

**LETTER TO THE EDITOR****TO THE EDITOR****Is Eagle Jugular Syndrome an Underestimated Potentially Life-Threatening Disease?**

**Keywords:** Eagle syndrome, Jugular vein compression, Headache, Deep vein thrombosis, Pulmonary embolism

We have read with interest the article titled “Eagle Syndrome as a Cause of Cerebral Venous Sinus Thrombosis” by Fu-Liang Zhang et al. published in the May 2019 issue of *The Canadian Journal of Neurological Sciences*.<sup>1</sup>

The authors describe a case of Eagle syndrome in a 15-year-old teenager with a 2-month history of headache that was caused by left transverse-sigmoid sinus thrombosis due to compression of the left jugular vein by an elongated styloid process (SP). This is quite unusual, since the classic Eagle syndrome is primarily caused by compression of the internal carotid artery, resulting in a wide spectrum of clinical and neuroradiological findings from cervicofacial pain to cerebral ischemia.<sup>2,3</sup>

The paper by Fu-Liang Zhang deserves merit since it describes an unusual effect of the presence of elongated SPs, i.e., compression of the jugular vein, leading to some interesting considerations regarding what has been defined as “Eagle jugular syndrome.”<sup>4</sup> It is well known that prolonged venous compression can lead to the development of deep venous thrombosis (DVT). DVT is much more frequent in the lower limbs, but it may occur in any section of the venous system, including arms, abdominal, and also the jugular veins.<sup>5</sup> For instance, thrombosis of the subclavian vein at the costoclavicular junctions, also known as Paget-Schroetter syndrome, is caused by intermittent venous compression due to muscular stretch in the thoracic outlet and may potentially result in pulmonary embolism initially classified as of unknown origin.<sup>6</sup>

Similarly, it has already been demonstrated that compression of the jugular vein caused by an elongated SP may result in jugular thrombosis, thus configuring the case of “Eagle jugular syndrome.” In such a scenario, clinical manifestations may vary from migraine to cerebral venous thrombosis.<sup>4,7</sup>

In light of such evidence, it is reasonable to hypothesize that jugular vein thrombosis may also lead to pulmonary embolism. It has been reported that in approximately 20% of cases of pulmonary embolism, the source of emboli cannot be identified, representing the so-called “isolated pulmonary embolism,” which is frequently caused by unusual forms of venous thrombosis and thrombophilia.<sup>8</sup> It could be speculated that jugular vein thrombosis in Eagle syndrome may be responsible for some of these cases of pulmonary embolism, given that this eventuality has been poorly investigated.

Our suggestion is to include also the investigation of jugular vein compression due to elongated SPs in cases of pulmonary embolism of unknown origin. As previously reported, we strongly support the necessity of a dynamic evaluation of compression of neck structures due to elongated SPs in Eagle syndrome.<sup>9</sup> The diagnostic accuracy of computed tomography angiography (CTA)/magnetic resonance angiography (MRA) can be increased through a “dynamic” evaluation of the spatial relationship

between SPs and neck vascular structures simply performing acquisitions using different orientations of the head. This could better identify the compression that occurs only with a specific head orientation (e.g. rotation) but not in a “neutral” head position.

In conclusion, we think that, amongst others, Eagle syndrome may result in compression and thrombosis of the jugular vein, thus causing life-threatening clinical manifestations, including cerebral venous thrombosis, as reported by Fu-Liang Zhang et al.,<sup>1</sup> along with isolated, recurrent, pulmonary embolism events, the origin of which may be misdiagnosed. The dynamic exploration of the spatial relationship between elongated SPs and jugular veins through CTA and/or MRA may lead to a prompt diagnosis of this eventuality and to an effective treatment of these patients. Further investigations are needed to confirm or deny our hypothesis.

**DISCLOSURES**


The authors have no conflicts of interest to declare.

**STATEMENT OF AUTHORSHIP**

ENS conception of the work; drafting the work and revising it critically for important intellectual content; final approval of the version to be published; agreement to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved. GR conception of the work; drafting the work and revising it critically for important intellectual content; final approval of the version to be published; agreement to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved. AG conception of the work; final approval of the version to be published; agreement to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved. FSDP conception of the work; final approval of the version to be published; agreement to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved.

\**Enrico Nastro Siniscalchi*

*Division of Maxillofacial Surgery, BIOMORF Department,  
University of Messina, Messina, Italy*

\**Giovanni Raffa* 

*Division of Neurosurgery, BIOMORF Department, University of  
Messina, Messina, Italy*

*Antonino Geranò*

*Division of Neurosurgery, BIOMORF Department, University of  
Messina, Messina, Italy*

*Francesco Saverio De Ponte*

*Division of Maxillofacial Surgery, BIOMORF Department,  
University of Messina, Messina, Italy*

\*The authors equally contributed as first authors.

*Correspondence to:* Giovanni Raffa, Division of Neurosurgery, BIOMORF Department, University of Messina, Via Consolare Valeria 1, 98125, Messina, Italy. Email: [giovanni.raffa@unime.it](mailto:giovanni.raffa@unime.it)

#### REFERENCES

1. Zhang F-L, Zhou H-W, Guo Z-N, Yang Y. Eagle syndrome as a cause of cerebral venous sinus thrombosis. *Can J Neurol Sci.* 2019;46(3):344–5.
2. Chung C-P, Chao AC, Hsu H-Y, Lin S-J, Hu H-H. Decreased jugular venous distensibility in migraine. *Ultrasound Med Biol.* 2010;36:11–6.
3. Galletta K, Granata F, Longo M, et al. An unusual internal carotid artery compression as a possible cause of Eagle syndrome – a novel hypothesis and an innovative surgical technique. *Surg Neurol Int.* 2019;10:174.
4. Galletta K, Siniscalchi EN, Ciccio M, Velo M, Granata F. Eagle syndrome: a wide spectrum of clinical and neuroradiological findings from cervico-facial pain to cerebral ischemia. *J Craniofac Sur.* 2019;30:e424–8.
5. Illig KA, Doyle AJ. A comprehensive review of Paget-Schroetter syndrome. *J Vasc Sur.* 2010;51:1538–47.
6. Martinelli I. Unusual forms of venous thrombosis and thrombophilia. *Pathophysiol Haemost Thromb.* 2002;32:343–5.
7. Porfidia A, Porceddu E, Feliciani D, et al. Differences in clinical presentation, rate of pulmonary embolism, and risk factors among patients with deep vein thrombosis in unusual sites. *Clinic Appl Thromb Hemost.* 2019;25:1076029619872550.
8. Siniscalchi EN. Dynamic imaging in suspected Eagle syndrome. *Eur Arch OtorhinoLaryngol.* 2020;277:307.
9. Zamboni P, Scerrati A, Menegatti E, et al. The eagle jugular syndrome. *BMC Neurol.* 2019;19:333.