# Eagle syndrome: rare cause of chronic neck pain – A case report and review of the literature

Oghenetejiri D Ogholoh,[1,2] Ngozi Nzekwue,[3,4] Joyce Ikubor[1,2]

**Abstract**

**Background:** Eagle's syndrome (ES) is a rare clinical disorder characterised by the aberrant elongation and ossification of the styloid process (SP), a narrow bony protrusion at the base of the skull or the calcification/ossification of the stylohyoid ligament and which may result in cervicofacial pain and dysphagia. It is also referred to as stylalgia, styloid syndrome, stylohyoid syndrome, or stylohyoid complex syndrome.

**Methods:** We report the case of a 58-year-old man who presented with an insidious onset of chronic bilateral neck pain of three years duration, worse on the right side, with recent progression and associated dysphagia. There is no prior history of previous neck surgery or trauma.

Craniofacial computed tomography (CT) done with axial, reformatted coronal and three-dimensional (3D) volume rendered images revealed elongation of the temporal bone styloid processes, with associated ossification of the stylohyoid ligaments bilaterally, extending inferiorly to abut the hyoid bone, and measuring about 6.60cm and 6.39cm in length for the right and left respectively (normal <3cm). There is no gross carotid arterial or jugular venous compression noted.

**Results:** A diagnosis of bilateral Eagle's syndrome was made. The patient declined surgical intervention and was managed conservatively with non-steroidal anti-inflammatory drugs, gabapentin, and amitriptyline. He was subsequently lost to follow-up.

**Conclusion:** To the best of our knowledge, this is one of the earliest reported radiologically diagnosed case of Eagle's syndrome in Warri, Delta State, Southern Nigeria.

**Keywords**: Styloid process, stylohyoid ligament, ossification, neck pain.

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## Introduction

Eagle's syndrome (ES) is a rare clinical disorder characterised by the aberrant elongation and ossification of the styloid process (SP), a narrow bony protrusion at the base of the skull, or the calcification/ ossification of the stylohyoid ligament.[1–6] It is also referred to as stylalgia, styloid syndrome, stylohyoid syndrome, or stylohyoid complex syndrome.[1,2,5,7]

The syndrome was named after the American otolaryngologist, Watt W. Eagle, who originally detailed it in 1937, although anatomical observations of the disease dates back to 1652.[1–4,8] It is a rare condition estimated at 4-8 per 10,000 people. This condition is predominantly diagnosed in women, with a 3:1 female-to-male ratio, usually between the ages of 30 and 60.[1,4,7,8] It is estimated that around 4% of most populations possess an extended styloid process; nevertheless, the majority remain asymptomatic, with only 4% to 10% of these individuals (accounting for less than 0.2% of the total population) exhibiting symptomatic ES.[1–5,9]

Eagle syndrome has a wide range of clinical presentations, ranging from acute pain in the throat or neck, otalgia, dysphagia, trismus and the sensation of a foreign body in the throat. Additional symptoms may comprise cephalalgia, facial discomfort and cervical pain. This varied and often complex presentation of symptoms may lead to inaccurate diagnosis of this condition.[1,2,4,7,10]

Eagle classified this syndrome into two categories: classic type and carotid artery type. The classic form of ES is frequently noted post-tonsillectomy, with the predominant manifestation being a sensation of a foreign body in the oropharyngeal area, accompanied by a dull, persistent discomfort devoid of a lancinating (i.e., neuralgic) character while swallowing. The carotid artery type manifests upon compression of the internal or external carotid artery by an elongated SP, but may also be associated with further dysfunction and symptoms resulting from stimulation of the sympathetic nerve plexus.[2,4,10]

The etiology of ES is thought to be complex and is still a topic of current clinical discussion, since it is unclear exactly what causes styloid process elongation.[1,2,11] Some theories include persistence and ossification of embryonic Reichert's cartilage, calcification of the stylomandibular ligament, and progressive osseous overgrowth which results in the styloid process elongating.[1,3,6,8] Additional contributing factors include genetic predisposition, endocrine changes particularly in postmenopausal women, and abnormal development of the stylohyoid complex, which has also been associated with atlanto-occipital joint malformations.[2,6] Procedures such as tonsillectomy

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may irritate the stylohyoid ligament and styloid process, triggering inflammatory changes, which may result in reactive ossification of the ligament and subsequent elongation of the styloid process.[2]

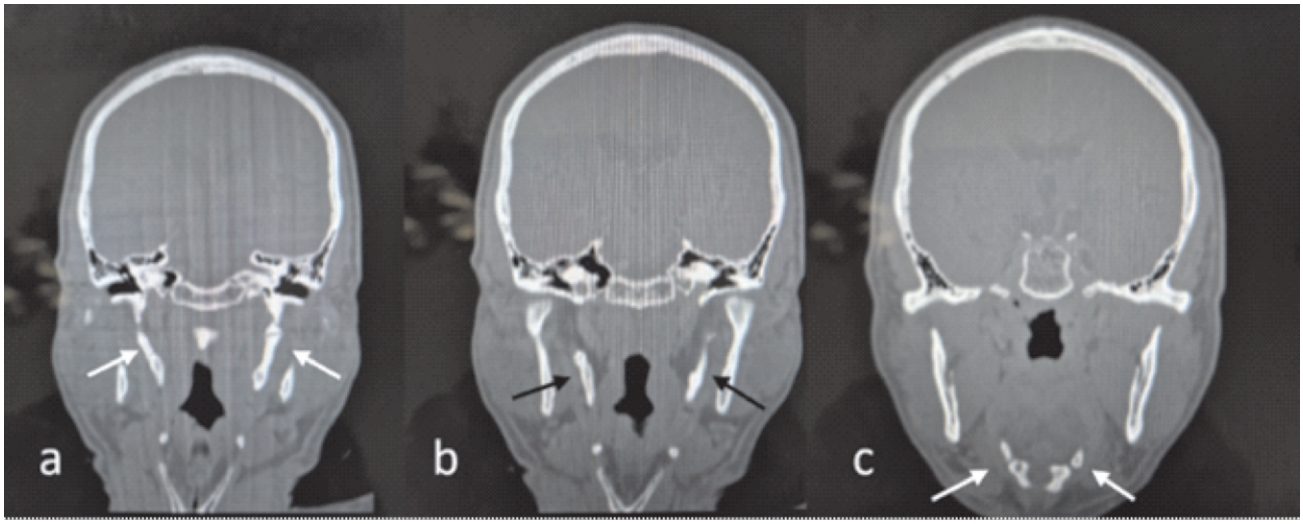
**Case Report**

We present a 58year old male presenting with insidious onset of chronic neck pain of about 3years duration, radiating to the face and neck, which was worse on the right. Pain was initially mild, but it gradually increased in severity, with associated recent onset difficulty in swallowing and feeling of foreign body sensation in the throat. There was no history of dizziness or fainting spells with changes in head or neck movements. There was also no prior history of previous neck surgery or trauma. He had taken several over the counter painkillers, predominantly non-steroidal anti-inflammatory drugs which offered transient relief.

Physical examination revealed a middle-aged man not in any obvious distress. He was obese and a known hypertensive (for about 8years) with good drug compliance. There was mild tenderness on moving the neck; however, the cervical, thoracic and lumbar spine showed normal flexion/extension movements. Other systemic examinations were not contributory.

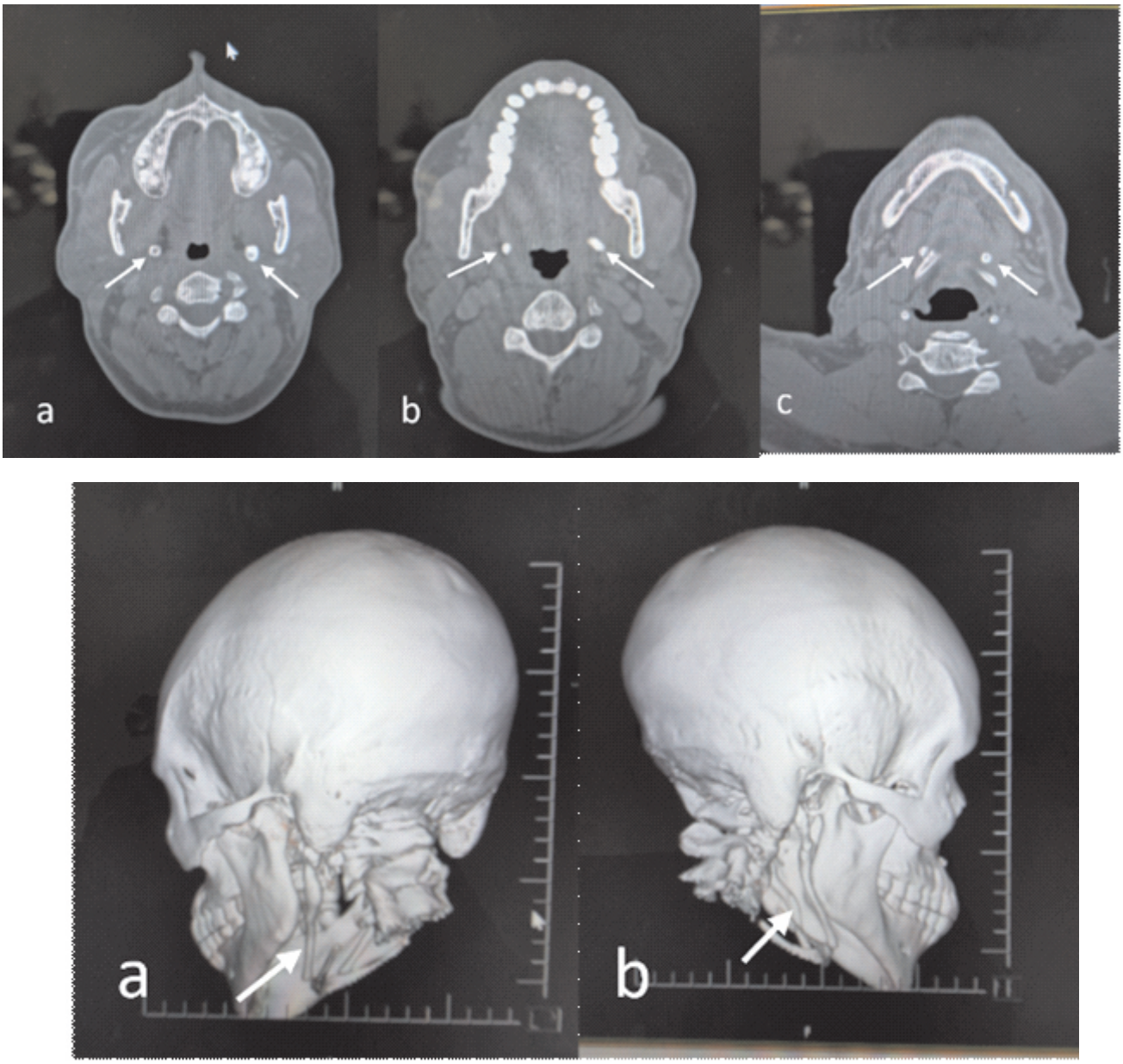
All of his laboratory blood test results (serum calcium, phosphorus, alkaline phosphatase, erythrocyte sedimentation rate, C-reactive protein, serum electrolytes and urea and complete blood count) were normal.

Radiologic evaluation was done using Computed tomography (Siemens Somatom go.Now 32-slice scanner). Craniofacial computed tomography (CT) was carried out from the vertex to the mid cervical region. The axial, reformatted sagittal and coronal images as well as three-dimensional (3D) volume rendered images were acquired, and these revealed elongation of the temporal bone styloid processes, with associated ossification of the stylohyoid ligaments bilaterally, extending inferiorly to abut the hyoid bone, as depicted in Figures 1 to 3, and measuring about 6.60cm and 6.39cm in length for the right and left respectively (normal <3cm). There is no gross carotid arterial or jugular venous compression noted.




Figures 1a to 1c: Coronal reformatted Craniofacial CT images (bone window). Figures 1a and 1b show elongation of the temporal bone styloid processes (white arrows), with associated ossification of the stylohyoid ligaments bilaterally (black arrows), while Figure 1c shows the distal aspect of both stylohyoid ligaments extending inferiorly to abut the hyoid bone (white arrows).

Figures 2a to 2c are axial Craniofacial CT images (bone window) taken at different levels, depicting elongation of the temporal bone styloid processes and associated ossification of the stylohyoid ligaments bilaterally, as well as showing the descent inferiorly of the distal aspect of both stylohyoid ligaments extending inferiorly to abut the hyoid bone (white arrows).




Figures 3a and 3b are volume rendered 3-D reformatted Craniofacial CT images, showing elongation of the temporal bone styloid processes and associated ossification of the stylohyoid ligaments bilaterally, as well as extension inferiorly to abut the hyoid bone (white arrows).

**Literature Review**

Previous case reports have demonstrated the range of clinical spectrum and diagnostic challenges associated with ES. Alaqeeli and co- researchers[1] reported a case of a 41-year-old male who presented with a three years history of headache, dysphagia, neck pain radiating to the ear, and tinnitus. The diagnosis was determined by utilizing a 3D computed tomography (CT) scan which revealed a markedly elongated and deformed left styloid process with a length of 8.5cm. A transcervical styloidectomy was conducted, with post operative examination establishing a complete resolution of the symptoms and post-surgery imaging confirming a surgically shortened styloid process.

Similarly, Boucher et al[2] described a case of a 51-year-old woman with a six-year history of retromandibular pain, foreign body sensation, and odynophagia. The patient underwent a cone beam CT scan that showed an asymmetric elongation of the styloid process with pseudo-articulation on the symptomatic side with respective maximal dimensions of 37.3 × 13.1 mm and 32.5 ×10.5 mm, located above the

inferior border of the mandible. A definitive diagnosis of ES was made based on the patient's medical history, clinical findings and imaging reports. A left stylectomy was performed, with almost complete pain relief being reported during the 3 months follow up visit. However, complete relief of symptoms was noted after 4 years with several follow up visits.

In contrast, Garg et al[3] presented a 37-year-old patient with bilateral neck pain radiating to the ears, which worsened on movement of the neck. Generally, no abnormality was reported upon physical and temporomandibular joint (TMJ) examination. Orthopantomogram and cone beam CT scan revealed bilateral styloid elongation, measuring over 4cm in length. Management of the patient was initiated with gabapentin, amitriptyline and paracetamol for six weeks. Afterwards, the patient reported reduction in pain, but however the pain during neck movement remained consistent. Continuation of the treatment was advised for another six weeks. The pain drastically reduced after three months with pain free intervals in the three month follow up.

Abdul Halim et al[4] described the case of a 43-year-old female who visited their health facility with symptoms of chronic neck pain, imbalance, and a persistent floating sensation. Prior to presentation at the clinic, the patient had been diagnosed with cervical spondylosis and was prescribed parenteral tramadol, which significantly improved her neck pain. After a short while, the pain reoccurred and she was referred to an orthopedic team that requested for a magnetic resonance imaging (MRI) scan of the cervical and lumbosacral spine which showed features of mild disc osteophyte complex, and cervical and lumbar spondylosis. After a month the neck pain progressively worsened, she was then referred to an otorhinolaryngologist who made a differential diagnosis of ES based on her previous medical history. Furthermore, a CT scan of the neck was performed which demonstrated bilateral elongation of the styloid processes. Considering the chronicity and severity of the symptoms, surgical management with bilateral tonsillectomy and styloidectomy was planned.

In Calabar, Nigeria, Mgbe and colleagues,[7] reported two cases with contrasting management approaches, one treated surgically and the other conservatively, further underscoring the heterogeneity of presentation and treatment response.

Vascular complications of ES have also been highlighted. Stopińska and Domitrz[10] described patients with the stylocarotid variant presenting with recurrent headaches and carotid artery compression, while Xhaxho et al.[9] reported an acute ischaemic stroke secondary to internal carotid artery dissection caused by an elongated styloid process, necessitating delayed surgical intervention.

In summary, these studies reported have demonstrated that ES presents with several clinical symptoms, which most times results in delayed diagnosis, and also requires a tailored diagnostic and therapeutic approach, with 3D CT being a preferred diagnostic modality and management planning.

**Treatment**

Patients with ES may be managed medically or surgically. The medical therapeutic options include pain control using oral analgesics, transpharyngeal infiltration with steroids, and administration of tonsillar fossa local anesthetic agents.[12] This index patient was managed medically with NSAIDs, gabapentin and amitriptyline. However, the surgical intervention is the treatment of choice, and it is styloidectomy or stylectomy, which may be done via the transoral or extraoral routes.

**Conclusion**

Eagle syndrome is a rare cause of facial or cervical pain. Therefore, it should be considered in the differential diagnosis of severe facial or neck pain with poor response to painkillers.

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