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Sinus Pericranii with Dominant Venous Outflow in the Superior Eyelid

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Abstract

Sinus pericranii (SP) located in the superior eyelid is an unusual clinical presentation. Here, we report a case of 72-year-old woman with an unruptured cerebral aneurysm presented with an SP located in the left superior eyelid. The SP was found to have a dominant venous outflow from the bilateral frontal region with an arterialized blood flow pattern on color Doppler ultrasonography (CDUS). During the aneurysmal surgery, intraoperative monitoring of the dominant venous outflow with CDUS was useful for the prevention of venous outflow obstruction. Physicians should carefully consider intracranial vascular anomalies in the differential diagnosis of vascular lesions of the superior eyelid.

Key words: sinus pericranii, superior eyelid, dominant venous outflow, color Doppler ultrasonography

Introduction

Sinus pericranii (SP) is a venous anomaly that represents a transosseous communication between the intracranial and extracranial venous drainage pathways.[1–3] SP is usually asymptomatic and most commonly occurs in the midline communicating with the superior sagittal sinus (SSS).[4] Off-midline locations are less common and have been reported to include the lateral,[5,6] parietal,[1] and occipital regions.[7] SP venous outflow patterns are classified as either dominant or accessory.[8] In some cases of SP with accessory venous outflow, surgical excision or endovascular embolization have been performed for cosmetic reasons, to prevent hemorrhage, or to minimize the risk of air embolism.[3,9,10]

Although rare cases of SP in the superior eyelid have been reported, most of these have had an accessory venous outflow.[11,12] In this report, we present a case of SP with dominant venous outflow located in the superior eyelid. The dominant venous outflow of the SP was monitored with color Doppler ultrasonography (CDUS) during aneurysmal clipping.

Case Report

History and examination

A 72-year-old woman with no previous head trauma or significant medical history was diagnosed with an unruptured left middle cerebral artery aneurysm by magnetic resonance imaging and referred to us for surgical treatment. Contrast-enhanced three-dimensional computed tomography (3DCT) reconstruction imaging was used to confirm the presence of the aneurysm and subsequently revealed unusual vascular structures under the left superior eyelid and in the skin over the left temporal area (Fig. 1A). The patient's left superior eyelid did not exhibit abnormal skin color, erosion, or swelling, but became enlarged when the patient's head was held in a downward position. Ultrasonography revealed compressible and hypoechoic tubular structures under the left superior eyelid. On CDUS, phasic flow under the left superior eyelid was detected communicating with the left temporal subcutaneous vein. Digital subtraction angiography in the venous phase demonstrated an unusual venous drainage route from the bilateral frontal region and occlusion of the anterior third of the superior sagittal sinus (SSS) (Figs. 2A-2C). The venous blood of the SP drained into the left cervical vein (Fig. 2D). Cranial radiography revealed an osseous route in the left frontal bone (Fig. 1B). A three-dimensional digital subtraction angiography surface-rendering image of the left carotid artery demonstrated a saccular aneurysm with wide neck and bleb arising from the bifurcation of the left middle cerebral artery (Fig. 3A). The aneurysm was 6 mm in diameter. Bilateral carotid angiography in the arterial phase did not indicate signs of any other vascular disease, such as ethmoidal or cavernous sinus dural arteriovenous fistula (Figs. 3B and 3C). Based on these findings, we concluded

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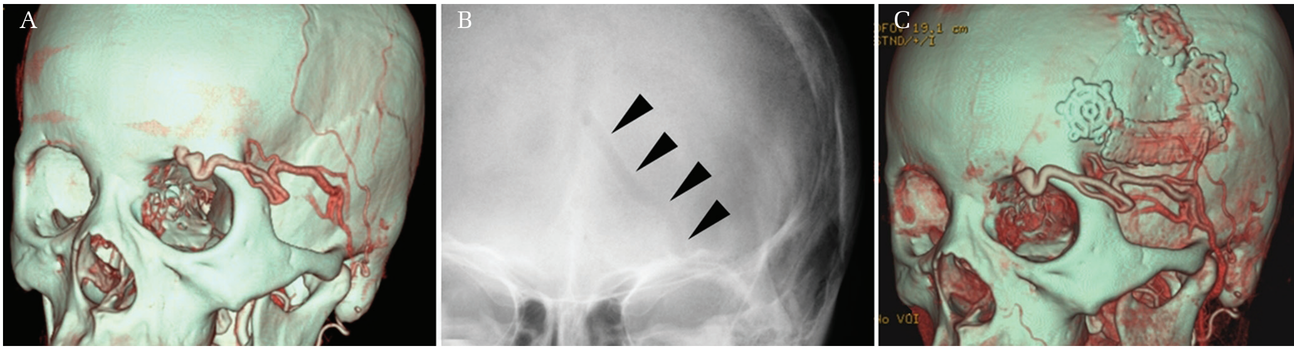

Fig. 1 Preoperative and postoperative imaging of the patient. (A) Preoperative contrast-enhanced three-dimensional computed tomography (3DCT) reconstruction image showing an unusual subcutaneous vein under the left superior eyelid. (B) Cranial radiography, anteroposterior view, showing a straight osseous route in the frontal bone (*arrowhead*). (C) Postoperative contrast-enhanced 3DCT reconstruction image showing preservation of the venous structures of the SP.

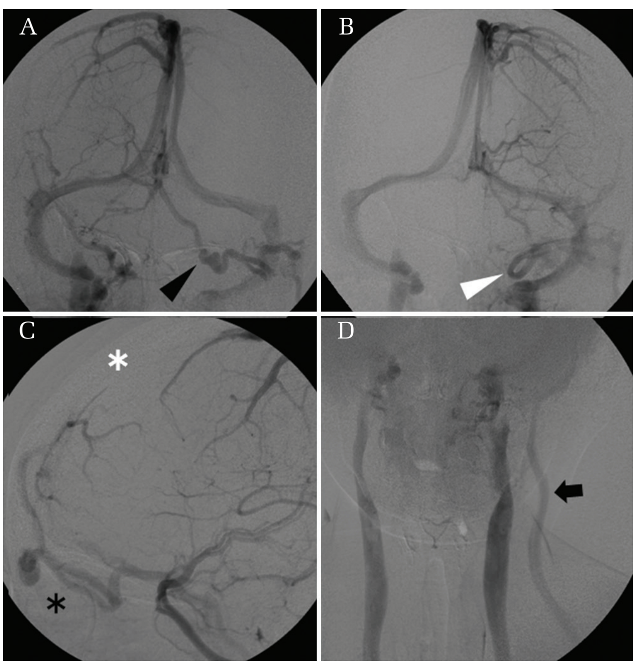

Fig. 2 Digital subtraction angiography in the venous phase. (A) Right carotid angiography, anteroposterior view, showing sinus pericranii (SP) drainage (*black arrowhead*) from the right frontal region. (B) Left carotid angiography, anteroposterior view, showing SP drainage (*white arrowhead*) from the left frontal region. (C) Left carotid angiography, lateral view, showing SP drainage (*black asterisk*) and occlusion of the superior sagittal sinus (*white asterisk*). (D) Cervical angiography, anteroposterior view, showing venous blood from the SP drained into the left cervical vein (*arrow*).

that the observed osseous venous route provided primary drainage from the bilateral frontal region leading to the left temporal subcutaneous vein, and the patient was diagnosed with a dominant SP located in the superior eyelid. For the treatment of the aneurysm, we selected surgical clipping rather than endovascular coiling based on the patient's age and medical history, and the location and configuration of the aneurysm.

Operation and postoperative course

Surgical clipping of the left middle cerebral aneurysm via a left pterional approach was performed to preserve dominant venous outflow from the SP. Under general anesthesia, the patient's head was rotated to the right with the vertex facing downward. A left semicoronal skin incision was made, and the skin flap was reflected anteriorly. A hockey stick CDUS probe (Hitachi, Ltd, Tokyo, Japan) fixed to a Sugita head frame was positioned on the left superior eyelid to monitor venous outflow from the SP (Fig. 4). Upon CDUS examination, both continuous flow and phasic flow were observed in the hypoechoic vascular structures. An arterialized phasic flow pattern was detected in a bone hole of the supraorbital rim with a peak systolic velocity of 34.9 cm/s, an end diastolic velocity of 14.1 cm/s, a time-averaged maximum velocity of 21.5 cm/s, and a pulsatility index of 0.966 (Fig. 5A). A strong reflection of the skin flap compressed the vascular structures and abolished phasic flow (Fig. 5B), whereas gentle reflection of the skin flap during surgery permitted consistent venous outflow. The left middle cerebral aneurysm was successfully clipped and the postoperative course was uneventful. Postoperative 3DCT demonstrated the preservation of the venous structures of the SP (Fig. 1C).

Discussion

The present case had two unusual vascular findings. First, SP in the superior eyelid is rare, as the most frequent site of SP associated with venous outflow from the SSS is the cranial midline. In addition, venous outflow of the SP in the present case provided a dominant venous drainage route from the bilateral frontal regions. Only two previous reports have documented SPs involving the superior eyelid.[11,12] One of these reports described an unusual location of an accessory SP in the frontal region involving the superior eyelid.[11] If dominant venous drainage had been affected by trauma or thrombosis, a life-threatening complication may have occurred, such as venous congestion and/or infarction, brain swelling, or hemorrhage. Several vascular

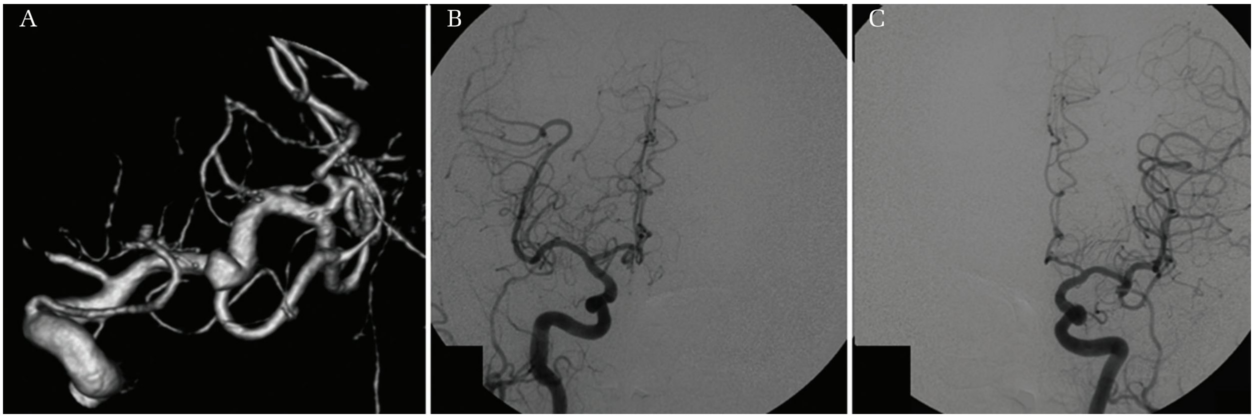

Fig. 3 Digital subtraction angiography in the arterial phase. (A) A three-dimensional digital subtraction angiography surface-rendering image of the left carotid artery, anteroposterior view, showing a saccular aneurysm with wide neck and bleb arising from the bifurcation of the left middle cerebral artery. (B) Right carotid angiography, anteroposterior view, showing no abnormal findings. (C) Left carotid angiography, anteroposterior view, showing a left middle cerebral artery aneurysm.

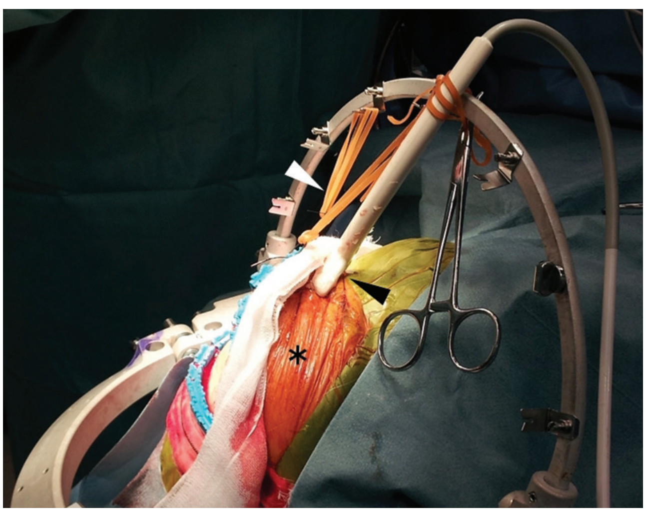

Fig. 4 Setup for monitoring the venous outflow from the sinus pericranii. A hockey stick probe for color Doppler ultrasonography (*black arrowhead*) was fixed to a Sugita head frame and positioned on the left superior eyelid. The skin flap (*asterisk*) was retracted with scalp hooks (*white arrowhead*).

diseases involving lesions of the superior eyelid have been described, including dilated subcutaneous eyelid vessels caused by a carotid-cavernous sinus fistula,[13] cavernous hemangioma associated with Sturge-Weber syndrome,[14] capillary hemangioma (the most common childhood benign periorbital tumor),[15] and Kaposi sarcoma (most often seen in patients with acquired immune deficiency syndrome).[16] Vascular entities, such as SP, should be carefully considered in the differential diagnosis of vascular lesions of the superior eyelid.

Second, an interesting point of focus in the present case was that SP outflow visualized on ultrasonography revealed an arterialized blood flow pattern, while cerebral angiography studies showed venous blood flow back into the SP (Figs. 2A and 2B). Considering that the volume of blood

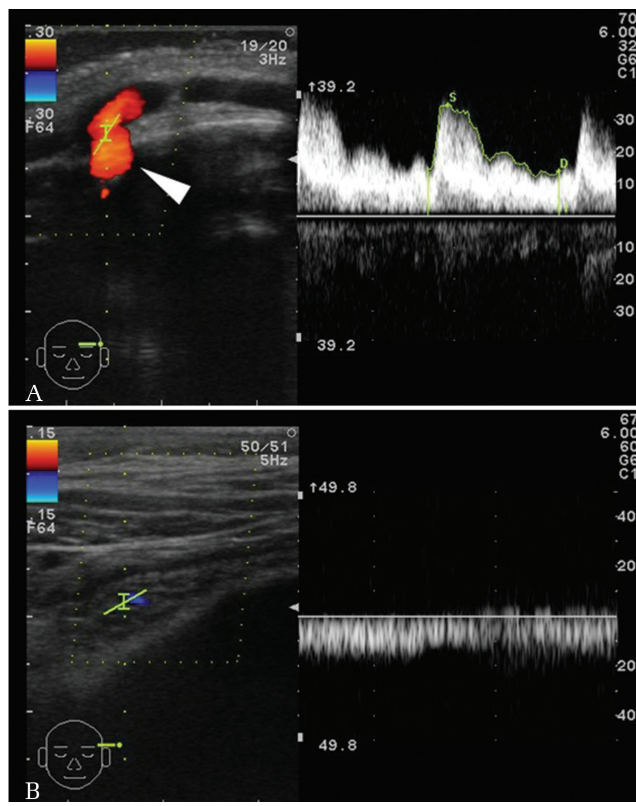

Fig. 5 Venous outflow from the sinus pericranii on color Doppler ultrasonography. Upper panel: An arterialized phasic pattern was detected in the left superior eyelid. The arrowhead indicates a bone hole in the left supraorbital rim. Lower panel: The vascular structures in the left superior eyelid were compressed by strong retraction of the skin flap, leading the temporary disappearance of venous flow and the phasic pattern.

flow from the intracranial region back to the extracranial region was constant in the present case, it can be argued that the flow velocity becomes larger when passing from the SSS with a large diameter through an osseous route with a smaller diameter penetrating the left frontal bone. Moreover, in the presence of cerebral pulsation, an arterial blood flow pattern could reasonably be indicated on CDUS. Generally, evidence of an arterial blood flow pattern on CDUS can indicate a superficial temporal artery aneurysm and/or a soft tissue tumor with an arteriovenous shunt; an SP exhibiting an arterialized blood flow pattern might be overlooked or misdiagnosed in these cases. Thus, if an SP is identified, ultrasonography alone cannot fully inform the extent of venous drainage that is accounted for or influenced by the SP. Accordingly, cerebral angiography is required to determine whether a SP is dominant or accessory.

In the present case, intraoperative monitoring with CDUS was useful for preventing the obstruction of dominant venous outflow from the SP. Awareness of both the patency and direction of flow in a dominant emissary vein is important for preventing life-threatening complications during surgery. Although intraoperative cerebral angiography or indocyanine green video-angiography can be used to evaluate the venous flow, these are invasive methods that only provide intermittent monitoring. Alternatively, ultrasonography is non-invasive and inexpensive relative to other imaging modalities, and can provide continuous evaluation of venous flow during surgery. As in the present case, ultrasonography can also detect extracranial venous tubular structures and visualize the emissary vein. Moreover, CDUS can confirm venous flow in these tubular structures and reveal both the direction and pattern of venous flow in the emissary vein.[17] Recent advances in technology have yielded a variety of ultrasonography probes suitable for diverse operative situations. The technique described herein may be useful for the real-time monitoring of blood flow during surgical as well as interventional endovascular procedures.

Three possible causes have been postulated with regard to the pathogenesis of SP: congenital, spontaneous, and traumatic.[18] The etiology of the present case may have been congenital given the observed occlusion of the anterior third of the SSS, which possibly led to the development of a transosseous venous route in the left frontal bone during the prenatal period.

Although the frequency of cerebral aneurysm is high in patients with connective tissue diseases, such as polycystic kidney disease,[19] Ehlers-Danlos type IV,[20] and neurofibromatosis type I,[21] the relationship between cerebral aneurysm and SP remains unknown. Coexisting diseases associated with SP have been reported, such as cerebellar venous angioma,[22,23] blue rubber bleb nevus syndrome,[24] arteriovenous malformation,[8] dural sinus malformation,[8] vein of Galen aneurysmal malformation,[8] cavernous hemangioma,[25] aneurysmal malformation of the internal cerebral vein,[26] vein of Galen aplasia,[8] PHACE syndrome,[27] craniosynostosis,[28] and subcutaneous venous cavernoma.[1] To this end, physicians should be vigilant for possible congenital vascular anomalies in patients with cerebrovascular disease.

Conflicts of Interest Disclosure

The authors report no conflicts of interest concerning the materials or methods used in this study or the findings specified in this paper.

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