

Sinus Pericranii Associated With Epilepsy

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Abstract: Sinus pericranii is a rare vascular lesion of the cranial vault. Abnormal diploic veins provide communication between the lesion and the intracranial venous sinus. Although sinus pericranii is well documented in the literature, we report a patient with sinus pericranii and epileptic seizures. To our knowledge, a

case of sinus pericranii associated with epilepsy has not been previously reported. The patient was seizure free after the removal of the lesion. The relationship between sinus pericranii and epilepsy is also briefly discussed.

Key words: Epilepsy, Sinus pericranii.

INTRODUCTION

Sinus pericranii is a subperiosteal vascular anomaly which is connected with the intracranial venous sinus through abnormal diploic or emissary veins. It was first described by Stromayer in 1850, as "a cyst containing circulating blood" (13). The lesion is usually located midline lying along the superior sagittal sinus. Patients are usually asymptomatic. To our knowledge, this is the first case of sinus pericranii associated with epilepsy.

CASE REPORT

First Admission (March 1981): A 34 year-old man was admitted to the Neurosurgery Service of Hacettepe University Hospital for evaluation of the aetiology of epilepsy. He had had generalized tonic clonic seizures following nausea, blurred vision and dizziness, since 1978. There was no remarkable history for epilepsy. On admission, physical examination revealed only a subcutaneous mass 1cm in diameter in the right frontal area which was considered to be a lipoma or sebaceous cyst. Neurological examination was entirely normal. Repeated electroencephalograms demonstrated nonspecific slow wave activity on the vertex and fronto-temporal areas. Percutaneous carotid

angiography and cranial computed tomography were performed. No pathology was found in these studies. The patient was discharged with diphenylhydantoin 300 mg per day. At follow-up, rare focal motor seizures either on the right or the left side of the body had continued with or without secondary generalization.

Second Admission (November 1990) - The patient was readmitted for a progressive swelling on his forehead. At physical examination; a painless, soft 3x3cm diameter tumour covered with normal skin was seen in the right frontal area. The mass disappeared with compression or in the sitting position. Other physical and neurological findings were normal. Plain skull x-rays showed a small bone defect in the right frontal area. Soft tissue density CT (Window width: 75) was almost normal (Fig.1) but in the bone window CT (Window width: 800), the subperiosteal mass and bone defect were clearly seen (Fig.2). Selective carotid angiography (with digital subtraction) was performed and revealed opacification of the mass and drainage to the superior sagittal sinus in the late venous phase (Fig.3). This lesion was diagnosed as a sinus pericranii in the preoperative period. At operation, a semicircular scalp incision was made around the tumour. It was seen beneath the periosteum. The lesion was like a varix and was filled with venous blood. Craniectomy

was performed and the tumour was totally extirpated. The few vessel communications between the tumour and the superior sagittal sinus were coagulated and cut. At the same operation, cranioplasty was performed with an autogen bone graft. The postoperative period was uneventful. Histopathological studies of the lesion showed irregular sinuses lined with a layer of endothelium in fibrotic tissue (Fig. 4). At five-year follow up, the patient was seizure free without antiepileptic medication.

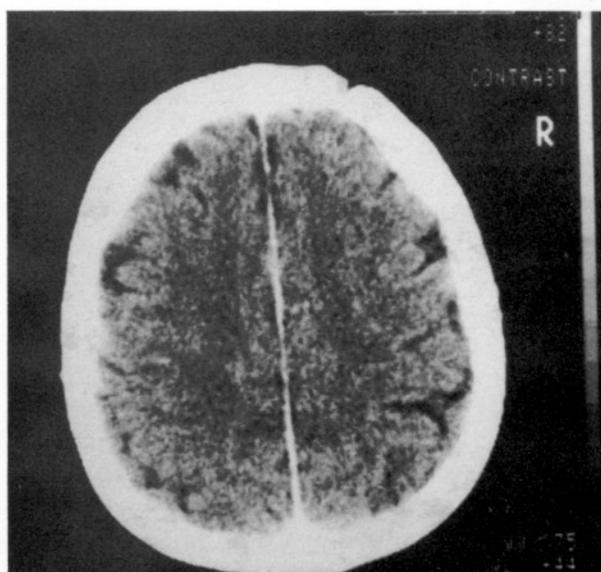


Fig. 1: Soft tissue density CT (Window width: 75) was almost normal.

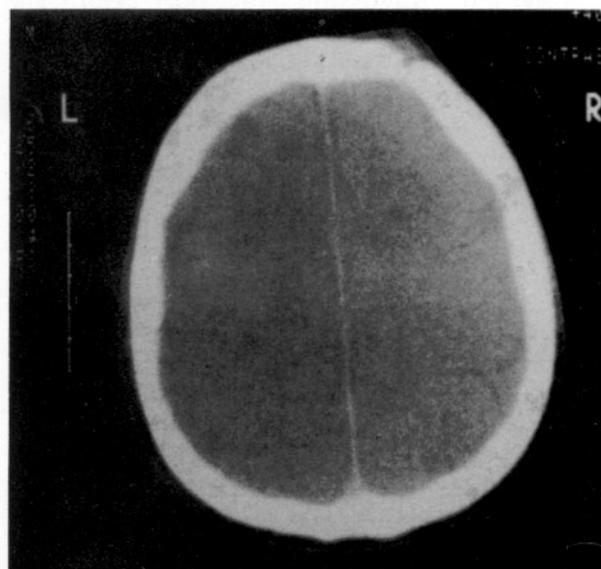


Fig. 2: "Bone window" CT (Window width: 800) clearly showed the subperiosteal mass and bone defect in the right frontal area (same slice as Fig.1).

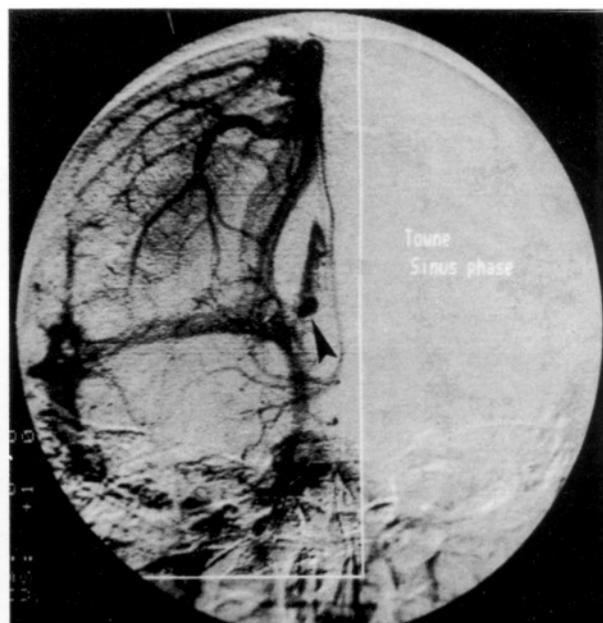


Fig. 3 : IV DSA Appearance: Vascular lesion (arrowhead) and drainage veins to superior sagittal sinus seen in the late venous phase.



Fig. 4 : Histopathological study showed irregular sinuses lined by a layer of endothelium in fibrotic tissue (H&Ex130).

DISCUSSION

Sinus pericranii, first described by Stromeyer in 1850 (13), is a rare vascular tumor that is caused by abnormal communications between intracranial and extracranial venous system. It is found beneath or in the periosteum of the cranial vault. Abnormal diploic veins provide communication between the lesion and intracranial venous sinus (5, 6, 10, 11). Formerly, this lesion was called "Fistula osteovasculare" or "Varix sprius circumscriptus venae diploicae frontalis" in the literature (7). According to Ohta, sinus pericranii is a collection of

non-muscular venous blood vessels or a venous hemangioma (11). There is no feeding of the sinus pericranii from the arterial system, therefore, it always contains venous blood and is not pulsatile (6, 10).

Etiologically, three types of the sinus pericranii was described (1, 10, 11, 12). One is head trauma. Fractures of the external table, tear of an emissary vein or venous extradural haematoma due to sinus tearing have been proposed as the pathogenesis of a traumatic origin. This traumatic haemorrhage extends below the periosteum and is then covered with the epithelium. Stromeyer stated that, subgaleal haematoma arose first from head trauma then communicated with the intracranial venous sinus via abnormal diploic veins (13). According to Nimura et.al, venous sinuses or feeding vessels are injured by linear fracture of the skull and then a vascular mass develops by bleeding (9).

Congenital origin of sinus pericranii has also been proposed. In addition sinus pericranii, coexisting with congenital anomalies support the congenital theory (2, 15). However head trauma in the course of labour may be responsible for the production of sinus pericranii in some so-called congenital cases (11). Histologically, Cohn (3) believed that the walls of the traumatic type had a connective tissue lining and congenital tumours have an endothelial lining.

Finally, spontaneous origin has been proposed. According to Mastin; osteitis and venous diseases are causative factors (7). In contrast, Arrues et al, stated that , spontaneous sinus pericranii is probably a variety of the congenital type which is revealed belatedly during the patient's lifetime (1).

Sinus pericranii is generally in the frontal region mostly in the midline. Lateral situated cases are rare (6, 10). The lesion is made more pronounced by the Valsalva manoeuvre or placing the head in a dependent position; it disappears with compression (6, 10, 11, 12). These clinical features are important in the diagnosis of the sinus pericranii.

CT is helpful for the diagnosis. According to Sadler et al., on noncontrast CT sinus pericranii is slightly hyperdense compared to the brain (12). In our case, the lesion was isodense and the mass was not clearly enhanced following intravenous contrast medium administration. This may be due a narrowing or thrombosis of some abnormal diploic veins. According to our experience, density of the CT is

more important than contrast medium administration. Especially "Bone window" CT (Window width: 700-900) clearly shows the cranial defect and the subperiosteal tumour.

Cerebral angiography demonstrates abnormal communicating veins and the accumulation of the contrast medium within the lesion. Multiangle views may be necessary to demonstrate the mass, especially the late venous phase as in our case. Some authors contend that direct injection into the lesion is necessary when there is non-visualisation by cerebral angiography (1, 6, 11, 14). We found that, "bone window" CT and cerebral angiography with digital subtraction gave enough information for a true diagnosis.

Meningocele, encephalocele, sebaceous cyst, eosinophilic granuloma and especially venous cavernoma of the scalp should be differentiated from sinus pericranii. Cavernoma is located in the subcutaneous space, and does not communicate directly with the intracranial venous sinuses.

Although a few spontaneous occlusions have been reported (4) , sinus pericranii must be treated surgically because, there is a danger of severe bleeding and air emboli due to trauma . After extirpation of the tumour, cranioplasty should be performed with bone graft or acrylic, if necessary.

Patients are usually asymptomatic but may present with headache (6, 11, 14). Vague symptoms of nausea, vomiting and vertigo have also been reported (11). We were unable to find a case of sinus pericranii with epileptic seizures in the literature. In 1975, Nakayama and Matsukado (8) reported a patient with predominant theta-ranged irregular pattern in electroencephalogram, but there was no epileptic discharge or clinical seizure. In our case, we can not explain the relationship between sinus pericranii and epilepsy. We postulate that both frontal lobes may be affected by transient impairment of blood flow in the superior sagittal sinus and changes of metabolism due to the indirect effect of sinus pericranii. In our patient, stopping the seizures with removal of the tumour is good evidence for the causative relationship between sinus pericranii and epileptic seizures.

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