

Case Report

# Single-session percutaneous embolization with onyx and coils of sinus pericranii

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## ABSTRACT

**Background:** Sinus pericranii (SP) is a rare vascular malformation consisting of an abnormal communication between the extra- and the intracranial venous system. It occurs due to the adhesion of vessels without a muscular layer or a hemangioma on the outer surface of the skull through diploic vessels, communicating with an intracranial venous sinus.

**Case Description:** A 10-month-old female presented with a pulsatile mass on the posterior parietal region. Investigation with brain vascular examinations showed a venous malformation communicating with the superior sagittal sinus under the scalp, without arterial feeder or nidus. An endovascular embolization with coils and a percutaneous embolization with Onyx ® were performed. The final venography showed complete exclusion of the lesion, which was gradually being absorbed.

**Conclusion:** This is the first case of an SP successfully treated in a single session by embolization with coils and onyx.

**Key Words:** Central nervous system vascular malformations, dural arteriovenous fistulas, endovascular procedures, sinus pericranii

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## INTRODUCTION

Sinus pericranii (SP) was first reported by Pott in 1760 when he discovered a soft tissue injury following a cranial fracture. As early as 1845, Hecker presented the disease with the name “varix spurius circumscriptus venae diploicae frontalis,” which became known as sinus pericranii only in 1850 when Stromeier defined the disease as “a subperiosteal cystic sac filled with blood that communicates with an intracranial sinus.”<sup>[4]</sup>

SP is considered a rare benign venous abnormality, comprising an intradiploic emissary vein derived from

an intracranial sinus,<sup>[7]</sup> causing increased subgaleal drain consisting of a network of thin-walled veins forming varicose veins in the skull outer table.<sup>[6,8,10]</sup>

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Due to the rarity of the disease, its pathogenesis and natural history remain obscure.<sup>[3]</sup> It is believed that the main etiology is congenital, which is justified by anomalous and late venous development during embryogenesis and an incomplete closure of the cranial suture, which is associated with an abundant number of diploic and emissary veins.<sup>[9]</sup> Rare traumatic associations have been reported due to the rupture of emissary veins associated with small cranial fractures or even by direct lacerations of the dural venous sinuses, allowing the development of aberrant communications between the epicranial and dural venous systems.<sup>[8]</sup> However, it is believed that, even in these cases, there is a genetic predisposition. This is corroborated by the frequent association with intracranial developmental venous anomalies.<sup>[4]</sup>

Regarding gender distribution, some authors believe that there is a slight predominance in females,<sup>[3]</sup> but others believe that, if we consider the SPs of traumatic origin, this distribution would be even and may even have a predominance in males.<sup>[9]</sup> The reported age ranges from 0 to 70 years being more common in children and young adults.<sup>[4]</sup>

The diagnosis can be made with Doppler ultrasonography/computed tomography (CT)/magnetic resonance imaging (MRI)/magnetic resonance angiography (MRA)/angiotomography (CTA), and digital subtraction angiography (DSA), with DSA being the gold standard.<sup>[8]</sup>

An SP is considered true when changes in shape, size, or vascularity are observed with maneuvers that cause elevated intracranial pressure (ICP), such as Trendelenburg position, sudden neck flexion, jugular vein compression, Valsalva, cough, and crying.<sup>[3]</sup> As the vessels are strongly adhered to the periosteum and there is a direct connection with the intracranial venous system, changes in ICP cause changes in the size of vessels, especially on the outside.<sup>[1,7]</sup>

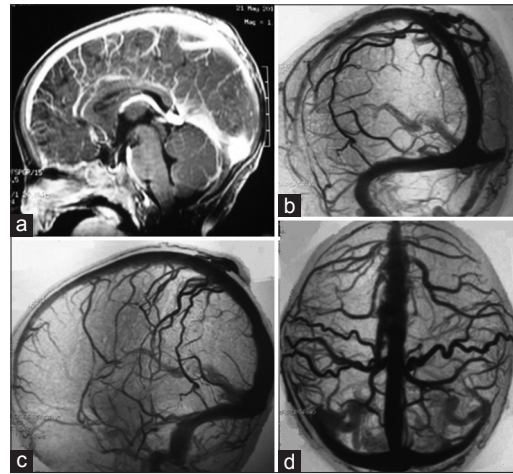
There is no scientific consensus about the choice of the best modality for treatment, which can include from open surgeries, such as the repair of the cranioestenosis and the manual ligation, until endovascular procedures, such as the percutaneous and transvenous embolization, or even percutaneous puncture direct and combined approaches.<sup>[8]</sup>

We report the case of a child with a large SP who was successfully treated only with embolization by percutaneous phlebography.

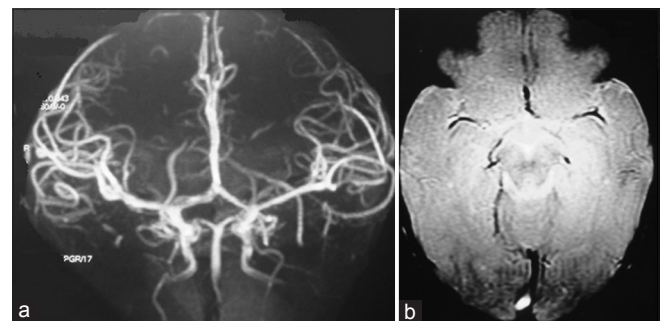
## CASE REPORT

A 10-month-old female child presented with a pulsatile mass on the posterior parietal region with fremitus in the midline. There was no head trauma history and no neurological deficit on physical examination. Brain CT,

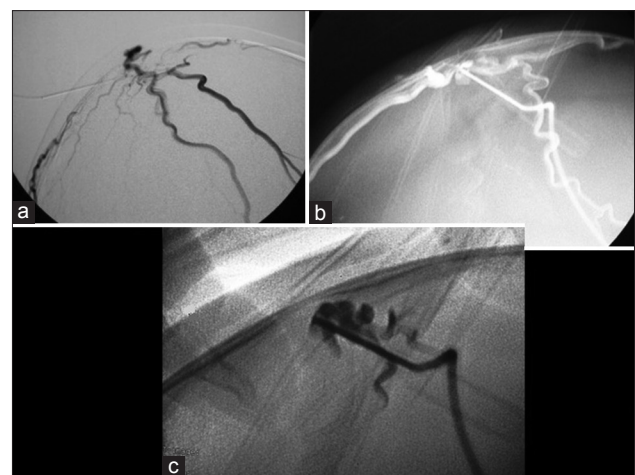
MRI, MRA [Figure 1] and CTA [Figure 2] demonstrated a venous malformation communicating with the superior sagittal sinus under the scalp, without arterial feeder or nidus. We performed an endovascular embolization with coils to decrease the lesion's flow. Following this,



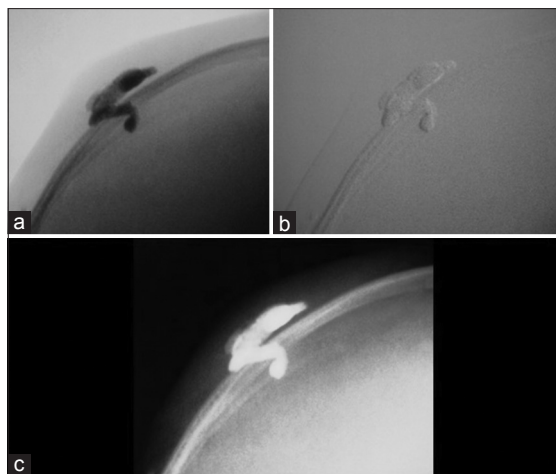
**Figure 1:** 3D reconstruction of brain MRA (venous phase). (a) Perfil view in T2. (b) Left posterior oblique view in T1. (c) Perfil view in T1. (d) Superior view in T1



**Figure 2:** CTA. (a) Right internal carotid artery in anteroposterior view, without arterial malformations. (b) Axial T1 with contrast, with the vascular lesion in posterior interhemispheric fissure



**Figure 3:** Percutaneous embolization with Onyx ®. Needle on the scalp and phlebography. (a) With subtraction. (b) Without subtraction. (c) Making an external compression around the lesion



**Figure 4: Final phlebography with exclusion of the vascular lesion. (a) Final view of the Onyx® without bone subtraction; (b) with subtraction and (c) inversion mode**

an intracranial standard venous phase catheter cerebral DSA was performed, while the lesion was temporarily obliterated by external compression, followed by a percutaneous embolization in one single-session with Onyx ® (ev3, USA) [Figure 3], a nonadhesive liquid embolic agent. The final phlebography [Figure 4] showed complete exclusion of the lesion. The infant achieved good recovery, and a late control DSA confirmed exclusion of the SP. After 6 months, there was no evidence of mass due to absorption. The girl remains asymptomatic and without visible aesthetic defects after 8 years of follow-up.

## DISCUSSION

This is a report of an extremely rare disease. At present, there are just over 100 reported cases of SP in the literature.<sup>[10]</sup>

The lesion in our patient is a typical example of this disease – it was unique, was located in the midline in the parietal region, and measured 3 cm. The main location of the SP is midline or parasagittal (95%), distributed mainly in the frontal (43%) and parietal regions (36%). Other less common locations include occipital (8%), frontoparietal (5%), parietooccipital (3%), occipitotemporal (3%), and temporal (2%). The size of the conglomerate of extracranial vessels may range from 1 to 13 cm, more commonly between 2 and 6 cm. The skull defect varies between 1 and 4 mm, and large defects are rare.<sup>[2]</sup>

Like most cases, our patient was asymptomatic, with only complaints regarding the aesthetic defect. However, there are reports of diverse clinical presentation, from mild symptoms such as local pain, nausea, headache, feeling of pressure, and dizziness to more complex and serious symptoms such as abrupt increase in ICP, bradycardia, ataxia, and even bradycardia.<sup>[9]</sup> It is also

worth mentioning that, although majority of patients have a benign evolution, some have complications such as massive hemorrhage (spontaneous or traumatic), infection, thrombosis, gas embolism, abrupt increase of ICP, and even severe heart failure.<sup>[8]</sup> There are also reports of association of anomalies, such as meningocele, hemangioma, macrocrania, cavernoma, craniosynostosis,<sup>[2]</sup> aneurysm, and arteriovenous malformations. Cavernous hemangiomas, in addition to angiomas, meningoencephaloceles, meningoceles, and cephalohematomas, are also the main diagnostic differences. Symptomatic SP or with a significant cosmetic deformity are strong candidates for surgical treatment.<sup>[9]</sup>

Surgical excision, in a majority of cases, besides endovascular embolization, has positive results (cure, stabilization or emergencies/complications resolution).<sup>[2]</sup>

SPs are classified according to their vascular architecture, which helps in determining treatment approach. When interfering with SP circulation, the circulation of the associated dural venous sinus is also compromised, they are called dominant SP (25% of the cases). However, when the associated sinus vascularization does not change, regardless of the maneuver, SP is said accessory (75%).<sup>[4,7]</sup> In the present case, through DSA and compression maneuvers of the scalp, it was possible to evaluate the repercussion of an intervention in the global venous drainage. The treatment was well indicated because, since it was an accessory SP, there was minimal chance of causing sequelae.

DSA plays a crucial role in the classification of SP and choice of optimal treatment. Although surgery is curative, some studies have reported significant hemorrhage because of dural sinus lacerations.<sup>[9]</sup> The endovascular approach is becoming increasingly relevant and has proven to be safe and effective.<sup>[8]</sup> Especially in the case of a young child, with a higher surgical risk due to blood loss, we opted for percutaneous treatment guided by endovascular view.

Endovascular treatment of SP with N-butyl cyanoacrylate glue is possible, but we opted for the liquid embolic agent Onyx ® (ethylene-vinyl alcohol copolymer). This has been used in the endovascular treatment of intracranial arteriovenous malformations, arteriovenous fistulas, and other venous malformations with excellent results.<sup>[9]</sup> The use of this agent in high-blood flow vessels is reliable and safe. Its use in low-flow vessels, such as veins, can be considered even safer.<sup>[9]</sup>

There is risk to skin ulceration beyond pulmonary or systemic embolization. The latter could be minimized with use of “armored concrete” embolization (use of coils and Onyx).<sup>[5]</sup>

## CONCLUSION

This is the first described case of SP treated in a single session by percutaneous embolization with coils (via endovascular) and Onyx (percutaneous). Although asymptomatic and only with an aesthetic objective, the treatment was well indicated when assessing the repercussion in the global venous drainage through angiography with maneuvers of compression of the scalp.

### Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient has given her consent for her images and other clinical information to be reported in the journal. The patient understands that name and initial will not be published and due efforts will be made to conceal identity, but anonymity cannot be guaranteed.

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Nil.

### Conflicts of interest

There are no conflicts of interest.

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