CASE REPORT

Bilateral and symmetric isolated dorsospinal arteries with prominent anterior radiculomedullary contribution

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SUMMARY

We present the case of a 15-year-old girl with a left T7 spinal epidural arteriovenous fistula (SEAVF), in whom bilateral and symmetric isolated T9 dorsospinal arteries (DAs) were incidentally documented, each providing a significant contribution to the anterior spinal artery. Recognizing the existence of isolated DAs of direct aortic origin is critical, both for the diagnostic evaluation and for the management of spinal vascular diseases. Notably, isolated DAs tend to branch off significant radiculomedullary arteries, which vascularize the normal spinal cord but have also been shown to supply spinal vascular malformations.

BACKGROUND

A unilateral intersegmental trunk is made of two or more consecutive intersegmental arteries (ISAs) that share a single aortic stem. These trunks are either complete—that is, each involved ISA includes a full set of branches—or incomplete, most commonly when one of the ISA lacks its dorsospinal component. In the latter case, the missing dorsospinal artery (DA) generally originates independently from the aorta.¹ It has been suggested that isolated DAs have a tendency to branch off significant radiculomedullary arteries,^{2,3} which may, in addition to supplying the normal spinal cord, participate in the vascularization of spinal vascular malformations.⁴

The present case report offers, to our knowledge, the first observation of multiple DAs in a single patient, and emphasizes the importance of recognizing these anatomical variants, both from a diagnostic and a therapeutic standpoint.

CASE PRESENTATION

Routine pulmonary auscultation revealed an abnormal mid-thoracic bruit in a 15-year-old girl presenting with increasing fatigue and left-sided back pain evolving for several months. Chest CT revealed the presence of abnormal vessels at the T7 vertebral level, suggesting a vascular malformation. The patient underwent spinal digital subtraction angiography (SpDSA), which revealed the presence of a spinal epidural arteriovenous fistula (SEAVF), but failed to document the normal vascularization of the spinal cord. It was suspected that the normal medullary supply arose from the vessel involved by the SEAVF (left T7), and the lesion was deemed untreatable.

The patient's mother consulted our team for a second opinion. Repeat angiography confirmed the

diagnosis of single hole, high flow left T7 SEAVF. The enlarged left T7 ISA showed no evidence of medullary contribution. Incidentally, bilateral isolated DAs with prominent anterior radiculomedullary branches were noted at the T9 level.

The SEAVF was treated endovascularly. A microcatheter was advanced through the arteriovenous connection into the venous pouch of the SEAVF. Detachable microcoils were then deployed within the venous pouch, the fistulous connection, and the distal segment of the feeding artery. The patient was discharged home the following day. She was symptom free at the 3 month follow-up, notably reporting a much improved level of physical activity (eg, climbing a flight of stairs without having to sit down and rest).

INVESTIGATIONS

A contrast enhanced chest CT (figure 1A) revealed a hypertrophied left T7 ISA associated with dilated epidural, azygos, and hemiazygos veins. SpDSA confirmed the presence of a single hole, high flow SEAVF (figure 1B). SpDSA also documented bilateral and symmetric isolated DAs at the T9 level, with prominent anterior radiculomedullary arteries joining over the anterior surface of the spinal cord in a diamond-shaped pattern (figure 1C). The variant was associated with a left T10 unilateral intersegmental trunk branching off the left T10 and T11 ISAs as well as the lateral branch (posterior intercostal artery) of the left T9 ISA (figure 1D). An incomplete right T10 unilateral trunk provided the lateral branch of T9 and the dorsospinal branch of T10 (the lateral branch of T10 branched off the right T11 ISA, not shown here) (figure 1E). The lesion was successfully embolized with detachable microcoils (Micrus, Codman, USA) (figure 1F).

DISCUSSION

Developmental anatomy

The typical anatomy of a thoracic or lumbar ISA includes an aortic stem with three branches: a lateral (or intercostal, subcostal, or lumbar) branch, a medial (or spinal) branch, and a dorsal (or muscular) branch.⁴ As the lateral branch tends to take off first, the ISA generally continues as a short trunk, which takes the name of DA and divides into spinal and dorsal branches. Embryologically, the aortic stem and the medial branch develop first (primary components), while the dorsal and lateral branches appear later (secondary components).⁵ Variations in this branching pattern and the

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Figure 1 A 15-year-old girl with a T7 spinal epidural arteriovenous fistula (SEAVF) and bilateral T9 isolated dorsospinal arteries (DAs). (A) Chest CT, axial view at the T7 level, showing a hypertrophic intersegmental artery (ISA) (large white arrow) with an enlarged radicular branch that travels along the left T7 nerve root towards the epidural plexus (small white arrow), which is also dilated. (B) Digital subtraction angiography (DSA), left T7 injection, posteroanterior view, confirming the existence of a SEAVF supplied by the left T7 ISA (black arrow), with early opacification of markedly enlarged internal (epidural) and external vertebral venous systems. (C) DSA, left T9 injection, posteroanterior view, documenting an isolated left DA (large black arrow) that provides both an anterior (small black arrow) and a posterior (small white arrow) radiculomedullary artery. A right isolated DA (large white arrow), opacified via collaterals, also gives an anterior radiculomedullary artery (white arrowhead) that joins its counterpart over the midline, forming a diamond-shaped pattern. As neither anterior radiculomedullary contributor is clearly dominant, both may be considered as arteries of Adamkiewicz. (D) DSA, left T10 injection, posteroanterior view, showing a T10 unilateral intersegmental trunk (black arrow) branching off the left T10 and T11 ISAs (both with dorsospinal components) as well as the lateral branch (posterior intercostal artery) of the left T9 ISA (without dorsospinal component). The T9, T10, and T11 labels, placed over the corresponding vertebral bodies, show the absence of a dorsospinal component at T9. (E) DSA, right T10 injection, posteroanterior view. The right T10 ISA (large black arrow) gives a lateral branch for the right T9 ISA (small black arrow) and a dorsospinal component for T10 (small white arrow). The T10 lateral component comes from the T11 ISA (not shown). The right T9 isolated DA (large white arrow) is opacified via small anastomotic connections (black arrowhead) back to its aortic origin (white arrowhead). This opacification was considered sufficient for diagnostic purposes, obviating the need to selectively catheterize the right T9 ISA. (F) DSA, left T7 injection, posteroanterior view, showing dense coil packing of the venous pouch, arteriovenous fistula, and distal segment of the feeding ISA. The SEAVF is obliterated, without residual abnormal opacification of the epidural plexus or other early draining vein.

existence of multiple longitudinal paravertebral anastomoses linking adjacent ISAs are at the basis of complex intersegmental anatomical variants, including unilateral intersegmental trunks and isolated DAs.

Angiographic and clinical implications

Chiras and Merland reported the first three angiographic observations of isolated DAs originating from the aorta in 1979.¹ Since then, five more cases have been published,^{2 3 6 7} while three additional observations of a slightly different, more primitive, type were described in 2013.⁴ The majority of reported cases were located at the low thoracic level (T8 to T10), most often at T9. Variants located on the left side have invariably been noted to branch off a major radiculomedullary artery.^{2 3} Importantly, these radiculomedullary branches can constitute the sole supply of a spinal vascular malformation, such as a perimedullary arteriovenous fistula.⁴ The absence of a hemivertebral blush is classically considered as an indirect sign of missing DA. Since a blush is not always observed under normal circumstances, especially in elderly patients, the strongest angiographic sign pointing to a missing DA is the non-identification of the radicular and retrocorporeal arteries constantly provided by its spinal branch. It should also be remembered that the partial thromboembolic occlusion of an ISA or DA might mimic an anatomical variant.

In order to be considered complete, a spinal angiogram must have investigated each and every pair of ISAs and documented the normal (or abnormal) vascularization of the spinal cord. Overlooking an isolated DA may of course lead to a falsely negative study when the variant itself supplies a vascular malformation,⁴ but ignoring the presence of a significant radiculomedullary artery from an isolated DA can also, as in the reported case, interfere with the correct management of a remotely located lesion. It is therefore important for neurointerventionalists to keep in mind the existence of complex anatomical variations that can participate in the normal and abnormal supply to

the spinal cord, notably when SpDSA apparently fails to document expected medullary contributors.

The case presented here is, to our knowledge, the first recorded observation of bilateral isolated DAs, arising at the same level and providing prominent radiculomedullary arteries (anterior on the right, anterior, and posterior on the left).

Learning points

- Complete spinal angiography requires the investigation of all intersegmental arteries (ISAs) to be of diagnostic value.
- An isolated dorsospinal artery (DA) originating from the aorta must be suspected when a vertebral level supplied by a unilateral ISA trunk lacks a hemivertebral blush or when the corresponding radicular and/or retrocorporeal arteries are not identified.
- In its classic form, an isolated DA emerges directly from the aorta and provides the spinal and dorsal components of the ISA. More complex configurations may be encountered due to the intricate anastomotic connections established between adjacent ISAs.
- Neurointerventionalists must have these anatomical variants in mind when performing spinal angiography, as they often participate in the supply of the normal spinal parenchyma and can be involved in various pathological conditions.

Contributors Both authors contributed equally to the writing of this manuscript.

Competing interests None declared.

Patient consent Obtained.

Ethics approval The study was approved by the Johns Hopkins Hospital institutional review board.

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