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Eagle's Syndrome in Children: A Case Report

Serigne Tawa Ndiaye[1*], Cheikh Diéne Niang[2], Ciré Ndiaye[2], Mohameth Mbodj[1], Ndeye Fatou Sow[2], Amadou Sow[3], Faty Fall[2], Papa Moctar Faye[1], Ibrahima Diagne[4], Ousmane Ndiaye[1]

[1]Albert Royer National Children's Hospital, Dakar, Senegal
[2]Cheikh Anta Diop University, Dakar, Senegal
[3]Abass Ndao Municipal Hospital Center, Dakar, Senegal
[4]Gaston Berger University, Saint Louis, Senegal
Email: *mabeyend@gmail.com

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Abstract

Eagle's syndrome is a collection of symptoms caused by styloid process elongation or calcification of the stylohyoid ligament, measuring more than 2.5 centimeters. It is a radio-clinical entity characterized by a heterogeneous polymorphic symptomatology most often involving headaches, facial pain, dysphagia and a foreign body sensation in the throat. Its management is mainly surgical. It is a rarely diagnosed condition in children. Here we report a typical case of Eagle's syndrome which was diagnosed in a 14-year-old child with a history of chronic right unilateral pharyngeal discomfort, odynophagia and oropharyngeal foreign body sensation. CT scan showed a long left styloid process. The patient underwent surgical removal of the elongated styloid process externally. The outcome was favorable after surgery.

Keywords

Eagle's Syndrome, Styloid Process, Child

1. Introduction

Eagle's syndrome, or stylo-carotid syndrome, is defined as an elongation of the styloid process or ossification of the stylohyoid ligament measuring more than 2.5 centimeters. The disease was described in 1937 by an ear, nose and throat (ENT) specialist named Watt Eagle [1]. It is a radio-clinical entity characterized by its heterogeneous and polymorphic ENT symptomatology. The three-dimensional cervical CT scan is the examination of choice for diagnostic confirmation. It is mainly managed by intraoral or external surgery. It is a little known pathology that affects 4% of the general population and is rarely symptomatic before the age of 30 [2]. Thus in children, Eagle's syndrome is extremely rare. Indeed, to our knowl-

edge, only five cases have been reported in the literature. The main symptom was glossopharyngeal pain in most cases [3]. Here we present a new case diagnosed in a 14-year-old boy.

2. Observation

A 14-year-old boy with no particular medical or surgical history, good psychomotor development, and full vaccination status according to the Senegalese extended programme of vaccination. He is the eldest of five healthy children with no past medical family history. He consulted the emergency unit of the Albert Royer Children's Hospital for a symptomatology made of unilateral right oropharyngeal discomfort, odynophagia and foreign body sensation, and intermittent right retro-auricular cervicalgia triggered by head movements, evolving since 12 months. Physical examination revealed a weight of 45 kg, a height of 158 cm (−1 SD; median), a BMI of 18 (−1; median), a temperature of 36.7˚C or 98.06˚F, a heart rate of 98 bpm. Systematic inspection of the throat with a tongue depressor was unremarkable. A full ENT examination was requested which revealed a right subdigastric pain on cervicofacial palpation. Oropharyngeal palpation in the right tonsillar fossa revealed a painful tapering induration in the right tonsil area. The rest of the examination was unremarkable. The cervical ultrasound was normal. The 3D cervical CT scan showed a right styloid process measuring 60 mm in length, the tip of which terminates at the styloid bone and is flush with the pharyngeal wall, with right parapharyngeal nodular calcification (**Figure 1**). The diagnosis of Eagle's syndrome was retained. An external styloidectomy was performed (**Figure 2**). No intraoperative complication was found.

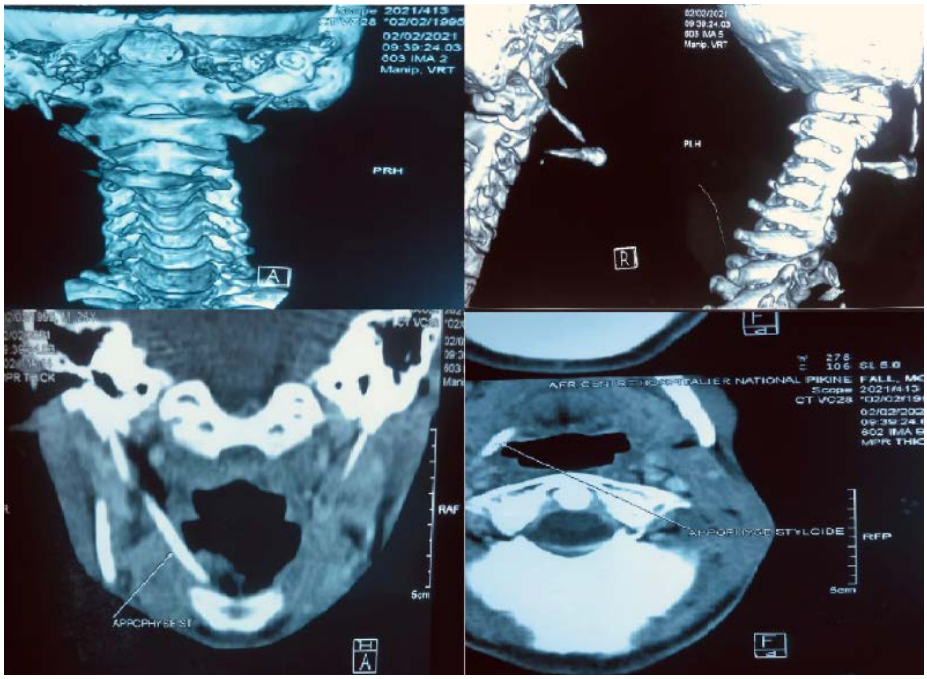

Figure 1. 3D scan images measuring the right styloid process at 60 mm (arrow).

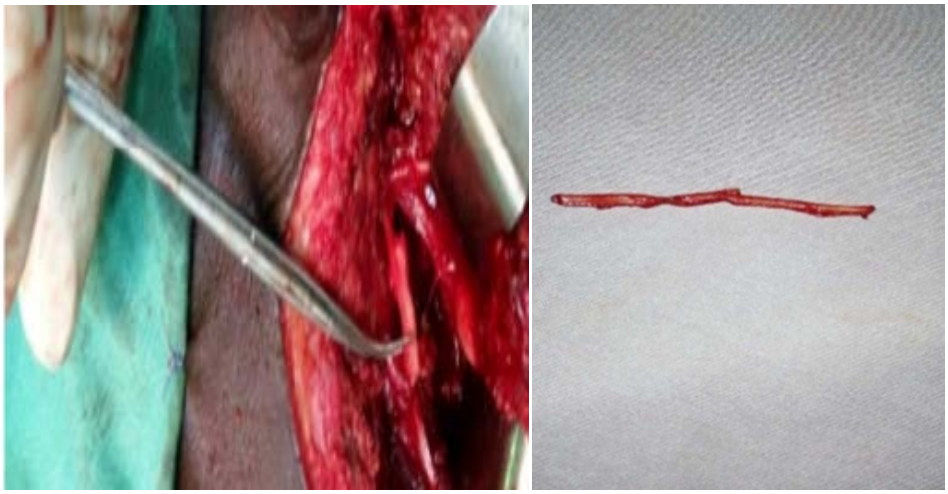

Figure 2. External approach to the styloid process.

However, after the surgical procedure, the patient suffered a transient peripheral facial paralysis which resolved rapidly after 10 days of oral anti-inflammatory treatment. Since then, the patient has been asymptomatic.

3. Discussion

The first mention of the pain syndrome associated with the elongated styloid process dates back to 1937, when it was described by Watt Weems Eagle [1]. The styloid process is a thin bony projection from the lower surface of the petrous portion of the temporal bone. This process originates from Reichert's cartilage of the second brachial arch and persists as a structure from the base of the skull to the lower horn of the hyoid, passing between the internal and external carotid arteries. It is also adjacent to the glossopharyngeal and vagus nerves [4]. The elongated styloid process causes compression of the cranial nerves (trigeminal V, facial VII, glossopharyngeal IX) and its branches, and results in pain on the face and with swallowing, and a sensation of a foreign body in the throat [5]. The incidence in the general population is 0.16% with a female predominance, and it is rarely symptomatic before adolescence [6]. However, a few paediatric cases have been reported in the literature [3] [7] [8] [9].

In this case of Eagle's syndrome in children, as in most reported paediatric cases, the circumstances of discovery are variable [3] [8] [9] [10] (**Table 1**). Indeed, Eagle described three types [1]. The first, "classic styloid syndrome", occurs mainly after neck trauma or tonsillectomy, with dysphagia, otalgia and pharyngeal foreign body sensation, the second, with pain along the external carotid artery and the third is asymptomatic. The symptoms of the classical syndrome were present in our patient, but no history of tonsillectomy was found.

The differential diagnosis of Eagle's syndrome in children includes third molar involvement, oropharyngeal foreign body, oropharyngeal tumours, chronic tonsillitis, trigeminal neuralgia, temporomandibular joint anomalies [8]. But the diagnosis is established on clinical examination if palpation of the styloid process in

Table 1. Reported cases of Eagle syndrome in children.

Author and year	Age	Gender	Circumstances of discovery	Diagnostic means
Holloway *et al.*, 1991	5	Female	Sensation of discomfort in the throat	Radiography
Riaño *et al.*, 1999	10	Female	Recurrent torticollis and dysphagia	Radiography
Gárriz M *et al.*, 2016	9	Male	Paraesthesia in the hemipharynx Pain in the tonsil fossa	3D scan
Our patient	14	Male	Unilateral right oropharyngeal discomfort Sensation of a foreign body in the oropharynx	3D scan

the tonsillar fossa reproduces the pain and the lidocaine injection test in this fossa relieves the pain in few minutes [11]. In our patient, this test came back positive.

The diagnosis of Eagle syndrome is confirmed by imaging. Several imaging techniques have been used, but currently CT scan is the examination of choice. 3D CT scan reconstruction of the neck allows the length of the styloid process and ossified stylohyoid ligament and the vascular-nervous relationships to be determined [2] (**Table 1**).

The authors state that the treatment of choice is surgical, based on either intraoral or external removal of the calcified process and release of the compressed vascular and nerve structures [12]. In our patient, a styloidectomy was performed externally. The postoperative course is generally simple with disappearance of the symptoms [13] as was the case with our patient.

4. Conclusion

Eagle syndrome is a rare radio-clinical entity in the paediatric population. As found in this case, the possibility of this diagnosis should not be overlooked in the setting of persistent unilateral pharyngeal discomfort in children with a normal endobuccal examination. The use of CT scans with three-dimensional reconstruction can confirm the diagnosis. Treatment is mainly surgical.

Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this paper.

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