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# Intravascular Embolization of a Congenital Subclavian Arteriovenous Fistula

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Congenital subclavian arteriovenous fistula (AVF) is an extremely rare entity, only one case has been documented previously [1]. In general, subclavian AVFs present with a bruit, thrill, or mass lesion [2–4]. However, unusual cases can present with intracranial hypertension or congestive heart failure [1, 5].

The presentation in our patient, with venous hypertension in the head and neck, is extremely unusual and partially due to previous surgical ligation. In this case, intravascular embolization was used to successfully treat this lesion.

## Case Report

A 3-year-old girl presented with a 6-week history of increasing facial and left neck swelling. Her medical history was significant for a history of a congenital left subclavian arteriovenous fistula, which was ligated at 1 month of age because of congestive heart failure. She was otherwise healthy, with no evidence of fatigue, shortness of breath, or cyanosis. Physical examination was remarkable for a head circumference of 53.5 cm ( $>+2$  SD) and a loud bruit over the left chest and supraclavicular region.

A head CT showed a moderately enlarged ventricular system and prominent sulci. Arch aortography demonstrated the previous surgical ligation of the proximal left subclavian artery and brachiocephalic vein (Fig. 1A). Multiple collateral channels (including enlarged intercostal arteries and retrograde flow through the left vertebral artery) supplied a proximal left subclavian artery pouch (Figs. 2B and 2C). Flow through the fistula was cephalad into the internal jugular vein.

During attempted entry into the left internal jugular vein, an inadvertent puncture of an enlarged collateral artery occurred, and retrograde catheterization of the arterial segment of the fistula with 5-French Hanafee tubing was performed (Fig. 1D). Twenty 0.038-inch Gianturco coils (lengths, 3–10 cm; coil diameters, 5–15 mm) were placed into the arterial pouch. Postembolization angiography showed persistent opacification of the fistula, but with decreased flow (Fig. 1E). The procedure was stopped at this point with the hope that progressive thrombosis would occur over the next several days. Two days after the procedure, the facial and neck edema were completely resolved with a marked decrease in the audible subclavian bruit.

Five months after the initial procedure, a follow-up CT scan revealed no change from the preembolization study. An angiogram

disclosed persistent slow flow through the subclavian fistula (Fig. 1F). A retrograde internal jugular vein catheterization was performed with placement of several Gianturco coils (sizes similar to those used in first procedure) into the venous pouch and distal jugular vein. Follow-up arteriograms after this second procedure showed complete occlusion of the fistula (Figs. 1G and 1H). The patient is presently doing well clinically.

## Discussion

Arteriovenous fistulae of the subclavian artery are rare. Etiologies include iatrogenic, posttraumatic, and congenital causes [1–7]. Most reported cases are a complication of venous catheterization of the internal jugular or subclavian vein [2–4, 6]. To our knowledge, only one previous report of a congenital subclavian AVF exists [1].

Surgical treatment of these lesions can be difficult [8]. Although no large surgical series of subclavian AVFs has been reported, a review of direct surgical treatment of similar vertebral AVFs revealed disappointing results [9]. As exemplified in this case, arterial ligation fails to prevent formation of collateral channels. In fact, proximal ligation usually makes subsequent treatment more difficult. This is due to recruitment of multiple arterial collateral vessels shunting blood to the AVF. For many AVFs, endovascular management is either the treatment of choice or should be used in conjunction with surgery [3, 9–11].

Previously, one case of congenital AVF has been reported, a 1-month-old neonate presenting with a congenital subclavian AVF and congestive heart failure [1]. Our case has several unusual features, including face and neck edema that was probably a result of increased venous pressure transmitted cranially because of outflow obstruction at the level of the left brachiocephalic vein. In fact, the patient's enlarged ventricles and large head circumference were possibly a result of long-standing elevated pressure transmitted in a retrograde fashion up the left internal jugular vein. This aspect was similar to the case reported by Lal et al. [5], which documented elevated CSF pressures resulting from a brachial arteriove-

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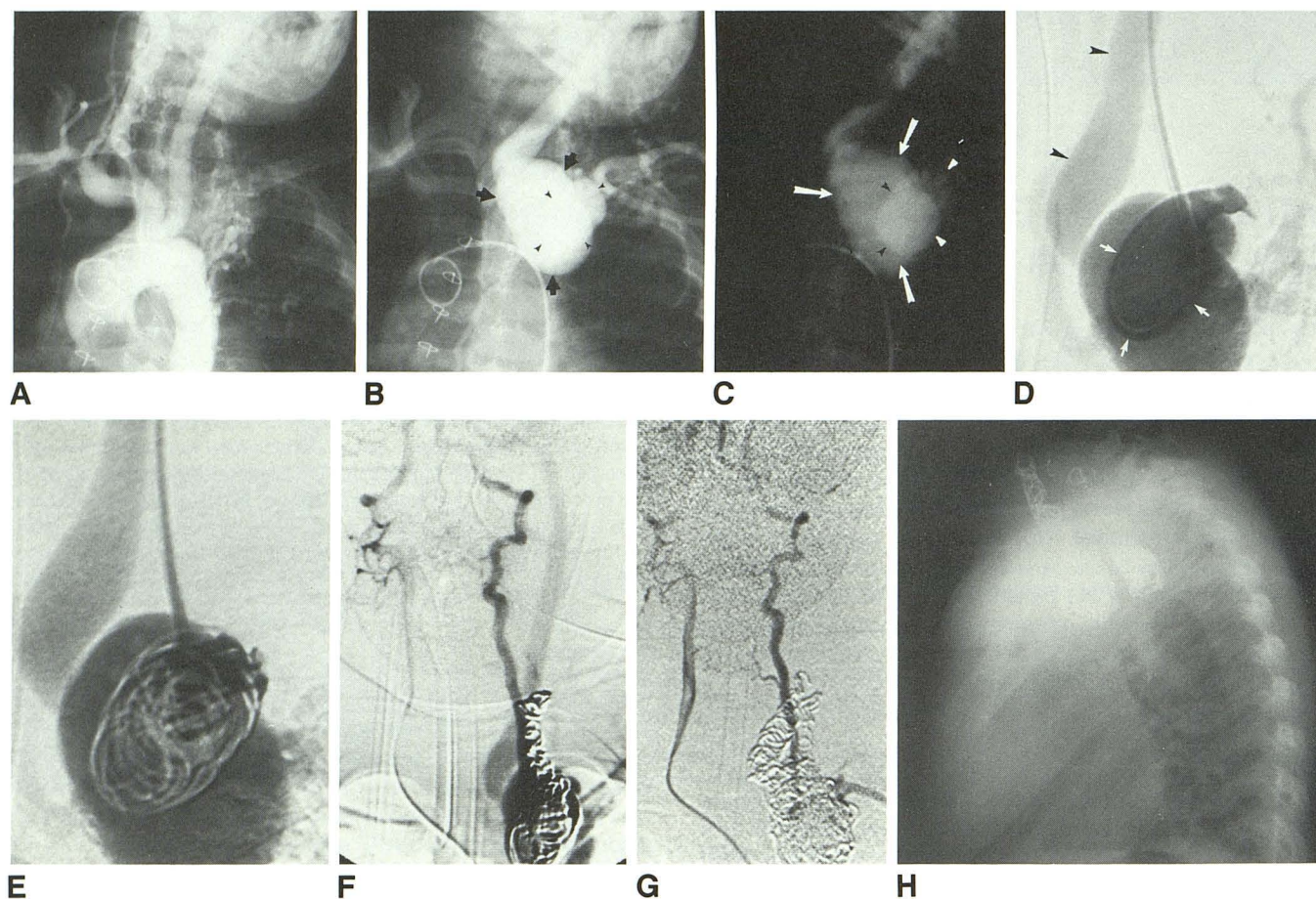


Fig. 1.—A, Arch aortogram, early phase. Prior surgical ligation of proximal left subclavian artery seen with many enlarged collateral vessels supplying arteriovenous fistula.

B and C, Arch aortogram, later phase. Arterial pouches (arrowheads) and venous pouches (arrows) are seen on each side of fistula. In C, image has been darkened to show separate pouches. Venous drainage is cephalad into left internal jugular vein.

D, Embolization, first-stage image. Retrograde collateral artery catheterization shows distal catheter in arterial pouch (white arrows). Early-phase subtraction angiography shows predominate venous drainage into internal jugular vein (arrowheads).

E, Postembolization image of arterial pouch with Gianturco coils. Stasis of contrast material in fistula and slower flow were seen.

F, Right vertebral artery injection. Persistent, but slow flow through the fistula is still seen.

G, Postembolization image of venous pouch with Gianturco coils. No fistula is seen. Retrograde flow through left vertebral artery supplies distal left subclavian artery.

H, Lateral view of the chest reveals placement of coils into anterior (venous) and posterior (arterial) pouches.

nous fistula (for dialysis) with posttraumatic thrombosis of the innominate vein segment. Since the CT findings did not reverse after the first procedure, the atrophic changes were probably a result of chronic venous hypertension that caused a communicating hydrocephalus and/or ischemic changes.

In our case, separate arterial and venous pouches were present on each side of the fistula. In the first stage of embolization, puncture of an enlarged arterial collateral provided access to the arterial pouch. Gianturco coils were used instead of detachable balloons because they could easily fill the pouch with less danger of distal embolization into the left jugular vein and head. Although this first procedure was successful in decreasing flow and improving the clinical symptoms, complete closure of the fistula did not occur.

A second embolization was performed by a technique similar to that of the first procedure. However, the Gianturco

coils were placed into the venous pouch via a retrograde transvenous approach. Complete occlusion occurred after the second embolization.

Subclavian AVFs are unusual and can present with a variety of clinical problems. When the principal draining vein is occluded, rerouting the flow may result in increased venous pressure transmitted to the face, neck, and intracranial compartment. We agree with others [9–11] that transvascular embolization is often the most effective and safe therapy.

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