is the most advantageous as it provides various, precise, three-dimensional information regarding the stylomandibular complex that may be diagnostically significant such as, for example, the length of the styloid process, its anatomic variance as well as its relations to the adjacent anatomic

structures (30, 31). All in all, there have been three major criteria determined in order to diagnose Eagle Syndrome, and they involve an elongated styloid process palpable in tonsillar fossa during the clinical examination, radiographically detected exceeded length (over 30 mm) of the styloid process and pain which severity increases during swallowing (2, 8, 9, 10, 32).

Differential diagnosis

If an elongated styloid process is concomitant to cervical or orofacial pain it is of the utmost importance to conduct differential diagnosis that ought to include a wide range of conditions such as TMD, inner ear diseases, chronic tonsillopharyngitis, hyoid bursitis, neuralgias, unerupted molars, cluster headache, migraine-type headache, Sluder's syndrome, cervical vertebral arthritis, temporal arteritis and esophageal diverticula (30, 33, 34).

Pathophysiology of symptoms

The pathophysiology of various symptoms of Eagle Syndrome has been thoroughly described by Murtagh et al. (23) and it is based on the division into four theories. The first one involves the theory of a traumatic fracture of the styloid process with a proliferation of granulation tissue which may force pressure on the adjacent tissues. Another one includes a compression of the mandibular branch of the trigeminal nerve, the chorda tympani or the glossopharyngeal nerve. In contrast, the essence of the third theory is an insertion of tendonitis as a result of an inflammation of the tendous part of the stylohyoid insertion, whereas the fourth theory includes the irritation of the pharyngeal mucosa by either straight-through compression or postoperative scarring after tonsillectomy. The final theory is explained by an irritation of the sympathetic nerves surrounding the carotid vessels.

Treatment methods

Treatment of an Eagle Syndrome is considered to be complex and dependent on the severity of symptoms as well as the patient's condition. It may involve a conservative approach or a surgical intervention (5, 29). If operation needs to be performed it is necessary to emphasize the importance of selecting the proper method as it may contribute not only to reducing the duration time of the procedure, but also the risk of potential complications, injuries and postoperative pharyngalgia (35).

Conservative approach:

Undergoing conservative management involves the application of analgesics, psychotropic and anti-inflammatory medications, physiotherapy and alternative therapies (36, 37, 38). According to Maher et al. (39) for some patients, conservative approach poses a sufficient form of treatment, however in order to obtain long-term, satisfactory effects regarding reduction the severity of symptoms it is necessary to implement various therapeutic agents or interventional modalities. Regarding neuropathic and inflammatory symptoms, they may be abated and reduced by various medicaments such as, for example, anticonvulsants, tricyclic antidepressants as well as NSAID therapy. Furthermore, the author highlights the positive outcome of therapy using fluoroscopy-guided blocks or intra-oral tonsillar pillar for some of the patients.

Surgical intervention:

Surgical management is recommended for patients who do not respond well to the conservative methods of treatment as well as for those with severe, intractable symptoms and it involves either stylectomy or microvascular decompression (40). Shortening of the styloid process may be performed using either intraoral or extraoral cervical approach. The latter one is preferred due to better visibility of the operated area as well as the lower risk of deep cervical infection. Nonetheless, the procedure is not without deficiencies that include a formation of postoperative scar (29). Another method that should be discussed is transoral resection, which advantage is aesthetics as it does not cause any external scars. However, it may pose an increased risk to the development of deep cervical infection as well as potential neurovascular injury (2, 10, 32). Wang et al. (35) conducted a study in which 103 medical individuals suffering from Eagle Syndrome took part. In order to determine a particular, the most appropriate method of surgical treatment, patients undergoing general anesthesia were examined and the proper operation method was selected based on oropharyngeal palpation, CT reconstruction of the styloid process as well as particular criteria. The external cervical approach was chosen for patients whose styloid process could not be perceptible through the mouth but could be perceptible under the jaw or when CT scan indicated that the inclination angle was not large. In contrast, if the styloid process could be perceptible during intraoral palpation or could not be directly perceptible but the CT scan indicated that the bone inclined towards oropharynx and

its distal part was in the vicinity of the oropharynx cavity, the choice of surgical method involved styloid process resection by performing transoral surgery.

Extraoral approach:

In the study by Wang et al. (35) surgical extraoral method entailed the creation of an arc-shape, curve incision which length measured circa 4 cm. It was executed 1-2 cm from the posterior edge of the ramus of the mandible and extended to its inferior edge. During the procedure the mandibular angle was thoroughly exposed in order to visualize the styloid mandibular ligament as well as styloid process and attached ligaments. Further, the styloid hyoid ligament was cut and the styloid process was shortened.

Transoral surgery:

According to Wang et al. (35) 82 out of 103 medical individuals underwent surgery that involved intraoral approach. The operation was preceded by a clinical examination. There were several methods of the surgery discussed, one of them including the tonsillar fossa approach selected when the tonsil was too large and affected the full exposure of the styloid process. In those cases, tonsillectomy was carried out. During the operation the styloid process was exposed, the styloid process periosteum was cut, and soft tissues were excised. The styloid process was cut and removed using bone-biting forceps, and the cut-off length estimated between 10-38mm. Further proceedings depended on the length of the residual styloid process and involved its stripping and shortening in case of the large length. Another surgical method involved palatoglossal arch – soft palate approach.

Conclusions

Chronic orofacial pain is a condition that may be caused by a wide range of various diseases and disfunctions, among which Eagle Syndrome ought to be taken into consideration. It is a rare yet significant condition that requires not only careful and thorough clinical examination, but also detailed imaging using radiographs in order to make an accurate diagnosis and undertake all the requisite measures. While conservative treatment may alleviate some symptoms, surgical intervention that involves resection of the styloid process is often required to obtain satisfying outcome and improve patient's condition. Furthermore, it is of the utmost

importance to increase social awareness in this matter, especially among healthcare professionals in order to improve diagnostic accuracy as well as selection of the most adequate treatment procedures. To achieve these goals, multidisciplinary approach is often necessary, thus cooperation between various specialists such as, for instance, dentists, neurologists, otolaryngologists, family physicians, maxillofacial oral surgeons and others is essential. Nevertheless, it is worth highlighting that due to the nature and complexity of the Eagle Syndrome, further research in this matter is required and necessary in order to develop both standardized as well as most effective diagnostic and treatment guidelines.

DISCLOSURE:

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