CASE REPORT

Onyx embolization of anterior condylar confluence dural arteriovenous fistula

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SUMMARY

The anterior condylar confluence (ACC) is a small complex venous structure located medial to the jugular vein and adjacent to the hypoglossal canal. To our knowledge, this is the first report of transvenous Onyx embolization for ACC dural arteriovenous fistula (DAVF). Three patients with ACC DAVF were treated using the Onyx liquid embolic agent with or without detachable coils. Complete angiographic obliteration of the fistulas was achieved in all cases without permanent lower cranial neuropathy. This report suggests that the controlled penetration of Onyx is advantageous in order to obliterate ACC DAVFs with a small amount of embolic material.

BACKGROUND

An anterior condylar confluence (ACC) dural arteriovenous fistula (DAVF) is a rare lesion (rate reported as 3.6% of all DAVFs)¹ but, up to a decade ago, some cases of this disease were reported under other names (eg, DAVF involving the hypoglossal canal, DAVF of the anterior condylar vein) or as the adjacent fistulas (eg, DAVF involving the inferior petrosal sinus, DAVF of the marginal sinus (MS)) due to the complicated venous structure at the craniocervical junction (figure 1).^{2–5} Recently, based on the contributions of high-resolution diagnostic tools and several articles about the detailed anatomy of this location, the terminology has been almost harmonized and successful endovascular treatments based on this accurate anatomical knowledge are being developed. At present, most ACC DAVFs can be successfully treated with transvenous coil embolization.^{1–3 6–11} However, it is not always possible to achieve complete obliteration using only coils in such a restricted small complex compartment without migrating into adjacent vital venous structures including the internal jugular vein, and compressing the hypoglossal nerve.^{1 3 9} Especially in high-flow lesions, the use of liquid embolic material such as Onyx (Covidien/ev3 Neuromuscular, Irvine, California, USA) could be advantageous over coils alone for the treatment of ACC DAVFs. We present three cases of transvenous Onyx embolization of ACC DAVFs and discuss the usefulness and disadvantages of this therapy with a review of the relevant literature.

CASE PRESENTATIONS

Case 1

A 51-year-old woman presented with left-sided pulsatile tinnitus for several months. MR angiography

at the outside hospital showed a suspected right-sided DAVF at the skull base. Subsequent diagnostic catheter angiograms showed a high-flow DAVF medial to the right jugular bulb (ie, ACC) fed by the neuromeningeal trunk of the right ascending pharyngeal artery, the intraosseous branches of the right occipital artery, the petrous branch of the right middle meningeal artery and the meningeal branches arising from the extradural right vertebral artery third segment. The DAVF also drained through the right ACC retrogradely into the right sigmoid sinus, antegradely to the right internal jugular vein, and which drained directly into the right posterior condylar vein (PCV), the right lateral condylar vein (LCV) and the right

Figure 1 Anatomic scheme of the anterior condylar confluence (*) and surrounding venous complex (anterior-posterior view) made with reference to several articles about the detailed anatomy.^{12–16} a, inferior petro-occipital vein; b, anastomotic channel with prevertebral venous plexus; c, anterior condylar vein; d, posterior condylar vein; e, lateral condylar vein; f, anterior internal vertebral venous plexus; g, vertebral artery venous plexus. CS, cavernous sinus; IPS, inferior petrosal sinus; JB, jugular bulb; SS, sigmoid sinus; IJV, internal jugular vein.

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suboccipital cavernous sinus (SCS) with some small drainage into the left side as well (figure 2A,B). A 6 F angled Envoy guiding catheter (Cordis Neurovascular, Miami lakes, Florida, USA) was advanced to the level of the right jugular bulb. Through the guiding catheter, a Hyperform balloon 7 mm×7 mm (Covidien/ev3 Neuromuscular) was then advanced over an Xpedion 10 guidewire (Covidien/ev3 Neuromuscular) via the right ACC into the right PCV. Control angiograms with balloon inflation in the ACC demonstrated a significant reduction of the arteriovenous shunting related to the fistula, indicating that this was indeed the location of the venous end of the fistula. An Echelon 14 microcatheter (Covidien/ev3 Neurovascular) was positioned in the distal aspect of the venous pouch (ie, the PCV). An additional Echelon 14 microcatheter was then placed in the proximal aspect of the venous pouch followed by sequential coil embolization of the PCV and the ACC using seven Axium coils (Covidien/ev3 Neurovascular). Once the loose coil mesh was formed in the venous pouch, Onyx embolization was performed using the coil mesh as a scaffold. The second Echelon 14 microcatheter was pulled back into the coil mass and Onyx was injected into the venous pouch. The standard technique was used, with preparation of the microcatheter with 0.34 ml dimethyl sulfoxide (DMSO) followed by slow injection of Onyx 34 embolic material. The Onyx successfully penetrated into the coil mass and took the shape of the irregular shaped venous pouch. Approximately 1.3 ml Onyx 34 was used to completely obliterate the DAVF. After the procedure the patient complained of some swallowing difficulty and very mild

tongue weakness probably due to hypoglossal nerve palsy. These symptoms disappeared after 2 weeks. The follow-up angiogram 7 months after the treatment demonstrated complete obliteration of the fistula without any recurrence (figure 2D).

Case 2

A 46-year-old woman presented with a persistent right-sided pulsatile tinnitus. The diagnostic angiograms showed a DAVF at the most medial aspect of the right ACC fed by the bilateral neuromeningeal trunk of the ascending pharyngeal artery, the transosseous branches of the right occipital artery and the meningeal branches of the vertebral artery third segment, draining retrogradely through the venous pouch into the right sigmoid sinus and antegradely to the right internal jugular vein, the right SCS via the right LCV and to the MS via the right anterior condylar vein (ACV) (figure 3A,B). A 6 F Envoy guiding catheter was advanced to the right internal jugular vein. An Echelon 14 microcatheter could be navigated into the venous pouch where the fistula opened. A superselective angiogram via the microcatheter showed relatively small drainers arising from the venous pouch, and the risk of Onyx migration was expected to be small. Onyx 34 was therefore slowly infused into the venous pouch without adjunctive use of coils (figure 3C). The post-embolization angiogram after injection of 0.1 ml Onyx demonstrated complete obliteration of the fistula. Her previously noted tinnitus was resolved immediately after the procedure without any new symptoms. There was no cranial neuropathy before or after the procedure. A follow-up angiogram 5 months

Figure 2 Right common carotid angiogram preoperatively ((A) anterior-posterior and (B) lateral views) showing the right-sided anterior condylar confluence (ACC) dural arteriovenous fistula (DAVF) (black large arrow) which drained directly into the right posterior condylar vein (PCV; arrowheads), the right lateral condylar vein (small black arrows) and the right suboccipital cavernous sinus (white large arrow). The balloon microcatheter was advanced via the jugular bulb into the ACC and the PCV. (C) Postoperative Xper CT scan showing the coils placed at the right hypoglossal canal and posterior condylar canal. (D) Follow-up angiogram 7 months postoperatively showing complete angiographic closure of the DAVF.

Figure 3 Preoperative right external carotid angiogram ((A) anterior-posterior and (B) lateral views) showing the right-sided anterior condylar confluence dural arteriovenous fistula (large black arrow) which drains into the right anterior condylar vein (arrowhead), the right lateral condylar vein (small black arrows) and the right suboccipital cavernous sinus (large white arrow). (C) Onyx 34 was injected into the venous pouch at the fistula site. (D) Postoperative Xper CT scan showing the coils placed just in front of the right hypoglossal canal. (E) Follow-up angiogram 5 months after embolization showing complete angiographic closure of the fistula.

after the procedure showed persistent obliteration of the fistula (figure 3E).

Case 3

A 68-year-old woman presented with left-sided pulsatile tinnitus and the MR angiogram suggested the presence of a DAVF at the left side of the skull base. The diagnostic angiograms prior to treatment showed a fistula at the left enlarged ACC adjacent to the left jugular bulb fed by the neuromeningeal trunk of the left ascending pharyngeal artery, the transosseous branches of the left posterior auricular artery, the meningeal branches of the left vertebral artery third segment and the left dorsal clival artery arising from the meningohypophyseal trunk of the left internal carotid artery. It drained retrogradely into the left sigmoid sinus and antegradely into the left internal jugular vein via the small venous pathway connecting the internal jugular vein and the ACC (figure 4A). The entire microsystem was advanced to the level of the left jugular bulb and subsequently into the ACC at the fistula site. Two Axiom 3D coils were deployed and roughly packed within the ACC to reduce the blood flow (figure 4B). The tip of the microcatheter was then placed into the coil mass and controlled infusion of Onyx 34 was performed which

penetrated the coil interstice without distal migration (figure 4C). The post-embolization angiogram demonstrated no evidence of a residual fistula and preservation of the normal venous flow of the left internal jugular vein (Figure 4D, E, F). The angiographic result had not changed in the follow-up angiogram at 6 months. No complications were noted during the procedure.

DISCUSSION

The ACC is an extracranial venous structure located anteromedially and inferiorly to the jugular foramen, and just anterolaterally to the hypoglossal canal. Its size is generally between 3 and 5 mm in an anterior view and approximately 2 mm in its ventro-dorsal extension; the structure is present in all humans.^{12–14} This venous confluence merging with the surrounding venous pathways was first reported as the petrosal confluence by Katsuta *et al* in 1997¹⁵ and the term ACC was coined by San Millan Ruiz *et al* in 2002.¹² The ACC connects with various venous structures including the MS and anterior internal vertebral venous plexus via the anterior condylar vein (ACV; actually, it is the venous plexus in the hypoglossal canal), the jugular vein or the suboccipital cavernous sinus (SCS, a vertebral venous

Figure 4 (A) Preoperative left CCAG showing the left-sided anterior condylar confluence (ACC) dural arteriovenous fistula (large black arrow) which drained into the left internal jugular vein via the small venous pathways (small black arrows). (B) The coils were deployed and roughly packed within the enlarged ACC. (C) Onyx 34 was penetrated into the coil interstice and the venous pathway connecting the ACC and the internal jugular vein. (D, E, F) The left CCAG postoperatively showed complete angiographic closure of the fistula and there was no evidence of Onyx migration into the internal jugular vein. (E) Postoperative Xper CT scan showing the coils placed just in front of the right hypoglossal canal CCAG, common carotid angiogram.

plexus surrounding the horizontal portion of the third segment of the vertebral artery) via the LCV, the inferior petro-occipital vein (coursing extracranially through the petro-occipital suture), the cavernous sinus through the foramen of lacerium (ie, internal carotid artery venous plexus of Rektorsik), as shown in figure 1.^{12–16} The ACC often connects with the junction between the inferior petrosal sinus and the jugular bulb via several emissary veins. Thus, understanding the complex venous anatomy at the skull base is essential for planning a proper endovascular approach and also for consideration of the symptoms of ACC DAVFs.

With respect to the arterial supply of the ACC DAVF, the neuromeningeal trunk of the ascending pharyngeal artery is the main feeder in almost all cases, and the mastoid branches of the occipital artery, the petrosal branch of the middle meningeal artery, the posterior auricular artery and the posterior meningeal branch of the vertebral artery also supply the ACC DAVF to varying degrees.⁹

The ascending pharyngeal artery consists of two major trunks, the pharyngeal trunk and the neuromeningeal trunk. From the neuromeningeal trunk arise the hypoglossal branches, jugular branch, internal auditory branch and clival branches, with the former two being the most important branches implicated in ACC DAVFs. The hypoglossal branch traverses the hypoglossal canal to supply the posterior fossa meninges and the vasa nervorum of cranial nerve XII. The jugular branch

extends to the posterior fossa to the jugular foramen and supplies the vasa nervorum of cranial nerves IX, X and XI.⁶ In addition, the neuromeningeal trunk of the ascending pharyngeal artery has an anastomosis with the vertebral artery through the odontoid arch, and with the cavernous internal carotid artery through the dorsal clival artery.¹⁷

Several treatment options can be used to obliterate ACC DAVFs. To our knowledge, four cases of transarterial embolization with a liquid embolic agent,^{6 11 18 24} cases of transvenous embolization with coils,^{1–3 6–11} one case of direct surgery and five cases of observation alone have been reported previously.^{1 19 20} Transarterial embolization can be performed, although this is generally considered less effective and carries a significant risk of lower cranial nerve palsies (IX–XII) and embolic stroke, as described above, particularly when using a liquid embolic agent.²¹ Neurointerventionalists must have a comprehensive understanding of the local anatomy and associated anastomoses to minimize these risks.²⁰ Transvenous embolization appears to be the most effective treatment for eliminating ACC DAVFs when venous access is available. In most cases the microcatheter is navigated to the ACC through the jugular vein and the ACC, which is considered to be the simplest approach to the ACC. Other approaches have been attempted because of the hypoplasia, tortuosity or stenosis of the above approach route.^{10 11} Although most ACC DAVFs can be successfully treated by transvenous embolization using coils,

it should be kept in mind that coil overpacking in the ACV may cause postoperative hypoglossal nerve palsy.^{1 3 9} Of 24 previous cases of transvenous coil embolization, three patients presented with transient or permanent hypoglossal nerve palsy postoperatively.^{1 9} The hypoglossal canal is an osseous tubular structure and the ACV, ascending pharyngeal artery and hypoglossal nerve are present together in this structure. The ACC DAVF resembles a cavernous DAVF. Several authors have reported the feasibility of transvenous Onyx embolization for a cavernous DAVF.^{22–24} They advocated that the non-adhesive and cohesive properties of Onyx are suitable for transvenous casting of the cavernous sinus, and it has the potential to avoid the undesirable compression of the adjacent cranial nerves and obtain sufficient packing rates.²² On the other hand, they point out potential adverse effects of Onyx embolization including direct neurotoxicity of DMSO on the cranial nerves within the cavernous sinus and retrograde reflux of the Onyx into the vital feeding arteries.^{22–24} In our series, one case treated with Onyx and coils presented with transient swallowing difficulty probably due to mild hypoglossal nerve palsy immediately after the procedure. Concerning the cause of this adverse event, we must consider not only coil overpacking but also DMSO neurotoxicity or retrograde reflux of the Onyx into the vasa nervorum arising from neuromeningeal trunk. With respect to venous migration of the Onyx, previously deployed coils can slow the fistula flow and provide secured anchoring to the Onyx cast and avoid inappropriate migration of the Onyx.^{22 23} This method is particularly useful for cases of high-flow DAVF (cases 1 and 3). In cases with a discrete venous pouch (case 2), the Onyx cast is stable so that the combined use of coils is not always necessary.

Learning points

- ▶ Onyx embolization of ACC DAVFs is feasible and, for high-flow lesions in particular, the combined use of coils seems to provide an adequate occlusive effect with acceptable stabilization.
- ▶ Conformability of the Onyx cast reduces the overall volume of embolic material needed to achieve complete obliteration of the DAVFs so that the chance of unnecessary obliteration of adjacent vital venous structures and the mass effect onto the lower cranial nerves may be reduced.
- ▶ Disadvantages include the possibility of DMSO toxicity and Onyx reflux into vital feeding arteries.
- ▶ Further investigations are needed to clarify fully the utility and safety of the procedures using Onyx.

Competing interests None.

Patient consent Obtained.

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