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Cavernous Malformation of the Trigeminal Nerve Manifesting as Facial Hypoesthesia: A Case Report and Literature Review

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Abstract

Cavernous malformations are relatively common vascular lesions of the central nervous system. These lesions can occur along the entire neuroaxis; however, they are very rare in cranial nerves. Trigeminal cavernous malformations can be classified into four types, according to their location: type G (Gasserian ganglion), type C (between the cisternal and intra-axial portions), type P (intra-axial trigeminal nerve root in the pons), and type S (spinal tract of the trigeminal nerve root below the medulla oblongata). In our review, we found a total of 27 cases of cavernomas affecting the trigeminal nerve. Most cases presented with pain as the main symptom. We present the case of a 23-year-old man with facial hypoesthesia over the left V2 region that had started three days before, with progressive intensity. No headache or facial pain was noted. His magnetic resonance imaging (MRI) scans showed a vascular lesion in the left pons involving the fibers of the trigeminal nerve. Since the patient was young and symptomatic and the lesion was surgically accessible, a retrosigmoid craniotomy was devised for resection, with the aid of neurophysiological monitoring. Histology revealed a cavernous malformation. The patient had an uneventful recovery, without new neurological deficits, and post-op MRI confirmed complete resection.

Categories: Neurosurgery
Keywords: brainstem cavernoma, cavernoma, cavernous angioma, cavernous malformation, cranial nerve, fifth cranial nerve, fifth cranial nerve palsy, pontine cavernoma, trigeminal nerve

Introduction

Cavernous malformations (CMs), also known as cavernous angiomas, cavernomas, or cryptic vascular malformations, are low-flow vascular lesions primarily found in the brain and in the spine less often. The supratentorial lesions account for approximately 65-80% of cases [1]. CMs can occur along the neuraxis, occurring in areas such as the brainstem, cerebellum, and cerebral hemisphere; however, cavernoma involving the cranial nerves is very rare [2-4].

Cavernomas have an estimated prevalence of approximately 0.4-0.9% in the general population [3]. In many instances, the CMs remain clinically silent. Indeed, large clinical-radiological series indicate that up to 40% of individuals with CMs are asymptomatic. Symptomatic presentations often include seizures, headaches, hemorrhage, or focal deficits [1].

Cerebral CMs are abnormal, low-flow, thin-walled, enlarged vascular channels with no intervening brain parenchyma. Cranial nerve CMs tend to have an aggressive clinical course, with a tendency to progressive neurological deficits following intralesional hemorrhage or, less commonly, subarachnoid hemorrhage [5]. In general, after surgery, patients with trigeminal nerve CMs experience improvement in the focal deficits caused by mass effect; however, persistent sensory deficits in the postoperative period are common [6].

Trigeminal neuralgia can be classified into three types: idiopathic, classical, and secondary. Idiopathic trigeminal neuralgia has no apparent cause. Classical trigeminal neuralgia is associated with neurovascular compression. Secondary trigeminal neuralgia is generally caused by multiple sclerosis, herpes zoster, or cerebellopontine angle tumors, such as meningiomas, vestibular schwannomas, epidermoid cysts, and cholesteatomas [4].

There are limited cases of CMs involving the fifth nerve that have been documented in the literature. The majority manifested clinically as trigeminal neuralgia. This case report details a rare presentation of a CM affecting the fifth cranial nerve, marked by progressive sensory decline, expanding our understanding of the varied clinical manifestations of CMs affecting cranial nerves. In addition, we conducted a review of all previously reported cases of cavernoma involving the trigeminal nerve published in the literature. In total, we identified 27 documented cases.

Case Presentation

A 23-year-old man with no significant past medical history presented with left-sided hypoesthesia in the V2 distribution that had begun three days earlier and progressively worsened, consistently respecting the midline. The patient denied headache, facial pain, trauma, recent infections, or vaccinations. He also reported no prior similar episodes and no visual disturbances, gait abnormalities, or motor deficits. On examination, the patient was alert with a Glasgow Coma Scale score of 15. Pupils were equal and reactive to light. Extraocular movements were intact, and visual fields were normal. Facial expression was preserved. Muscle strength was grade 5 throughout, reflexes were normal and symmetric, and no pathological reflexes were observed. Sensation in other body regions was intact.

Magnetic resonance imaging (MRI) revealed a CM in the left pons, consistent with the patient's symptoms (Figure *1*). Surgical resection of the lesion was subsequently performed.

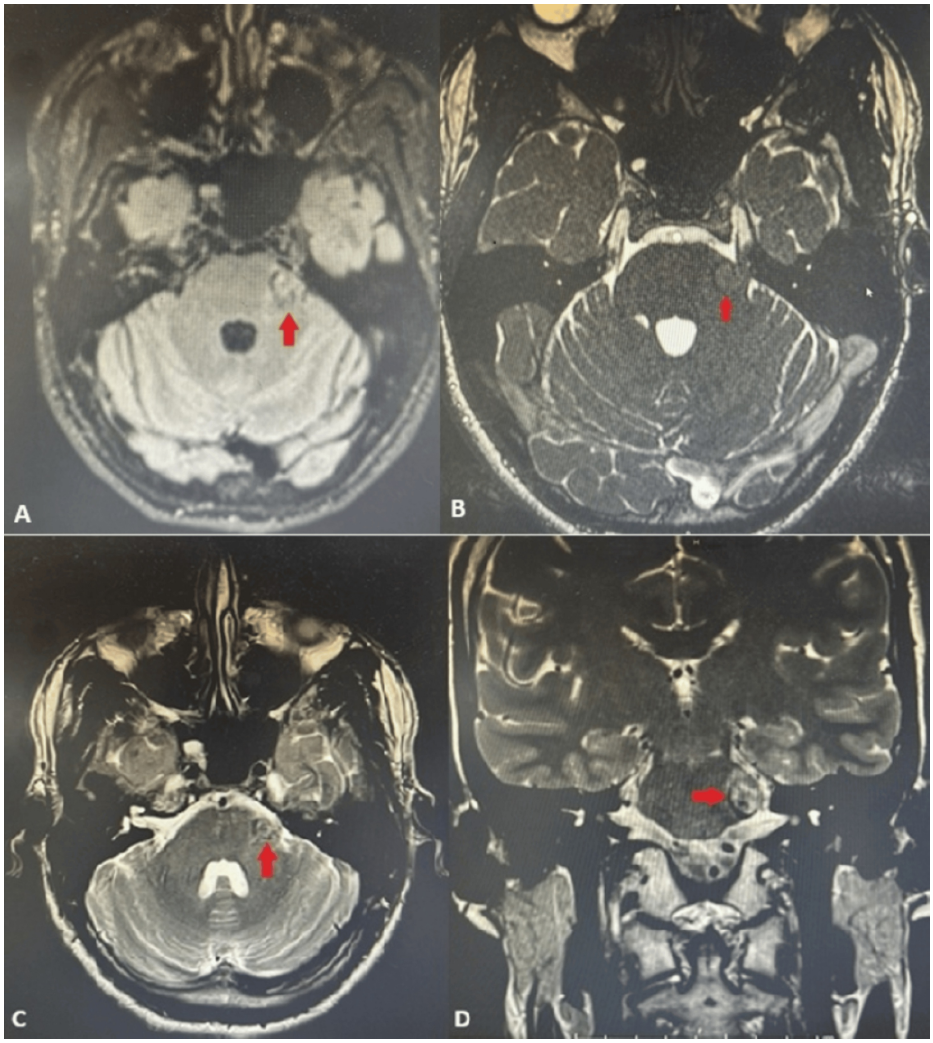

FIGURE 1: The cavernous malformation is indicated by the red arrow. (A) Axial T1-weighted MRI demonstrating a mildly heterogeneous lesion within the left pons, surrounded by a hypointense rim (red arrow). (B) Axial CISS-weighted MRI showing a well-defined hypointense lesion adjacent to the cisternal segment of the left trigeminal nerve (red arrow). (C) Axial T2-weighted MRI demonstrating a heterogeneous lesion with a peripheral hypointense rim (red arrow). (D) Coronal T2-weighted MRI demonstrating a heterogeneous lesion with a peripheral hypointense rim (red arrow).

MRI: magnetic resonance imaging; CISS: constructive interference in steady state

During surgery, the patient was placed in the supine position with the head rotated to the right. A left retrosigmoid approach was performed. Cranial nerves were identified with the aid of intraoperative electrophysiological monitoring. Neuronavigation was used to accurately localize the lesion. A pial incision was made at the site where the lesion was closest to the brainstem surface, and microsurgical resection of the cavernoma was carried out (Video *1*).

VIDEO 1: Operative video demonstrating the surgical resection of the lesion.

View video here: https://vimeo.com/1198820086?share=copy&fl=sv&fe=ci

Postoperatively, the patient progressed well without new neurological deficits. Postoperative imaging performed one month after surgery demonstrated complete resection of the lesion (Figure *2*). Histological analysis confirmed a central nervous system (CNS) CM (Figure *3*). After one year of follow-up, the patient remained clinically well.

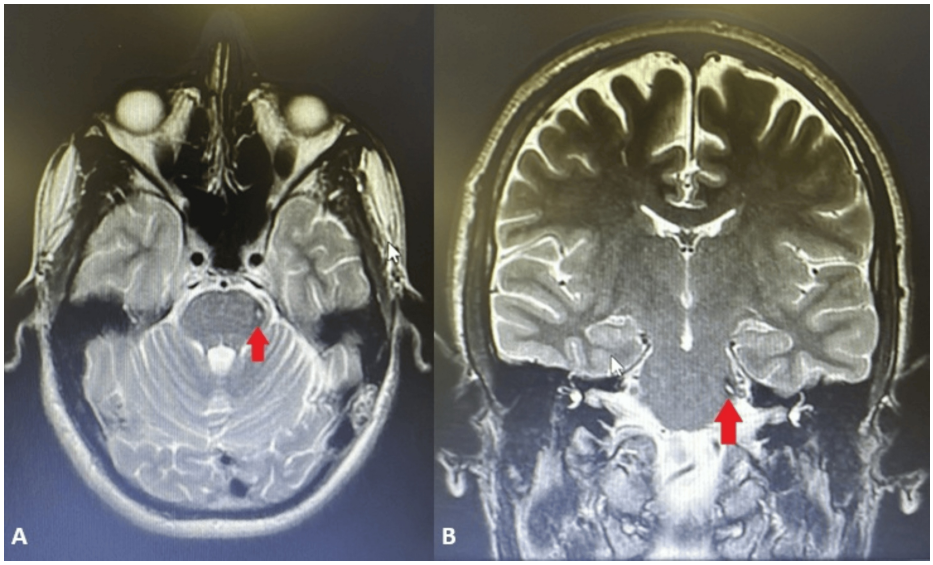

FIGURE 2: Postoperative T2-weighted MRI demonstrating expected postoperative changes in the left pontine region. No residual cavernous malformation is visible, confirming gross-total resection (red arrow). (A) Axial T2-weighted MRI. (B) Coronal T2-weighted MRI.

MRI: magnetic resonance imaging

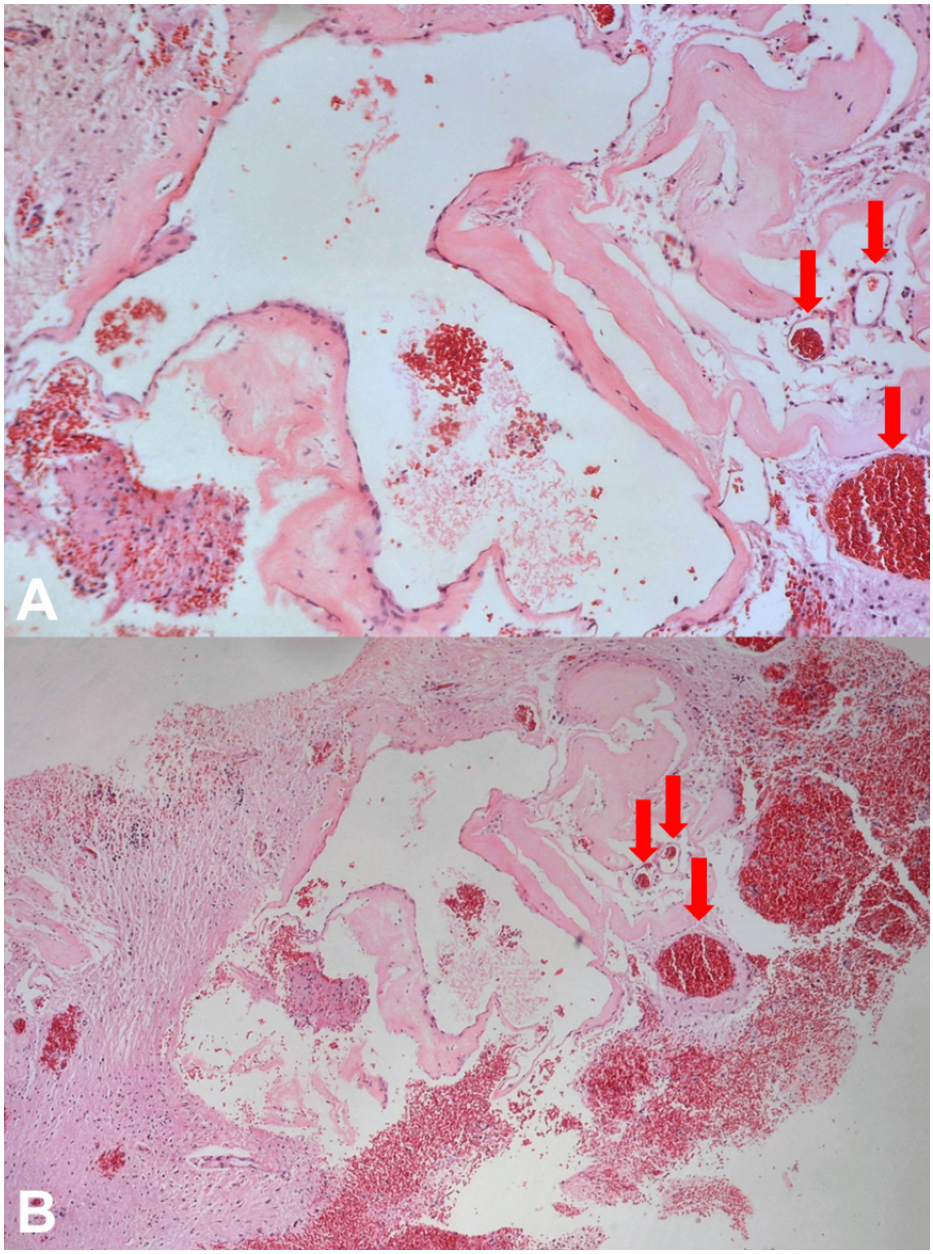

FIGURE 3: Histopathological findings of the lesion demonstrating multiple dilated, thin-walled vascular channels lined by flattened endothelial cells, associated with intraluminal blood products, consistent with a cavernous malformation (red arrow) (hematoxylin and eosin stain). (A) Original magnification ×100. (B) Original magnification ×40.

Discussion

Trigeminal CMs are extremely rare. We performed a literature review of previously reported cases. A search was performed in the PubMed and Embase databases using the following terms: ((cavernoma) OR (cavernous malformation) OR (cavernous angioma) OR (cavernous hemangioma)) AND ((trigeminal neuralgia) OR (trigeminal nerve)). Only case reports with full-text availability were included.

A total of 27 cases have been reported in the literature, and our patient represents the 28th reported case (Table *1*). The first case was described by Fehlings and Tucker in 1988 [7]. The youngest reported patient was 10 years old [5], whereas the oldest was 80 years old [8]. Considering all 28 cases, the mean age was 46.9 years, and the median age was 53 years. The gender distribution was balanced, with 15 males and 13 females. Eighteen patients presented with left-sided involvement, whereas 10 had right-sided lesions.

Author (year)	Gender	Age	Symptoms	Type	Treatment	Outcome
Fehlings and Tucker (1988) [7]	M	33	Left trigeminal neuralgia, facial hypoesthesia, and weakness of the masticatory muscles	G	Craniotomy	Cure. No pain, partial recovery of sensation, and persistence of weakness
Saito et al. (1989) [9]	F	45	Left trigeminal neuralgia, left limb weakness, and right C2-C7 hypoesthesia	S	Craniotomy	Cure. No pain. No motor weakness. Left facial and right body hypoesthesia
Kuntzer et al. (1992) [10]	F	29	Right trigeminal neuralgia, facial hypoesthesia, and weakness of the masticatory muscles	G	Craniotomy	Cure. No pain. Persistence of weakness and hypoesthesia
Komiyama et al. (1993) [11]	M	59	Left facial hypoesthesia, ataxia, and weakness of the masticatory muscles	P	Conservative treatment (patient declined surgery)	Recovery of ataxia. Hypoesthesia did not improve
De Benedittis (1996) [12]	M	62	SUNCT syndrome on the left	P	Craniotomy	Death
Shimpo (2000) [13]	M	67	Right trigeminal neuralgia and hypoesthesia	P	Pharmacotherapy (carbamazepine)	Effectiveness. No pain
Deol et al. (2001) [14]	M	17	Left facial hypoesthesia	G	Craniotomy	Cure. No pain
Vitek and Tettenborn (2002) [15]	M	61	Left trigeminal neuralgia	P	Pharmacotherapy (gabapentin)	Effectiveness. No pain
Deshmukh et al. (2005) [2]	F	52	Left trigeminal neuralgia, hypoesthesia, dizziness, and mild hearing loss	C	Craniotomy	Cure. No pain or dizziness. Facial hypoesthesia and hearing loss did not improve
Mascarenhas et al. (2006) [16]	M	54	Left trigeminal neuralgia and hypoesthesia	G	Craniotomy	Cure. Partial recovery of pain and hypoesthesia
Stellmann et al. (2007) [17]	F	55	Left trigeminal neuralgia, hypoesthesia, and ptosis	S	Spontaneously disappeared	Cure. No pain. No ptosis. Slight hypoesthesia persisted
Seçkin et al. (2009) [18]	M	56	Right trigeminal neuralgia and hypoesthesia	G	Craniotomy	Cure. No pain. Hypoesthesia did not improve
Cenzato et al. (2010) [19]	M	45	Left trigeminal neuralgia	P	Craniotomy	Cure. No pain
Cho et al. (2011) [20]	M	20	Right facial hypoesthesia. Palsies of III, IX, X, and XII cranial nerves on the right	C	Craniotomy	Cure. Cranial nerve palsies fully resolved, while hypoesthesia partially improved
Pereira de Morais et al. (2012) [21]	F	49	Right trigeminal neuralgia	C	Craniotomy	Cure. No pain
Adachi et al. (2014) [3]	M	62	Left trigeminal neuralgia and hypoesthesia	C	Craniotomy	Cure. No pain. Left facial hypoesthesia remained
Shuhui et al. (2016) [22]	F	61	Right trigeminal neuralgia, cervical pain, and right cervical itch	S	Surgery (C2 laminectomy)	Cure. No pain
Frossard et al. (2016) [23]	F	56	Initial left trigeminal neuralgia, later facial anesthesia, masticatory weakness, and gait ataxia	G	Craniotomy	Small stable residual lesion. No additional clinical data postoperatively
Giacobbo						

Scavo et al. (2018) [24]	F	62	Left trigeminal neuralgia, hypoesthesia, and gait ataxia	P	Craniotomy	Cure. No pain, hypoesthesia, or gait ataxia
Pease et al. (2019) [8]	F	80	Left trigeminal neuralgia and hypoesthesia	P	Radiosurgery	Effectiveness. Pain partially improved, while hypoesthesia remained stable
Zhang et al. (2020) [25]	M	37	Right trigeminal neuralgia and hypoesthesia	P	Percutaneous balloon compression	Effectiveness. No pain. Slight hypoesthesia persisted
Thompson et al. (2022) [5]	F	10	Left otalgia and trigeminal neuralgia	G	Craniotomy	Cure. No pain. Mild residual V1 sensory loss
Liu et al. (2022) [4]	F	29	Right trigeminal neuralgia and mild hearing loss	C	Craniotomy	Cure. No pain, but mild hearing loss persisted and right facial hypoesthesia developed
Harty et al. (2023) [26]	F	11	Left otalgia, headache, and syncope	C	Craniotomy	Cure. Facial hypoesthesia persisted
Wu et al. (2023) [27]	M	59	Left trigeminal pain, hypoesthesia, left facial weakness, speech slurring, and lack of right-sided coordination in the limbs	G and lesion in the left insular lobe	Craniotomy	Cure. No facial pain, significantly reduced speech slurring, and 3/5 right upper limb muscle strength
Connelly et al. (2024) [28]	F	49	Right trigeminal pain and hypoesthesia	S	Craniotomy	Cure. Partial improvement of pain
Hanks et al. (2025) [6]	M	70	Right trigeminal hypoesthesia, hearing loss, and lateral gaze nystagmus of the right side	C	Craniotomy	Cure. Facial hypoesthesia persisted. Hearing loss and nystagmus improved
Our case (2026)	M	23	Left facial hypoesthesia	P	Craniotomy	Cure. Slight hypoesthesia

TABLE 1: Clinical features and management of all reported cases of trigeminal nerve cavernomas in the literature.

"Effectiveness" was defined as successful symptom relief without lesion removal. "Cure" was defined as complete surgical resection of the lesion.

F: female; M: male; SUNCT: short-lasting unilateral neuralgiform headache attacks with conjunctival injection and tearing; type G: Gasserian ganglion; type C: between the cisternal and intra-axial portions; type P: intra-axial trigeminal nerve root in the pons; type S: spinal tract of the trigeminal nerve root below the medulla oblongata

Trigeminal CMs can be classified into four types, according to their location: type G (Gasserian ganglion), type C (between the cisternal and intra-axial portions), type P (intra-axial trigeminal nerve root in the pons), and type S (spinal tract of the trigeminal nerve root below the medulla oblongata) [3]. In total, there were seven cases of type C, eight cases of type G, nine cases of type P, and four cases of type S. Most cases presented with trigeminal neuralgia. Interestingly, our case presented only with hypoesthesia.

For cases that did not undergo surgical treatment, including medical therapy, percutaneous balloon compression, and radiosurgery, the term "effectiveness" was used to indicate successful symptom relief without lesion removal. In contrast, the term "cure" was reserved for cases in which complete surgical resection of the lesion was achieved.

Of all reported cases, 22 underwent neurosurgery, two received pharmacotherapy, one underwent percutaneous balloon compression, one was treated with radiosurgery, one showed spontaneous improvement, and one underwent conservative management due to refusal of surgical treatment. For the treatment of our case, we chose surgical intervention because it offers a definitive approach with the potential for complete lesion removal. It is important to note that the natural history of CMs in the brainstem is worse than in other CNS locations. Brainstem CM has a higher hemorrhage rate and is more likely to hemorrhage again than CMs located elsewhere [3]. In addition, it has been reported that radiation-related complications and hemorrhage may frequently occur after radiosurgery [3].

It is important to note that complete resection of cavernomas located in the brainstem is crucial, as incomplete resection may increase the risk of rebleeding by up to 43% [4]. In the follow-up of the 22 surgically treated cases, 20 achieved cure (with no residual lesion on MRI), one died [12], and one had a small residual lesion during follow-up [23].

All cases showed improvement in pain; however, most patients continued to have some degree of hypoesthesia during follow-up. Therefore, surgery is an excellent therapeutic option, yielding favorable outcomes.

Conclusions

Trigeminal CMs are exceedingly rare lesions. We present the 28th reported case of a trigeminal nerve cavernoma, with good postoperative evolution. Although facial pain is the most common presenting symptom reported in the literature, our patient presented primarily with facial hypoesthesia, highlighting the variability in the clinical manifestations of this condition. Given its rarity, this case may assist neurosurgeons in the diagnosis and management of similar lesions. Furthermore, it demonstrates that, despite the challenging anatomical location and technical complexity of the procedure, microsurgical resection can be a safe and highly effective treatment option. To the best of our knowledge, this is the first recorded surgical video of a trigeminal nerve cavernoma. We demonstrate a step-by-step approach to the surgical procedure.

There are some limitations to this case report and literature review. In addition, there is a small number of cases due to the rarity of this condition. Therefore, it is difficult to determine the ideal therapeutic approach. However, surgical treatment appears to be a safe and effective option and should be considered the first-line therapy whenever feasible.

Additional Information

Author Contributions

All authors have reviewed the final version to be published and agreed to be accountable for all aspects of the work.

Acquisition, analysis, or interpretation of data: Tiago K. K. Braga, Gabriel M. Ribeiro

Drafting of the manuscript: Tiago K. K. Braga, Gabriel M. Ribeiro, Marcelo V. Santos

Critical review of the manuscript for important intellectual content: Tiago K. K. Braga, Gabriel M. Ribeiro, Rodrigo I. Pongeluppi, Matheus F. M. Ballestero, Ricardo Santos de Oliveira

Concept and design: Rodrigo I. Pongeluppi, Marcelo V. Santos, Matheus F. M. Ballestero, Ricardo Santos de Oliveira

Supervision: Ricardo Santos de Oliveira

Disclosures

Human subjects: Informed consent for treatment and open access publication was obtained or waived by all participants in this study. Institutional Review Board of the Clinics Hospital of Ribeirão Preto, Medical School, University of São Paulo issued approval 77856623.4.0000.5440. **Conflicts of interest:** In compliance with the ICMJE uniform disclosure form, all authors declare the following: **Payment/services info:** All authors have declared that no financial support was received from any organization for the submitted work. **Financial relationships:** All authors have declared that they have no financial relationships at present or within the previous three years with any organizations that might have an interest in the submitted work. **Other relationships:** All authors have declared that there are no other relationships or activities that could appear to have influenced the submitted work.

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