

Congenital Aneurysm Presenting as Peripheral Facial Paresis

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Key Words

Congenital aneurysm • Facial paresis • Intracerebral hematoma

Abstract

The incidence of intracranial aneurysms in infancy is relatively rare. They are infrequent in children less than 1 year old, and are an exception in the neonatal period. They usually originate in the internal carotid artery bifurcation, posterior circulation and seldom in the territory of the distal middle cerebral artery distribution. Infants often present with nonspecific symptoms: irritability, lethargy, vomiting, seizures and coma. Sometimes, they may present with unexpected clinical symptoms such as peripheral facial palsy. Subarachnoid hemorrhage is the most frequent presentation in this kind of aneurysms. Intracerebral hematoma is unusual. We present the case of a 10-week-old child harboring a middle cerebral artery aneurysm. To our knowledge, this is the first case of an aneurysm presenting as a peripheral facial paresis in a pediatric patient.

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in the territory of the distal middle cerebral artery distribution [1–4]. Infants often present with nonspecific symptoms: irritability, lethargy, vomiting, seizures and coma [5]. Subarachnoid hemorrhage is the most frequent presentation in this type of aneurysms.

We present the case of a 10-week-old child harboring a middle cerebral artery aneurysm. To our knowledge, this is the first case of an aneurysm presenting as a peripheral facial paresis in a pediatric patient.

Case Report

We report the case of a 10-week-old infant who presented to the Emergency Department with vomiting, diarrhea and hypoxia for the last 2 days. Past medical history and family history were unremarkable. Physical examination revealed irritability, drowsiness and seventh nerve peripheral paresis in the left side unnoticed until this moment by his family. He was able to move all extremities equally. Muscular tone and reflexes were all normal.

Computed tomography of the head revealed a left frontal lobe intracerebral hematoma associated with subarachnoid hemorrhage (fig. 1). Subsequent arteriography revealed a cerebral aneurysm arising from the left middle cerebral artery bifurcation (fig. 2).

Elective surgery was chosen and a left frontotemporal craniotomy performed. The hematoma was evacuated and the aneurysm sac was clipped and resected after coagulation and modeling of the neck. Postoperative course was uneventful. At the time of discharge he was asymptomatic. Facial paresis resolved completely within 3 months. No related congenital disorders have been found in the work-up.

Introduction

The incidence of intracranial aneurysms in infancy is relatively rare. They usually originate in the internal carotid artery bifurcation, posterior circulation and seldom

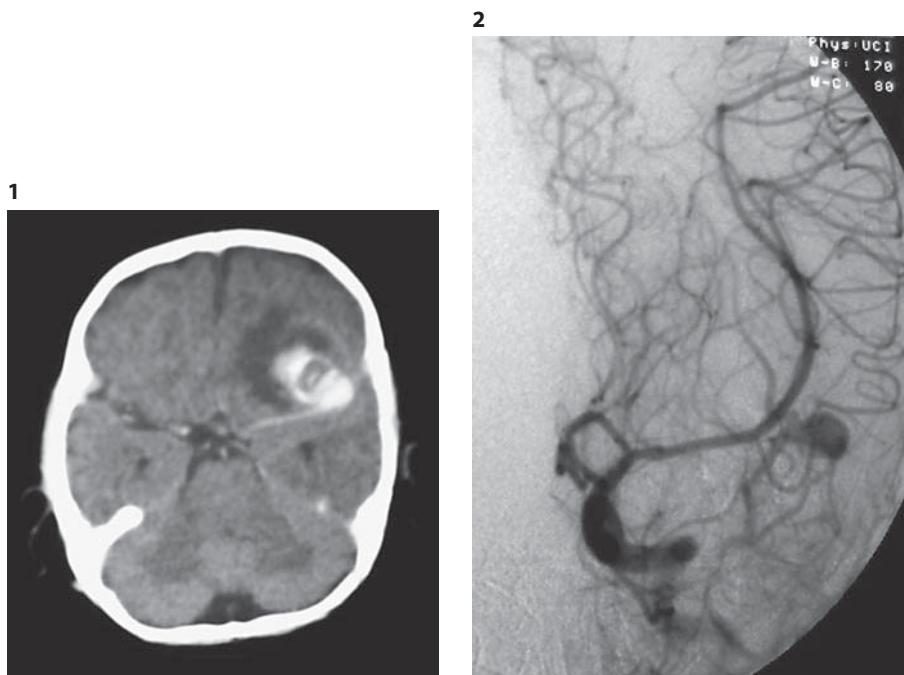


Fig. 1. Axial unenhanced computed tomography scan demonstrating left frontal lobe intracerebral hematoma and associated subarachnoid hemorrhage.

Fig. 2. Left ICA angiogram, lateral view, revealing a cerebral aneurysm arising from the left middle cerebral artery bifurcation.

Discussion

Intracranial aneurysms are relatively infrequent in the pediatric population, with a reported prevalence ranging from 0.5 to 4.6% of total intracranial aneurysms in large series [1–4]. In children less than 1 year old, they are even less frequent, accounting for 18% of childhood aneurysms and approximately 1% of aneurysms at any age [5].

Aneurysms in infants exhibit some well-known special features. Male predominance and predilection for internal carotid artery bifurcation or distal middle cerebral artery location have been reported [1–3, 5]. Posterior circulation aneurysms occur more frequently in children than in adults. Infants also seem to have a higher incidence of large/giant aneurysms and increased risk of aneurysms rupture [2–5]. Etiology is more likely to be traumatic or infectious. The most frequent presentation is subarachnoid hemorrhage. Intracerebral hematoma, as in the present case, is unusual [1, 4, 5].

It is well known that aneurysms in infants are sometimes associated with congenital disorders. Polycystic kidney disease, aortic coarctation, sickle cell anemia, type IV Ehlers-Danlos syndrome, type IV collagenopathy and pseudoxanthoma elasticum are some of them [4–6].

Clinical presentation often consists of irritability, lethargy, vomiting, seizures and coma [5]. To our knowledge, a prior case in which peripheral facial paresis was a clinical feature of aneurysm in a pediatric patient has not been documented.

There are several explanations in the literature regarding pathophysiological aspects of peripheral facial paresis in the context of intracranial diseases.

Peripheral facial palsy could be secondary to direct compression of neural structures. A few reports of adult aneurysms and one report of cerebral venous thrombosis support this hypothesis [6–9].

Although not directly related to the current case, in a second theory, infections and other inflammatory diseases of the central nervous system have been associated with cranial nerve paralysis by a direct injury mechanism.

Yet another theory purports intracranial hypertension as the main factor in peripheral seventh nerve paresis by means of two different mechanisms. The increased intraluminal pressure in the nerve's satellite veins could either cause a reversible compromise in oxygen or glucose consumption or lead to venous blood brain barrier dysfunction with leakage of fluid and ions into the endoneurial space. Both mechanisms could account for neurapraxia. This theory could explain cases related to cerebral venous thrombosis or hydrocephalus reported in the literature [9, 10].

Some authors refer to arterial hypertension as one of the rare causes of peripheral facial paralysis in children, without relation to cerebral vascular diseases [11, 12].

We suggest intracranial hypertension as the main pathophysiological feature in the present case.

The therapeutic options for pediatric aneurysms are the same as those applied in adult patients. These options con-

sist of surgery and interventional neuroradiological techniques [13, 14]. Embolization performed using Guglielmi detachable coils is associated with a lower risk of complications and a lower percentage of complete occlusion mainly in giant aneurysms where recanalization or coil compaction may occur [2, 3]. Surgical clipping with parent artery preservation is an elective option, when is possible [5, 13]. Hunterian ligation of the afferent artery or trapping of the arterial segment harboring the aneurysm may be necessary in some cases with bypass or reconstruction of the artery. It should also be kept in mind that aneurysms in infants can spontaneously thrombose [2, 3, 5, 13, 14].

Conclusion

Congenital aneurysms are rare and sometimes may present with unexpected clinical symptoms. We should be aware of the association of peripheral facial paresis with vascular diseases by different pathophysiological mechanisms because it could prevent delays in the diagnosis. The election of surgical clipping versus interventional endovascular techniques may vary depending on the surgeon's preference and experience, availability of endovascular treatment and characteristics and topography of the aneurysms to treat.

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