



Perioperative anesthesia management in a patient with Eagle's syndrome

Özgenur BACAŞIZ *^{ORCID}, Elif Bengi ŞENER ^{ORCID}

Department of Anesthesia and Reanimation, Faculty of Medicine, Ondokuz Mayıs University, Samsun, Turkey

Received: 21.03.2022

Accepted/Published Online: 16.07.2022

Final Version: 29.10.2022

Abstract

Eagle syndrome, a prolonged styloid syndrome, is a clinical disorder caused by an elongated, enlarged, and angulated styloid process. Most of the cases are asymptomatic. Symptomatic cases are presented as continuous, intermittent pain in the face and anterolateral neck region, depending on position; reflected pain may be seen in the ipsilateral ear and temporomandibular joint. Patients with a bilateral prolonged styloid process may present unilateral complaints. Symptoms can manifest themselves with foreign body sensations in the throat, episodes of dysphagia, and syncope. Syncope episodes are one of the most serious clinical manifestations of the syndrome and are called 'Carotid-Styloid Syndrome'. Patient anamnesis and physical examination are the most important steps for diagnosis. Palpation of the styloid process in the tonsillar fossa is a clue for Eagle syndrome. Imaginary examinations should confirm the diagnosis. The local anesthetic injection technique into the tonsillar fossa can be used for differential diagnosis. The reduction of post-injection pain is a finding favoring Eagle's syndrome. Furthermore, surgical and medical methods can treat Eagle's syndrome. In this article, the perioperative anesthesia management of a 38-year-old woman with Eagle syndrome planning to undergo axillary lymph node biopsy in Ondokuz Mayıs University Hospital is presented as a case report.

Keywords: Eagle syndrome, elongated styloid process, pain syndromes, stylohyoid syndrome, neurological monitorization

1. Introduction

The Styloid process (SP) is part of the temporal bone, located anterolaterally, and forms the lower part of the bone. It is lateral to the tonsillar fossa between the internal and external carotid arteries (1). SP lies adjacent to the styloglossus, stylopharyngeus, and stylohyoid muscles. Embryologically, the structures found in this region originate from Reichert's cartilage (second branchial arch). The Styloid process is located in the maxilla-vertebra-pharyngeal space where the carotid artery, internal jugular vein, facial nerve, glossopharyngeal nerve, vagus nerve, and hypoglossal nerve pass. The tip of the styloid process is connected to the lesser horn of the hyoid bone via the stylohyoid ligament. The clinical disorder resulting from the elongation of the styloid process, calcification or ossification of the ligaments it is associated with (stylohyoid, stylomandibular ligament), is called Eagle's syndrome (2).

W. W. Eagle described the clinic, radiological diagnosis, and treatment of Eagle's syndrome in 1937 (3). Eagle defined the normal length of the styloid process as 25 mm (millimeters) and named protrusions longer than 25 mm as long styloid process. Despite that W. W. Eagle was the first to describe the syndrome, Italian surgeon Pietro Marchetti detected styloid process elongation and ligament calcification in 1652 but did not mention the clinical reflections of this

pathology (4).

Surgical trauma, blunt trauma, chronic inflammation of the ligaments (especially the stylomandibular ligament), styloid process osteitis, tendinitis, and mucositis may be responsible for the syndrome's pathogenesis (5). However, the etiological factor may also be calcification due to aging (6). In endocrine diseases or chronic renal failure, hypercalcemia may also cause ligament calcification. Apart from these etiological reasons, prosthesis elongation can also be congenitally observed with the malformation of Reichert's cartilage in the fetal period (7).

Most people with an elongated styloid process are not diagnosed with Eagle's syndrome if they are asymptomatic. Even if they are not symptomatic, the perioperative anesthesia approach should be known, and necessary precautions should be taken in cases with an elonged SP.

2. Case Report

A 38-year-old, 50 kg (kilogram) female patient was referred to us for axillary lymph node biopsy to prepare for preoperative anesthesia. In her anamnesis, the patient who had Eagle's syndrome, asthma without attacks and untreated follow-up, and history of hypotension attacks several times a month did not describe syncope. She had undergone

*Correspondence: ozgenur15@hotmail.com

tonsillectomy and bilateral styloidectomy with intraoral approach twice in chronological order. Enquiring the visible symptoms due to Eagle's syndrome, the patient did not describe pain when the head was in the supine and neutral position; there was a pain in the neck and tongue root in the deviations of the head to the left and right, and the pain reflected in the bilateral ears. The patient did not describe syncope attacks, vertigo, or vision-related symptoms. After the preoperative evaluation, we planned the operation day and took the patient to the operating room. Following the ASA (American Society of Anesthesiologists) standard monitoring recommendations, we performed electrocardiography, pulse oximetry, and noninvasive blood pressure monitoring. We enquired the patient again about the positions of her head she did not feel pain in and repeated her neurological examination. We fixed the patient's head in a neutral supine position and took measures to prevent deviance from right to left. We placed cerebral oximetry sensors appropriately and took the basal value to monitor cerebral blood flow closely. We recorded cerebral oximetry values for the left and right hemispheres intraoperatively every ten minutes. After recording the cerebral oximetry basal values, the patient was preoxygenated. We administered 1mg midazolam intravenously (i.v.), and started 0.2 µg/kg/min remifentanyl infusion. We administered 40 mg i.v. lidocaine, 2mg/kg i.v. for induction propofol, and 1mg/kg i.v. rocuronium and performed mask ventilation for one minute so that the position of the head was not distorted. We successfully performed fiberoptic intubation, avoiding anteflexion and extension of the head. We provided intraoperative anesthesia maintenance with a MAC (minimum alveolar concentration) value of one, sevoflurane, oxygen-air mixture, and 0.2-0.5µg/kg/min remifentanyl infusion. The operation lasted for 75 minutes. We observed no intraoperative hemodynamic instability and recorded no pathological value in the cerebral oximetry follow-up. The patient's muscle relaxation was reversed by 2mg/kg sugammadex and taken to the recovery unit for close postoperative follow-up at the end of the operation. The patient had no problems in the postoperative follow-up, and we transferred her to the recovery room with the head in a neutral position, hemodynamically stable, conscious, with normal neurological examination, and a modified Aldrete score of nine.

3. Discussion

According to the Genetic and Rare Diseases Information Center (GARD) data, 4% of the entire population has elongated SP. 4-10.3% of cases with an elongated styloid process are symptomatic. The incidence of Eagle syndrome is estimated to be 0.16%. Yavuz et al.'s (8) study in 2008 evinced that the length of the styloid process in Turks varies between 3.5 and 8 cm, with an average of 5 cm on the left SP and 5.2 cm on the right SP.

Clinical complaints are usually seen in patients aged 50 years and older and detected three times more in women than

men (7). Studies have shown that the syndrome is more symptomatic in women than men (9) and more common in older women. This suggests that it may be associated with menopause (10). The syndrome generally progresses asymptotically. In addition to the clinical course of an asymptomatic character, only unilateral complaints can be observed in patients with bilaterally prolonged SP. Clinically, earache, foreign body sensation in the throat, a sharp and blunt pain aggravated by swallowing, and neck rotation may be seen on the side with an elongated SP. If the elongated process presses on the carotid, syncope episodes are encountered. There are two types of the syndrome: the classical type (stylohyoid syndrome) is usually seen after tonsillectomy or pharyngeal surgery, and symptoms occur when fibrotic tissues compress the cranial nerve (5, 7, 9, and 10th cranial nerve). Pain-related complaints are in the foreground in the classical type (11). The second form, called carotid syndrome (Stylocarotid syndrome), occurs due to the end part of elongated SP or pathological ligaments compressing the sympathetic fibers adjacent to the carotid artery. It progresses with syncope attacks, and radiating pain can be present in the carotid zone (11). Symptoms are related to SP's length, width, angulation, the side of deviation, and the ligaments' ossification level (12). There is no relationship between patient age and symptoms (9). The technique of pain formation or exacerbation of pain by palpating the styloid process in the tonsillar fossa can be used in the physical examination. The styloid process, whose length is within normal limits, is not palpable in the tonsillar fossa (13). After the physical examination of patients considered to have Eagle's syndrome, an otolaryngologist should visualize the process with maxilla facial computed tomography or three-dimensional computed tomography. Radiologically, computed tomography is the most valuable method in diagnosis (14).

The primary treatment for Eagle's syndrome is the surgical shortening of the prolonged SP (styloidectomy). Analgesics, anti-inflammatory drugs, antidepressants, and corticosteroid-containing drugs can be used in medical treatment. Relieving pain by injecting local anesthetic agents into the tonsillar fossa is a method that can be used in diagnosis and treatment (15).

Eagle's syndrome is mostly asymptomatic, and the clinical status of symptomatic cases can be confused with many diseases. While creating the differential diagnosis list, Eagle's syndrome must be added to the list. Even if asymptomatic, if being taken to surgery for other reasons, there are points to be considered in patients with an elongated styloid process;

- The subtype of Eagle syndrome should be determined.
- Neurological examination and neurological monitoring should be performed (especially in cases with the carotid syndrome).

- Preoperatively, the position in which the patient's pain aggravates should be determined, and the intraoperative position should be planned according to this data.

- If the treatment of Eagle's syndrome is planned medically, antidepressant and steroid-containing drugs should be questioned, and preoperative drug regulation should be performed.

- If there is ligament calcification in the syndrome's pathogenesis, it should be kept in mind that chronic renal failure and endocrinological diseases may cause this condition.

Anesthesiologists should be aware of this syndrome and should take the necessary perioperative measures for the patient's medical well-being. Although it is a rare disorder, it requires finesse in anesthesia management.

Conflict of Interest

There is no conflict of interest between the authors.

Funding

None.

Acknowledgment

None.

References

1. Kurmann PT, Van Linthoudt D. Eagle syndrom: a rare cause of lateral neck pain. *Praxis*. 2007; 96(8): 297-300.
2. Fini G, Gasparini G, Filippini F, Becelli R, Marcotullio D. The long styloid process syndrome or Eagle's syndrome. *Journal of cranio-maxillofacial surgery*. 2000; 28(2): 123-127.
3. Eagle WW. Symptomatic elongated styloid process report of two cases of styloid process-carotid artery syndrome with operation. *Archives of otolaryngology*. 1949; 49(5): 490-503.
4. Fusco DJ, Asteraki S, Spetzler RF. Eagle's syndrome: embryology, anatomy, and clinical management. *Acta neurochirurgica*. 2012; 154(7): 1119-1126.
5. Eagle WW. Elongated styloid processes: report of two cases. *Arch Otolaryngol*. 1937; 47:584-87.
6. Beder E, Ozgursoy OB, Ozgursoy SK, Anadolu Y. Three-dimensional computed tomography and surgical treatment for Eagle's syndrome. *Ear, nose & throat journal*. 2006; 85(7): 443-445.
7. Prasad KC, Kamath MP, Reddy KJM, Raju K, Agarwal S. Elongated styloid process (Eagle's syndrome): a clinical study. *Journal of Oral and Maxillofacial Surgery*. 2002; 60(2): 171-175.
8. Yavuz H, Caylakli F, Yildirim T, Ozluoglu LN. Angulation of the styloid process in Eagle's syndrome. *European archives of oto-rhino-laryngology*. 2008; 265(11): 1393-1396.
9. Piagkou M, Anagnostopoulou S, Kouladouros K, Piagkos G. Eagle's syndrome: a review of the literature. *Clinical Anatomy: The Official Journal of the American Association of Clinical Anatomists and the British Association of Clinical Anatomists*. 2009; 22(5): 545-558.
10. Rizzatti-Barbosa CM, Ribeiro MC, Silva-Concilio LR, Di Hipolito O, Ambrosano GM. Is an elongated stylohyoid process prevalent in the elderly? A radiographic study in a Brazilian population. *Gerodontology*. 2005; 22(2): 112-115.
11. Roy AK, Reader ST, Das SK, Mahmud SA, Readre NS. Case Study of Asymptomatic Eagle's Syndrome, *International journal of Current Research*. 2018;10(1): 64186-64189.
12. İlgüy M, İlgüy D, Güler N, Bayirli G. Incidence of the type and calcification patterns in patients with elongated styloid process. *Journal of international medical research*. 2005; 33(1); 96-102.
13. Rechtweg JS, Wax MK. Eagle's syndrome: a review. *American journal of otolaryngology*. 1998; 19(5): 316-321.
14. Eagle WW. Elongated styloid process: further observations and a new syndrome. *Archives of otolaryngology*. 1948; 47(5): 630-640.
15. Glogoff MR, Baum SM, Cheifetz I. Diagnosis and treatment of Eagle's syndrome. *Journal of Oral Surgery*. 1981; 39(12): 941-944.