

CLINICAL CASES

Dent. Med. Probl. 2015, 52, 3, 366–370
ISSN 1644-387X

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Elongation of the Styloid Process – Eagle Syndrome – Case Reports of Patients on Dialysis

Wydłużenie wyrostka rylcowatego – zespół Eagle'a – opisy przypadku pacjentów dializowanych

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A – research concept and design; B – collection and/or assembly of data; C – data analysis and interpretation;
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Abstract

Eagle syndrome or syndrome of the prolonged styloid process of the temporal bone is a rare syndrome, whose cause is an extended styloid process and often concomitant with an ossification of stylohyoid ligaments. A patient typically reports to a doctor because of sudden severe pain in his ear, throat, floor of the mouth, the oral side of the neck or jaw angle around the tonsillar fossa. This pain occurs when you open your mouth, turn the head or swallow. Additional symptoms which are coexisting may be drooling, dysphagia,odynophagia, trismus, sensation of something in the throat. Pain symptoms occur suddenly and last from a few seconds to several minutes. The method of treatment for Eagle Syndrome can be twofold and depends on the presented type of Eagle Syndrome. Nonsurgical treatment methods, such as injecting the tonsillar fossa after the palatal tonsil with local anesthetic agents and steroids, are used in the classic type. Unfortunately, in most cases, the results are not satisfactory. Two patients recently came to the clinic presenting the above-mentioned symptoms of varying severity. Panoramic radiographs and CBCT revealed much elongated styloid processes (**Dent. Med. Probl. 2015, 52, 3, 366–370**).

Key words: styloid process, Eagle syndrome,odynophagia.

Słowa kluczowe: wyrostek rylcowaty, zespół Eagle'a, odynofagia.

Eagle syndrome was first described in 1937 by the American otorhinolaryngologist Watt W. Eagle [1]. Based on the analysis of patients, Eagle distinguished two categories of the syndrome: the classic one and carotid artery syndrome [1–5].

The classic styloid process syndrome is usually associated with tonsil removal when the cranial nerve endings in the tonsillar fossa are compressed between fibrous tissue and an elongated styloid process. Irritation of cranial nerves V, IX and X leads to the pathognomonic symptoms of Eagle syndrome. These include pharyngeal pain localized in the tonsillar fossa after tonsil removal, swallowing difficulty, foreign body sensation in

the throat, pain upon swallowing and, rarely, voice change.

The second type, i.e. styloid-carotid artery syndrome, is due to compression of the sympathetic chain in the carotid sheath by the elongated styloid process. The main symptoms include pain in the eye, visual disturbances and unilateral headaches. In rare cases syncope may also occur.

In 1986, Langlais et al. [6] classified elongated styloid process and mineralized stylohyoid ligaments into three types (Table 1):

- Type I (Uninterrupted): the elongated styloid process measuring above 30 mm is uninterrupted.

Table 1. Classification of elongated styloid process and mineralized stylohyoid ligaments by Langlais

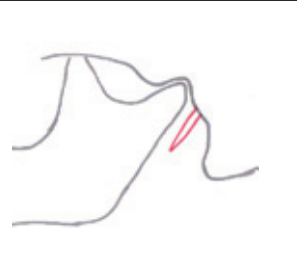
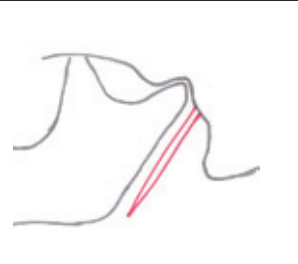
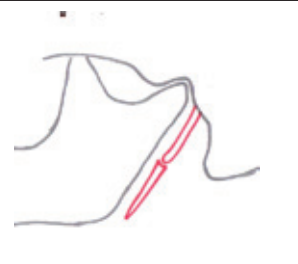
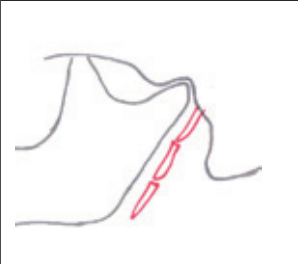
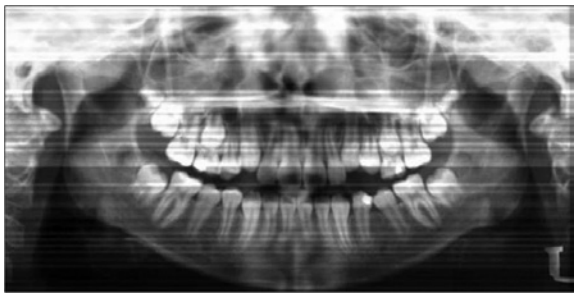

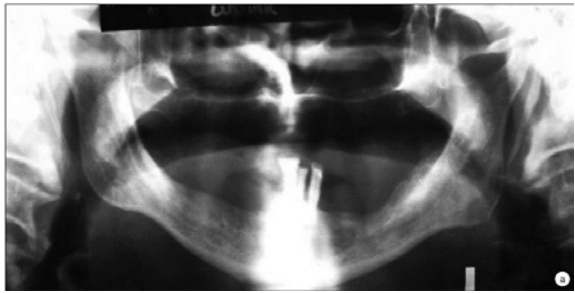
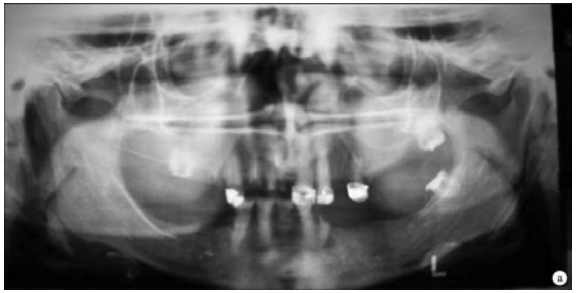
			
Normal	Type I – elongated	Type II – pseudoarticulation	Type III – segmented

Table 2. Panoramic picture of patients from article K. Thun-Szretter et al.: “Evaluation of styloid process on the panoramic radiographs – preliminary report”

	
Panoramic picture of 13-year-old female patient. Type “O” – styloid process not visible on OPG.	Panoramic picture of 45-year-old female patient. Type “A” – styloid process ends above the mandible foramen.
	
Panoramic picture of 42-year-old male patient. Type “B” – styloid process ends between mandible foramen and mandible angle.	Panoramic picture of 66-year-old male patient. Segmented processes. Type “C” – styloid process ends below mandible angle.

- Type II (Pseudoarticulation): characterized by a pseudoarticulation.
- Type III (Segmented): consisting of 2 or more segments.

O’Carroll’s classification, modified by Thun-Szretter [7], is also helpful. It distinguishes four ‘varieties’ of styloid processes on a pantomogram (Table 2):

- O – styloid processes not visible on an orthopantomogram,
- A – the apex of the styloid process projecting above the mandibular foramen,
- B – the apex of the styloid process situated between the mandibular foramen and the angle of the mandible,
- C – the apex of the styloid process placed below the mandibular angle.

Studies of the anatomical measurements of the styloid process have been performed and published several times and the gathered data shows population and ethnic differences [8–12]. The findings of research by Thun-Szretter et al. [7] indicate that variety C, in which the apex of the styloid process is placed below the mandibular angle, is rare. They noted the occurrence of divided (split) processes in the case of all patients diagnosed with variety C and in the case of one patient whose apex of the styloid process was situated between the mandibular foramen and the angle of the mandible (variety B).

Eagle syndrome can be treated in two different ways and the treatment depends on the type of syndrome presented. Non-invasive methods such as local anesthetic and steroid injections to the ton-

sillar fossa are applied in the classic type. Unfortunately, in most cases the results are not satisfactory.

Apart from pharmacological treatment, surgical methods via an intra-oral or extra-oral approach are applied [13, 14]. Both methods have advantages and disadvantages. The main disadvantages of the extra-oral approach involve surgery under general anesthesia and a scar that is left on the neck. However, good visibility of the surgical site is a huge advantage of the method [1–4]. The trans-oral approach, which leaves no visible scarring, involves risk of iatrogenic injury to the anatomical structures localized around the surgical site as well as a greater risk of infection of the parapharyngeal space. Regardless of the approach chosen, the surgery consists of removal of the ossified ligament and/or resection of the styloid process.

Surgical methods are undoubtedly the most effective in removing symptoms, with efficacy up to 80%.

One method of non-surgical treatment which is considered highly risky due to possible complications is a manual fracture of the styloid process. The procedure is performed without opening the neck tissues, through skin layers. This method is rather hazardous due to a number of anatomical structures located in the area which can become damaged. In addition, the method does not bring satisfactory therapeutic results.

Case Reports

Case I

A 56-year-old male, M. Sz., reported to a dental surgery clinic due to left-sided facial pain and swelling in the second and third molar area. The patient was given immunosuppressive treatment due to kidney transplantation. A clinical examination revealed inflammation around tooth 37, which was extracted with antibiotic prophylaxis, and tooth 38, which was to be extracted during the following appointment. A few days afterwards, the man reported due to facial pain and swelling. A soft-tissue ultrasound imaging of the cheek was performed which revealed the presence of a foreign body. Under local anesthesia, the patient underwent removal surgery. Histopathology results revealed no changes and no foreign body, the patient, however, reported relief from pain. 2 weeks following the procedure, the patient complained of pain in the ear and slight trismus, which may indicate TMJ inflammation. A CT revealed exacerbated inflammation of the left TMJ. Anti-inflammatory drugs (Diclac®) and an antibiotic were prescribed. Additionally, the CT showed

bilaterally elongated styloid processes and ossified stylohyoid ligaments, which may have been the cause of the symptoms reported by the patient. The location of the split of the styloid process on the left side (Fig. 1) and a segmented type on the right side (Fig. 2) were well visible, which qualifies the said processes as type II and III (as per Langlais) as described by Bozyk et al. [9]. This type – pseudoarticulation – is characterized by the presence of a pseudoarticulation (Fig. 3). The occurrence of this kind of anatomical anomaly is extremely rare in males (6%). Based on the examination and clinical picture, the patient was diagnosed with Eagle syndrome. The patient will be advised about the diagnosis and possible treatment methods in addition to a required laryngological examination.



Fig. 1. Panoramic picture, left side processus styloideus type 2 (as per Langlais), variety C (as per O'Caroll)



Fig. 2. Panoramic picture, right side, processus styloideus type 3 (as per Langlais), variety C (as per O'Caroll)



Fig. 3. CBCT left side, styloid processus type 2

Case II

A 65-year-old female was referred to a dental surgery clinic by the Department of Immunology in order to eliminate any possible causes of inflammation in the oral cavity. The patient had been suffering from glomerulonephritis for 40 years and the disease had recently become exacerbated. A clinical examination revealed the need to review the endodontic treatment of teeth 31 and 34 or their extraction if the endodontic treatment proved ineffective. An OPG revealed bilaterally elongated styloid processes reaching the mandibular angle on the right side (Fig. 4) and the middle of the ramus on the left side (Fig. 5). Both pro-

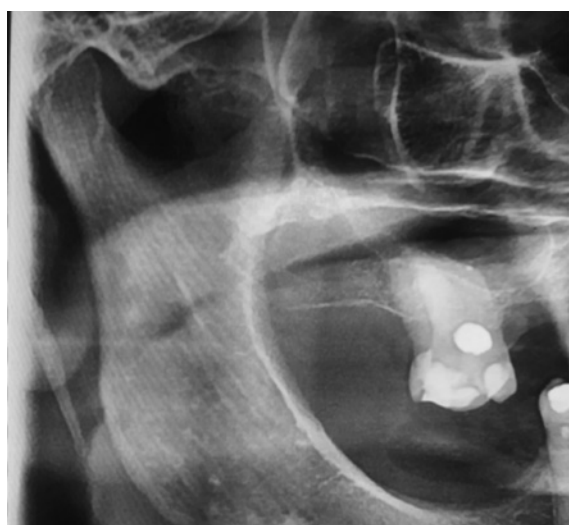


Fig. 4. Panoramic picture, right side type 1 (as per Langlais), variety B (as per O'Caroll)



Fig. 5. Panoramic picture, left side type 1 (as per Langlais), variety B (as per O'Caroll)

cesses were classified as type I. Both the process on the right side and the one on the left were classified as variety B as per O'Caroll. Subsequently, upon additional examination, the patient reported pain exacerbated by gentle palpation of the carotid triangle.

Discussion

Eagle syndrome is a rare condition which is difficult to diagnose due to its uncharacteristic symptoms such as: pharyngeal pain, odynophagia, headache and foreign body sensation in the throat. Diagnosis can be made by computed tomography, which reveals elongated styloid processes and forms the basis for diagnosis.

Both patients had been affected by the consequences of glomerulonephritis, the first of whom was given immunosuppressive treatment. There is no direct evidence to suggest that electrolyte imbalance and proteinuria in the course of renal failure could have an influence on the excessive growth of the styloid process. The existing hypothesis, which requires confirmation, suggests that there is a correlation between a higher risk of renal insufficiency and an excessively elongated styloid process as a genetic defect. The hypothesis requires confirmation on a larger group of patients.

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Conflict of interest: None declared

Received: 19.02.2015

Revised: 04.03.2015

Accepted: 04.04.2015