Cirsoid Aneurysm Treatment by Percutaneous Injection of Sodium Tetradecyl Sulfate

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BACKGROUND
Cirsoid aneurysms are uncommon arteriovenous fistulas of the scalp. Surgery for these lesions can be difficult; transarterial embolization is rarely curative, while embolization of the venous pouch with permanent agents usually necessitates subsequent surgical removal of the embolic material. The ideal embolic agent would be one that is safe and effective, commercially available, and would not require subsequent removal.

METHODS
We treated an arteriovenous fistula of the scalp with direct puncture and injection of sodium tetradecyl sulfate, a commercially available sclerosing agent.

RESULTS
Control angiography immediately following percutaneous injection of sotradecol into the fistula showed decreased flow but not complete closure of the lesion. However, within several days of the embolization, the patient’s scalp pain and mass resolved. Four months after embolization, MRA demonstrated no evidence of residual or recurrent fistula. Color doppler flow imaging demonstrated only slightly decreased vascular resistance in the distal superficial temporal artery, possibly indirect evidence of persistent micro-fistulae. Twenty-three months after the procedure, the patient continued to be asymptomatic and had no palpable lesion.

CONCLUSIONS
Percutaneous injection of sotradecol can be considered as one of the treatment options for arteriovenous fistula of the scalp. Further experience is needed to compare the safety and effectiveness of sotradecol with other agents currently used in the treatment of scalp arteriovenous fistulae. © 1996 by Elsevier Science Inc.

KEY WORDS
Cirsoid aneurysm, scalp arteriovenous malformation, embolization, sodium tetradecyl sulfate.

Cirsoid aneurysm of the scalp can be a difficult lesion to treat surgically [1–8]. Treatment of these lesions by transarterial and/or percutaneous embolization with various agents including PVA particles, coils, balloons, silk, acrylic material, and ethanol has been described [5–8]. Thcc treatment methods have been used alone or in conjunction with surgery with varying degrees of success. In this report, we present a case of cirsoid aneurysm treated by percutaneous injection of a commercially available, painless, and relatively safe agent, sodium tetradecyl sulfate, and discuss the findings immediately after treatment and at 23 months follow-up.

CASE REPORT
This 19-year-old male presented with complaint of intermittent, localized, pulsatile pain centered just above the left ear; he reported that this symptom had begun gradually 12-18 months prior to presentation. There was no history of trauma. Physical examination showed a pulsatile mass approximately 4 cm in diameter located 2 cm above the left ear. An MRI study from another institution showed discrete regions of signal void in that region of the scalp consistent with abnormally prominent, high-flow vasculature (Figure 1A). Subsequent angiography demonstrated a high flow fistula between the left superficial temporal artery and a varix (cirsoid aneurysm) that drained via several scalp veins (Figure 1B). There was no internal carotid supply or intracranial drainage. A Fastracker 18 infusion catheter (Target Therapeutics, Fremont, CA) was advanced into the superficial temporal artery within a few centimeters of the lesion. The artery was then embolized with several milliliters of PVA particles (500–700 μ) suspended in contrast material without any apparent reduction of flow through the fistula.
The left parietal scalp overlying the arteriovenous fistula was then shaved and prepped in sterile fashion. Direct puncture of the varix was performed with a 20 gauge angiocath (Becton Dickinson, Sandy, UT). The rim of a sterile plastic denture cup was utilized as a compression device to prevent arterial inflow and venous outflow during percutaneous injection, and a fistulogram was performed.
Two ml of a solution consisting of 3% Sotradecol (Elkins-Sinn, Inc., Cherry Hill, NJ) combined in a 1:3 ratio with nonionic contrast material (Omnipaque 300, Sanofi/Winthrop Pharmaceuticals, New York, NY) with a small amount of Avitene (MedChem Products, Inc., Woburn, MA) was then directly injected into the scalp fistula while the compression device was applied for 5 minutes; the percutaneous injection was followed by a control angiogram via the superficial temporal artery. This procedure was repeated using 2 ml of a 1:2 and finally 2 ml of a 1:1 Sotradecol contrast solution, pausing approximately 10 minutes between injections. Control angiography showed reduction of the apparent luminal diameter of the draining veins and slowed flow through the fistula, but did not demonstrate immediate complete closure of the lesion. Physical examination the following day revealed the lesion to be indurated but faintly pulsatile.

Three months later the patient was seen in clinic follow-up and reported that beginning several days following the embolization, the local scalp pain resolved and the mass likewise had resolved. He was asymptomatic and examination of the scalp revealed no palpable or visible abnormality. Four months after embolization, a follow-up MRI exam including MRA (Figure 1C,D) obtained with surface-coil technique [9] demonstrated no abnormality. Color doppler flow imaging performed on the same date failed to demonstrate any residual varix or vessel enlargement, but the flow pattern in the distal left superficial temporal artery was consistent with abnormally low vascular resistance for an external carotid artery branch. Twenty-three months after the procedure the patient continued to be asymptomatic and had no palpable lesion.

**Discussion**

Arteriovenous fistulas of the scalp are direct connections between a branch of the external carotid artery in the scalp and a scalp vein. They are uncommon, most often occurring spontaneously or after trauma [1,8]. Though not entirely accurate, the term "cirsoid aneurysm" has been used in the past by numerous authors because this lesion resembles a varix, or kirsos (Greek) [7].

Complete surgical removal of these lesions can be difficult [5-8]. Ligation of feeding arteries has been found to be ineffective; also, it leads to recruitment of additional vascular supply, and it may result in loss of vascular access [1,6,10]. Embolization of feeding arteries decreases flow and has been a useful adjunct to surgery, but is rarely curative [6,7]. Embolization of the venous pouch with acrylics or coils may be effective, but almost always necessitates subsequent surgical removal of the embolic material because of overlying skin necrosis [6-8]. Skin necrosis is also a possible complication after injection of the venous pouch with absolute alcohol [8], and though uncommon, cardiopulmonary collapse during intravascular injection of ethanol for treatment of peripheral arteriovenous malformations has been reported (Abstract, Cardiopulmonary collapse: a sequelae of ethanol embolotherapy, Yakes WFJ, presented at 1993 Annual Meeting Western Angiographic and Interventional Society).

Sotradecol (sodium tetradecyl sulfate) is a commercially available, mild sclerosing agent that has been widely used to obliterate varicose veins of the legs [11]. Intravenous injection causes intimal inflammation and thrombus formation that usually occludes the injected vein, subsequent formation of fibrous tissue results in partial or complete vein obliteration. Tissue necrosis may occur following extravasation, and allergic reactions have been reported (package insert; sodium tetradecyl sulfate injection; Elkins-Sinn, Inc., Cherry Hill, NJ). Unlike ethanol, Sotradecol causes no pain upon injection [12].

For this case, we utilized a ring-like compression device to achieve stasis, so as to maximize concentration of the embolic agent at the fistula site and promote thrombosis. Alternatively, it may be possible to obtain stasis by means of temporary balloon occlusion of the feeding artery.

Regarding the case presented here, after initially failing to completely close the fistula, we had anticipated that additional endovascular or surgical therapy would be required to achieve complete closure. Apparently, the full effect of injecting the drug into the varix was not immediate. This may indicate that failure of immediate closure of a fistula with Sotradecol or other sclerosing agent should not necessarily be construed as a treatment failure.

Other investigators treating scalp arteriovenous fistulas with other agents have used clinical criteria to judge outcome, as long as their patients were asymptomatic [6,8]. Likewise, the outcome in the case presented here was a clinical cure; the patient has remained completely asymptomatic. However, the low resistivity in the distal superficial temporal artery as measured by doppler ultrasound suggests that some shunting via micro-fistulae may have persisted. Apparently, it is possible for an abnormal doppler flow pattern to persist, at least for some period of time, in a patient whose signs and symptoms attributed to the fistula have completely resolved. At present, we do not know if this ultra-
sound finding is of clinical significance (i.e., if it indicates the possibility of eventual recurrence). For this reason, we feel that continued follow-up is advisable. We advocate angiographic follow-up; however, because the patient remained asymptomatic, he declined additional angiography.

Considering the excellent clinical result achieved in this case, and that Sotradecol is commercially available, painless upon injection, and does not require subsequent surgical removal, we feel that its use for the treatment of cirsoid aneurysms deserves consideration among the various agents available to the endovascular therapist. Further experience is needed to compare the safety and effectiveness of Sotradecol with other agents currently used in the treatment of scalp arteriovenous fistulas.

REFERENCES

COMMENTS
This report details the treatment of a single case of a scalp arteriovenous fistula (cirsoid aneurysm) by percutaneous injection of a sclerosing agent, sodium tetradecyl sulfate (Sotradecol). In their first attempt to treat this symptomatic fistula, the authors used transarterial embolization with particles, but almost as expected, the particles went through the fistula and it did not close. They then compressed the draining vein with a cup and injected the varix with a sclerosing agent. They have long-term follow-up of 17 months that demonstrates that the patient is asymptomatic. However, at that time, some of their imaging studies would indicate there may be a small residual fistula.

This case report details many of the important aspects of the treatment of these lesions. Primarily, the authors note that transarterial embolization or arterial ligations may be ineffective. The direct percutaneous puncture they performed is a variation of transvenous embolization, which has the highest likelihood of success in this type of case. This paper is the first report of a percutaneous injection of Sotradecol for this problem, and that is a reasonable use.

It would have been interesting if the authors had included their fistulogram at the time of the embolization. Presumably it would have shown reflux from the varix into the superficial temporal artery through the fistulous connection. It is probably important that the fistula itself be sclerosed rather than just the draining vein, unless all of the draining veins are sclerosed. This technique may not be as applicable for larger, high-flow fistulas, but for this lesion, it was a very appropriate therapy.

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The authors have elegantly treated a cirsoid aneurysm of the scalp with percutaneous injection of sodium tetradecyl sulfate. This is actually the fastest, easiest, and safest way to treat these malformations. Intraarterial embolization is usually difficult when the lesion is far away from the origin of the feeding arteries, which are usually distal branches of the occipital and superficial temporal arteries. When it is decided to treat the lesion with percutaneous injection of a sclerosing agent (pure ethanol or Sotradecol) or acrylic glue, great care should be taken not to inject too much embolic material in the soft tissues adjacent to the malformation because of the risk of skin necrosis (one of the 12 cases that I have treated).

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This paper by Hendrix et al. is an interesting one, because it concerns a rare disease in an uncommon location: an arteriovenous malformation in the scalp area.

Superficial vascular malformations are rare diseases, and they are of various types: arterial, venous, arteriovenous, capillary-venous, lymphatic. Each has its own natural history, complications, and methods of treatment.

Thanks to the great number of patients we follow at the multidisciplinary group for superficial vascular anomalies at the Lariboisière Hospital in Paris, we have identified three different types of angioarchitecture in AV shunts: (1) the arteriovenous type, which consists of a direct large AV shunt with a single lumen (AVF); (2) a type of AV fistula, which has until recently been confused with the third type, in which the draining vein, which is large, receives multiple small arteries which communicate directly with the vein; we call this the arteriolo-venous type (aVF); and (3) a nidus type made of a plexiform structure of small arteries and small veins connected to each other: the arteriolo-venular type (avF). The case presented in this paper is of the second type of AV malformations.

We distinguish between these three types of architecture, not only for descriptive purposes, but because of the different approaches to the lesions, the different methods of treatment, and the varying prognoses.

The three structures listed above have different approaches: (1) the arteriovenous type (AVF) has a single point of shunting that can be reached in three ways: arterial navigation, retrograde venous navigation, or, if accessible, direct puncture; (2) the arteriolo-venous type (AVF) may be reached by retrograde venous navigation, which is easy compared to the multiple arterioles that have to be catheterized, or by direct puncture; and (3) the third group (the so-called "nidus") may only be reached by the arterial approach.

The AV fistula type that is presented in this article can be treated in one session by any permanent agent to occlude the lumen. The occluding agent should be chosen according to the diameter of the lumen and by the rate of flow. To reduce the flow, an annular compression is very useful, and it allows one to use a liquid sclerosing agent if the contact with the endothelial wall is long enough. Ethanol is efficient even in large AVFs, but it is painful and carries potential risks (local or general) if used in large volume. Sodium tetradecyl sulfate can also be used; in our experience it is more efficient in small AV fistulas. If the fistula is larger, or if closure is not achieved after two injections of pure liquid agent under flow control, we prefer to complete the treatment with another agent, such as glue, Ethibloc, or coils.

In the arteriolo-venous type (AVF), the principle of treatment is the same, except that the arterial approach cannot cure the whole lesion; therefore, direct puncture or retrograde venous catheterization is the best approach to achieve a cure.

Arteriolo-venous shunts (AVF) are treated exclusively by the arterial approach, filling the whole nidus with a permanent liquid agent (glue or Ethibloc; pure liquid agents carry more risk of neighboring necrosis). In such a nidus, we must always discuss the possibility of a surgical excision; in that case, preoperative particle embolization can be performed.

In most cases of scalp AVMs, the architecture is one of the first two types of AV fistulas: direct arteriovenous or arteriolo-venous. Such a structure carries a major prognostic factor: if it is such a pure shunt, it does not include the risk of angiogenesis of a true nidus type, which means that embolization can be a curative treatment by itself, and can be performed without reserve.

The nidus type is the opposite—in our experience, it carries a high risk of evolutivity after partial treatment; we don't propose any kind of treatment if the AVM is nonevolutive and asymptomatic. A quiescent AVM may have an area of reddish skin (similar to a port-wine stain); which is the "marker" of the AVM. We must also keep in mind that any trauma or endocrine events are factors of evolutivity found in most cases of evolutive AVMs; this is not the case for the two types of true AVF. Therefore, evolutivity related to one of these factors is the marker of a nidus; a pulsatile mass of dilated vessels that is associated with red discoloration of the skin in the same area is also a marker of a nidus—these must be considered before any therapeutic planning. This type, if evolutive, is a candidate for large surgical excision, prepared by skin expansion and immediate presurgical embolization (respecting the normal arterial feeders), allowing for a safe surgical procedure.

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