Combined sagittal and lateral sinus dural fistulae occlusion

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The clinical course of three patients who had combined sagittal and lateral sinus dural fistulae is described. One patient, with impending blindness and a short life span, underwent right lateral sinus thrombosis in one stage. The second patient underwent sagittal and right lateral sinus thrombosis in four stages, and her remaining lateral sinus spontaneously occluded. A small untreated fistula persists in the right lower sigmoid. The third patient underwent sagittal sinus, left lateral sinus, and left parietal sinus thrombosis in six stages. A small untreated far-anterior fistula persists. All “satellite” fistulae have spontaneously disappeared. The small asymptomatic untreated residual fistulae have no demonstrable retrograde venous drainage and, therefore, require no treatment at this time.

Key Words • dural sinus fistula • sagittal • lateral • occlusion in situ

MULTIPLE locations of spontaneous dural sinus occlusion in association with a single site of dural sinus fistula are well-recognized. Contiguous dural fistulae, as in the superior petrosal and in the adjacent lateral sinus, are not infrequent; however, multiple isolated sites of dural fistulae are less common. We describe our experience with three patients who had both sagittal and lateral sinus fistulae. If it is assumed that fistulae in the sinus are preceded by a spontaneous and tolerated sinus occlusion, then it may be presumed that an induced permanent occlusion, as a therapeutic measure, could be tolerated without adverse effect on cortical or ophthalmic venous drainage. This line of reasoning would apply to combined sagittal and lateral sinus fistulae as well as to a more limited fistula. If, on the other hand, this assumption were erroneous, then induced therapeutic thrombosis of both the sagittal and lateral sinuses could be dangerous. An experience with these three cases of combined sagittal and lateral dural sinus fistulae recognized the hazard of the untreated state and explored the safety of the proposed therapy.

There was no initial overall plan to thrombose both the sagittal and lateral sinuses simply because they were there. The plan arose as a step-by-step response to the clinical progression of the disease, the symptoms and hazards that were generated, our previous experience with dural venous fistula occlusions, and the sequential response shown by each of these patients following each staged occlusion. No sinus was thrombosed until symptoms or threat to function demanded it. For example, in the first patient, the lateral sinus was occluded, but the sagittal sinus was left untreated because the symptoms within the expected life span were controlled. In the second patient, intervention was delayed for 4 years until the disease progressed to a stage that presented a serious threat to vision and to arm function. In the second and third patients, a trace fistula persists because at the moment there is no retrograde intracranial or intraorbital back pressure giving rise to symptoms or to a threat of loss of function. The first case is presented because it demonstrates the serious loss of function that may ensue if this condition is left untreated and alerted us to prepare for intervention in the two subsequent cases before such loss occurred. The final outcomes indicate that, in selected cases of extensive sagittal and lateral sinus fistulization, the disease can be safely controlled and the symptoms eliminated by staged deliberate occlusion of the sinus system.

Illustrative Cases

Case 1

This 53-year-old man, in the terminal stages of amyotrophic lateral sclerosis with brainstem involvement, assumed that his progressive blindness was part of his disease. He had difficulty identifying faces and complained of a distressing noise in his head, which led to the discovery of severe papilledema and to the additional and separate diagnosis of combined right lateral sinus and sagittal sinus fistulae. Angiography obtained in this patient, illustrated but not described, has been presented in previous publications. A standard obliteration of the very high flow right lateral sinus fistula without treatment of his two
smaller sagittal sinus fistulae stabilized his vision for the remaining year of his life.

Case 2

This somewhat overweight 48-year-old woman presented with headache and papilledema in 1981, 10 years after bilateral mastectomy, with intervening episodes of deep venous thrombosis of her legs.

Examination. Physical examination revealed that vision in the right eye was 20/100 and in the left 20/40; the fields of vision were intact. Radiological examination showed slightly enlarged scalp and meningeal vessels in relation to the posterior sagittal sinus and to the right lateral sinus. There appeared to be some right internal carotid artery (ICA) input and a small vertebral artery contribution. It was believed that dural fistulae existed, but they were too small to account for the papilledema or visual loss. There was a more clearly defined vascular malformation posterolateral to the right side of the spinal cord at C-2. A prominent mastoid emissary vein was seen on either side. The sagittal sinus was patent. The left lateral sinus was obstructed from the midline up to but not including the vein of Labbé. Pseudotumor cerebri was diagnosed and treated by lumbo-peritoneal shunt, and symptoms and papilledema receded over a period of 4 months. A review of the radiological evidence found that there was slight lumen irregularity in the posterior sagittal sinus. The mastoid veins probably indicated bilateral jugular bulb insufficiency, although these were not adequately visualized. An anterior bilateral anastomosis between the parasagittal and the middle cerebral veins suggested that there may have been some posterior venous impairment. At this stage, the parasagittal cortical veins had full access to the sinus. The reconstructed diagnosis was intracranial venous hypertension (Fig. 1 left, center, and right).

Reexamination. Four years later, in 1985, her headaches returned. Then in 1986, she experienced severe dizziness and her left arm became weak. The occlusion of the left lateral sinus between the torcular vein and the vein of Labbé persisted. There was extensive fistulous formation in the walls of the sagittal sinus, especially the right wall, behind the bregma and in the right lateral sinus. The left external carotid arterial input to the fistulous sagittal sinus wall did not empty into the sinus lumen, but rather into a vastly expanded network of right lateral occipital cortical veins (Fig. 2 upper), and from these it drained into the right cavernous sinus. The right external carotid fistulous flow to the sagittal wall followed the same pathway into the right occipital veins (Fig. 2 lower). The left ICA did not supply the fistula. Its venous return was into the left cavernous sinus anteriorly and posteriorly through the vein of Labbé into the left lateral sinus anterior to its posterior obstructed segment (Fig. 3 left). It was excluded from the sagittal sinus. The ICA on the right contributed to both the posterior sagittal and to the right lateral sigmoid sinus fistulae. Its fistulous return was into the right occipital veins and into the right lateral sinus (Fig. 3 right). Its nonfistulous return was directed into the cavernous sinus anteriorly in much the same manner as on the left and into the occipital network of veins posteriorly where it was obscured by the rapid fistulous washout. Although there was no longitudinal flow into the sagittal sinus from front to back, there were multiple pockets of patency. The straight sinus, which had been visible in 1981, was no longer seen. The malformation seen in relation to the high cervical cord in 1981 was no longer visible. The previously identified enlarged mastoid emissary veins were again prominent.

First Operation. In the latter part of 1986 the patient underwent initial surgery which effected obliteration of the sagittal sinus fistula from the bregma to a point two-
thirds of the way back to the lambda. This segment did not drain any cortical blood. Postoperatively the patient was neurologically well, but had an unexplained weakness of respiratory drive, which required reintubation and maintenance of respiratory support for 24 hours. Her headaches continued intermittently. There was no visual change. The arm weakness disappeared.

Second Operation. In March, 1987, the patient underwent a craniotomy, and the scalp arteries were divided through a small flap. The middle meningeal arterial branches were identified and embolized with glue. Postoperatively there was no change in symptomatology.

Third Operation. In April, 1987, the patient again underwent surgery, and the right lateral sinus was occluded. In view of the high fistulous flow and high lumen pressure, it seemed unlikely that this segment drained any cortical or cerebellar blood. Certainly the greatly distended cortical veins drained into the cavernous system, but because there was longitudinal flow (unlike the sagittal flow), it was not possible to exclude some minimal drainage that could be obscured by the high fistulous flow. Because of the previous experience, the patient remained intubated for 1 day. Postoperatively, there was a temporary left homonymous hemianopsia.

Postoperative Interval. During early 1988, there were several 24-hour admissions to a neighboring hospital for episodes of unconsciousness in which hypnea or apnea were major components. These episodes lasted less than 24 hours. The problem was considered to be of an epileptic nature although no obvious epileptic convulsions were recorded. On April 20, 1988, she experienced one of these episodes from which she did not awaken. She was in light coma, moving all limbs easily to painful stimulation; her eyes were closed, and there was no element of “vigil” coma. Multiple electroencephalographic recordings indicated a very disturbed cerebral rhythm, with frequent evidence of epileptic activity. She did not have a continuous
status epilepticus. The tentative diagnosis was that of increased venous pressure due to further spontaneous deep cerebral venous occlusion, especially around the brain stem; however, there was no new arteriogram to substantiate this. Poor respiratory drive remained a consistent feature, but within 2 weeks, there was some eye opening without person or object contact.

Fourth Operation. In May, 1988, the patient underwent surgery to occlude the remaining posterior sagittal sinus down to the torcular herophili because the fistulous malformation of the posterior sagittal sinus continued to drain into the right cortical veins, and thereby increase the intracranial venous pressure. It was hoped this would lessen the venous pressure on the brain stem. It had been known since 1986 that this sagittal segment did not drain any cortical blood.

Postoperative Course. There was no immediate postoperative change. The patient remained in an obtunded state with poor respiratory drive, although her conscious level first improved. She became alert within 2 months. Six months elapsed before she regained adequate respiratory control. Her renewed homonymous hemianopsia, detected upon awakening, receded to the upper quadrant.

She has returned to full activity, participating in running a small business despite some difficulties with memory. A small fistulous segment persists in the lower sigmoid, which was not occluded. There are no retrograde filling veins. It is asymptomatic but there is a detectable auditory bruit.

Case 3

Examination. This 40-year-old obese but active woman presented with fairly rapidly progressive numbness and weakness of her right extremities over a period of 1 month. She had had previous difficulty with the veins of her legs for which she had varicose vein ligations. Angiography was obtained and it revealed that she had extensive fistulization of her sagittal sinus from a point anterior to the hairline to one close to the lambda. This fistulous formation was supplied mainly by the bilateral external carotid vessels, but also from the right ICA through numerous “satellite” fistulae in the right cortex (Fig. 4A to D). Most of the external carotid supply came directly to the sinus, but some came through a few “satellite” sites within the bone. In all, 11 secondary “satellite” sites were counted (mostly internal carotid sites). In addition, she had a large fistula on the left in relation to bone and dura, between the parietal eminence and the midline. This was supplied mainly by the left middle cerebral artery with some assistance from the anterior cerebral artery (Fig. 5 upper). Lastly, there was a fistula in the left lateral sinus, supplied by left external carotid vessels and by contributions from the vertebral and from the tentorial branches of the left ICA (Fig. 5 lower). The fistulous drainage (unlike in Case 2) was into the sinus lumen at all sites and only partly retrograde into the cortical veins. The hemispheric venous return was largely directed into the middle cerebral veins and hence to the cavernous sinuses. Further drainage, especially on the left, was directed toward the superior ophthalmic vein, which could be palpated as a hard pulsating mass above and medial to the globe. There was some proptosis.

First Operation. In October, 1990, the patient underwent an initial operation in which the left parietal fistula was successfully occluded (Figs. 5 upper and 4A).

Second Operation. In November, 1990, the patient again underwent surgery and her left lateral sinus was occluded (Fig. 5 lower and 4A).

Postoperative Interval. The signs of weakness and loss of skill in the right arm and leg ceased to progress and gradually improved. The proprioceptive loss disappeared. Although visual loss was not an initial complaint, it became a problem, but was not severe. Simultaneously a mild papilledema increased, and headaches became a problem. The mid and posterior fistulous areas of the sagittal sinus acquired an added blood supply from the
ICA. Existing external carotid vessels increased in size and new branches became apparent especially posteriorly, as though the fistulization was progressive posteriorly.

Third Operation. In February, 1991, the patient underwent surgery to occlude the anterior third of the fistulous sagittal sinus. Postoperatively, her headaches continued and subjective visual impairment increased.

Fourth Operation. In a fourth operation, performed in May of 1992, the patient’s midsagittal fistulous sinus was obliterated. Postoperatively, a wound infection responded to treatment; visual symptoms persisted, and headaches improved.

Fifth Operation. In March, 1993, the patient underwent surgery to pack the posterior third of the fistulous sinus; however, postoperative angiography revealed that the obliteration was not complete.

Sixth Operation. In the sixth and last operation, performed in June of 1993, the patient’s posterior fistulous sinus was finally obliterated to a point below the lambda, beyond which no further fistula was evident.

Postoperative Course. In the course of the earlier surgical interventions, there were embolizations on at least three occasions using coils and polyvinyl alcohol. Postoperative angiography confirmed obliteration of the sagittal sinus from the hairline back to the torculus, together with absence of the left lateral sinus. The straight sinus, visible in 1990 was no longer visualized (Fig. 6A and B). A remnant of fistula persisted in the sagittal sinus anterior to the hairline, an area that had not been surgically exposed and in which fistula was not initially recognized (Fig. 6C and D). No retrograde filling cortical veins could be identified (as with the sigmoid remnant in Case 2). Because there were no symptoms, no further action has been taken. All satellite fistulae disappeared, although no effort was made to obliterate them directly (Fig. 6C). The patient’s headaches, visual symptoms, and papilledema have completely disappeared. Her handwriting and other fine motor skills in the right hand have almost, but not quite, returned to normal. Her only limitation is a persistent varicose ulcer of her leg.

Operative Techniques

The basic technique of lateral sinus occlusion has been described elsewhere. Sagittal occlusion is more difficult. The procedures were conducted with the patient in the semisitting position. Air embolism is not a likely problem in a high-pressure sinus, although a jugular tourniquet was in place in case of need. A narrow rectangular flap was drilled out from over the sinus with meticulous bone wax hemostasis. Unlike the lateral sinus, the sagittal sinus contained many pockets and septae making balloon isolation of less value and also requiring packing of multiple channels. The thick fistulous walls required additional cauterization. In Case 2, it was possible to enter the distended cortical veins from within the sinus wall and pack their termination in a retrograde manner. The parietal fistula in Case 2 presented a special problem in the first operation in that it lay mainly within the bone rather than in the dura. A thin, unrecognized inner table persisted and the sac tore when the bone was elevated: visibility

![Fig. 5. Angiograms obtained in Case 3. Upper: Left internal carotid angiogram showing the left parietal fistula fed by the left middle cerebral artery and to a lesser extent by the left anterior cerebral artery. Lower: Left external carotid angiogram showing the left lateral sinus fistula fed by the left external carotid and to a lesser extent by small vertebral and internal left carotid meningeal branches.](image-url)
was obscured and it was not certain that the dura had not been torn. Therefore, pressure could not be applied because it might drive the blood intradurally. When hemostasis was finally achieved (16 U with cell saver), the dura was found to be intact. The patient recovered quite uneventfully and was ambulatory the next day.

**Discussion**

In Case 1, occlusion of the sagittal sinus was not considered because of the patient’s terminal debilitated state (atrophic lateral sclerosis) and because his visual deterioration had held in check. In Cases 2 and 3, it was demonstrated preoperatively that the hemispheric drainage had found alternate pathways to the sagittal sinus. Figure 3 illustrates this in the left hemisphere of the patient in Case 2. It was equally clearly demonstrated in the right hemisphere and in both hemispheres of the patient in Case 3. It was thus calculated that the sinus could be obliterated safely. It was reasoned that the alternate channels which had developed would be able to drain the hemisphere better if they no longer had to carry an additional fistulous load. This proved to be the case. Occlusion was performed in stages, as the disease and symptoms progressed, partly as a cautious approach to the problem, partly to avoid blood transfusion, and partly because occlusion of each small segment was in fact a very major time-consuming surgical enterprise. Blood transfusions were needed only for the first operation in Case 3 when the intrasosseous component of the fistula became torn. This excessive blood loss emphasizes the potential for an enormous and dangerous blood loss if an unexpected complication should arise. For subsequent similar cases, one would advise drilling off the bone over the bulge and were thus assumed to be normal vessels that had been captured by the fistula, rather than (as in a congenital arteriovenous malformation) congenitally abnormal vessels. It was thus believed that they would safely tolerate occlusion using hypotension. There was no major fear that any of the “satellite” malformations would bleed. They represented a dilated vascular bed, which developed secondarily. They were not incorporated during an immature fetal state. Because they were derived from a normal vascular bed, they had drainage through normal venous channels, even when a “dedicated” draining vessel which led into the fistula was occluded. It might be postulated that because they were part of a normal vascular bed, the pressure within them was potentially lower than would occur in a bed which was exclusively fistulous, but this has not been measured.

In Cases 2 and 3, there were problems in the deep leg veins before any cerebral fistula was apparent. In both, thrombophlebitis of the legs during hospitalization required inferior vena cava filter placement. An extensive medical search was made for some underlying hematological or vascular factor that predisposed to widespread thrombosis: none was found but the suspicion remains that one must exist.

The presence of a cortical arterial blood supply to a dural fistula is uncommon. The presence of so many as were present in Case 2 is most unusual. Although the dural sinus receives copious venous blood from the cortical veins, it is generally recognized that a fistulous sinus receives its fistulous blood from the cortical mantle of
meninges, skin, bone, and related muscles and not from the underlying cortical arteries. This is in keeping with its embryological history. The development of the vasculature of the brain coverings, which includes the sinus, proceeds quite independently of the vasculature of the pia arachnoid and is well advanced before the forebrain even begins to expand. The cortical veins, which eventually connect the two, are relatively late in the maturation process. They constitute the only anatomical bridge between the two systems. In keeping with this, the developing fistula reaches out into its embryologically compatible territory to create vascular hypertrophy and new vessel development. It does not reach across the subarachnoid space. Endothelium is a potent source of vasoregulatory activity, and conceivably it is the source of an effective vasodilatory and vasogenic hormone. In a rare instance in which the dura had been surgically breached, cortical “satellite” arteriovenous malformations, together with efferent connections to a dural fistula have been seen on angiography to develop and then to disappear entirely upon surgical occlusion of the fistula. Case 3 in this series is quite exceptional in that multiple cortical arteriovenous malformations and connections were present without any previous disruption of the dura. The larger left parietal “satellite” fistula site supplied by the internal carotid vessels was in the dura and it is possible that the right middle cerebral connections in Case 2 may have been as well. It should be noted that in Case 2, this followed an intervention (Operation 2) whereas, those in Case 3 were spontaneous. One in the right frontal region was explored by opening the dura: it was subarachnoid and there was no dural adhesion.

It must be noted that this experience with multiple sinus occlusion is a very limited one. At each step there was good reason to assume that the next proposed occlusion could be safely tolerated. Each subsequent case will similarly need individual consideration of its specific risks. The experience verifies three assumptions. One is that cure can be obtained without excision; removal is unnecessary. It is easy to understand why excision has in the past carried such a high complication rate in view of the very extensive technical efforts demanded by this relatively simple concept of occlusion in situ. The second assumption also proved correct, which was that the “satellite” malformations would spontaneously disappear once the true fistula site, namely the sinus wall, was obliterated. Third, it added substance to the belief that a dural sinus fistula may be closed down without incurring cerebral or ocular ischemia.

References


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