CASE REPORT

Anterior condylar confluence dural arteriovenous fistula: a rare cause of hoarseness

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SUMMARY

Hoarseness secondary to an anterior condylar confluence (ACC) dural arteriovenous fistula (DAVF) has not been previously described. We present a 58-year-old patient with a 3-week history of progressive unilateral left-sided headaches and hoarseness. Nasolaryngoscopy and CT neck showed the presence of unilateral vocal cord palsy with no identifiable cause along the expected course of the recurrent laryngeal nerve. MRI revealed an incidental finding of abnormal serpiginous vessels in the left hypoglossal canal which led to a diagnostic cerebral angiogram, confirming the presence of an ACC DAVF. The patient underwent transvenous coil embolisation with subsequent resolution of arteriovenous shunting and symptoms. Follow-up MRI at 6 months showed no recurrence and there was complete resolution of clinical symptoms.

BACKGROUND

Dural arteriovenous fistula (DAVF) is described as an abnormal arteriovenous shunt occurring in the dura. It is supplied primarily by the meningeal arteries and drains via the meningeal veins, cortical veins or the dural venous sinuses.¹

The anterior condylar confluence (ACC) is located at the external orifice of the hypoglossal canal and provides venous anastomotic connections between the anterior condylar vein, internal jugular vein, lateral condylar vein, inferior petrosal sinus, inferior petro-occipital vein and the prevertebral venous plexus.² Presence of a DAVF in the ACC is known to be associated with hypoglossal nerve palsy.³ Other associations include ocular symptoms⁴ as well as pulsatile tinnitus.⁵

We present a rare case of unilateral vocal cord and vagal nerve palsy secondary to an ACC DAVF which has not been previously described. Patient's symptoms of hoarseness and headaches subsequently resolved after successful embolisation of the DAVF.

CASE PRESENTATION

A 58-year-old Chinese man presented at the emergency department with 3 weeks of unilateral left-sided headaches and hoarseness of voice. No significant past medical, surgical or family history was noted. Physical examination was unremarkable. No neurological deficit or evidence of intracranial hypertension was detected.

INVESTIGATIONS

Left-sided unilateral vocal cord palsy was noted on nasolaryngoscopy. CT neck was performed

showing imaging features of left-sided unilateral vocal cord palsy (figure 1A). No identifiable aetiology was detected along the expected course of the recurrent laryngeal nerve. It was on retrospective review of the images and clinical examination that paralysis of the ipsilateral soft palate was also noted (figure 1B).

MRI brain stroke protocol was performed to exclude an intracranial aetiology. The MRI brain was largely unremarkable with no infarct or signal abnormality detected in the brain stem or cerebellum. It was only on the time-of-flight magnetic resonance angiography (MRA) that abnormal flow-related enhancement in the left hypoglossal canal as well as transosseous communication with the jugular foramen (figure 2) was detected, suspicious for a vascular malformation.

Additional clinical history was obtained based on the MRA findings, revealing a transient 1 month history of pulsatile tinnitus which spontaneously resolved a few months prior. No dysphagia, hypoglossal nerve or other cranial nerve palsy was elicited. Visual acuity and fields were intact and there was no chemosis or ocular hypertension.

Digital subtraction cerebral angiogram showed an ACC DAVF with multiple dural arterial feeders including the hypoglossal branch of the anterior meningeal artery arising from the left vertebral artery as well as the bilateral neuromeningeal trunks of the ascending pharyngeal arteries from the external carotid arteries (figure 3). Venous drainage was primarily via the left anterior condylar vein into the anterior condylar plexus as well as into the left internal jugular vein. Reflux of venous drainage intracranially into the sigmoid and left transverse sinus via the left occipital emissary vein was noted. There was no cortical venous reflux in keeping with a Cognard type IIA DAVF.

Figure 1 (A) CT neck demonstrates ballooning of left laryngeal ventricle, in keeping with unilateral left vocal cord palsy. (B) CT neck demonstrates asymmetry and paralysis of the ipsilateral soft palate.

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Figure 2 (A) MRA shows abnormal flow signal in the region of the left hypoglossal canal (arrow). (B) Time-of-flight MR angiography shows abnormal flow signal and a tangle of vessels at the left hypoglossal canal with transosseous flow to the jugular foramen (black arrow). The prominent left anterior condylar vein (white arrow) is also demonstrated. MR, magnetic resonance ; MRA, magnetic resonance angiography.

TREATMENT

Right common femoral vein and left common femoral artery access was obtained. Arterial inflow was controlled via the left and right ascending pharyngeal arteries. The venous pouch in the ACC was entered with a Prowler 10/Terumo GT12 microcatheter and wire system and embolised with a total of 11 detachable coils.

OUTCOME AND FOLLOW-UP

Postembolisation angiograms confirmed cessation of arteriovenous shunting and preservation of arterial parent vessels

(figure 4). Patient reported improvement of symptoms shortly after embolisation with resolution of hoarseness on clinic follow-up at 2 weeks after discharge. Complete resolution of DAVF was confirmed on MRI repeated at 6 months after discharge. Laryngoscopy examination at 6 months also documented recovery of the left vocal cord palsy.

DISCUSSION

Intracranial DAVF constitutes 5–15% of all intracranial vascular malformations. The majority occurs in the transverse–sigmoid sinus or the cavernous sinus. Intracranial DAVFs involving the hypoglossal canal and the ACC are a rare occurrence, forming only 5% of intracranial DAVF.⁶ Postulated aetiologies of intracranial DAVF include venous thrombosis, transcranial surgery and head trauma.

The main feeding arteries of the ACC DAVF typically arise from the ascending pharyngeal and occipital artery of the ipsilateral external carotid artery as well as branches from the vertebral arteries, specifically, the hypoglossal branch arising from the neuromeningeal trunk of the ipsilateral ascending pharyngeal artery and anterior/posterior meningeal arteries arising from the vertebral artery forming the odontoid arcade which anastomose with the hypoglossal branch. Contribution from the contralateral ascending pharyngeal artery is via the anastomosis of the clival branches. The shunt point typically occurs at the anterior condylar vein, draining into the ACC. Similar main arterial feeders to the ACC DAVF and shunt point was also seen in our patient (figure 3).

Figure 3 (A) Oblique view of the left vertebral artery demonstrates the fistula supplied by the hypoglossal branch of the anterior meningeal artery arising from the left vertebral artery with early venous shunting into the ACC, IJV and anterior internal vertebral venous plexus (arrow). (B) Lateral projection of the superselected left ascending pharyngeal artery shows the branches arising from the neuromeningeal trunk supplying the fistula with early venous drainage into the anterior condylar confluence, suboccipital plexus and anterior internal vertebral venous plexus. (C) Lateral projection of the superselected right ascending pharyngeal artery shows branches arising from the pharyngeal and neuromeningeal trunks supplying the fistula with early venous shunting into the anterior condylar confluence. ACC, anterior condylar confluence; IJV, internal jugular vein.

Figure 4 (A) Anteroposterior projection of the left vertebral artery post embolisation shows resolution of the DAVF with coil mass in the left sigmoid sinus and internal jugular vein. (B) Oblique projection of the left ECA post embolisation shows resolution of the DAVF. (C) Lateral projection of the right ascending pharyngeal artery post embolisation shows resolution of the DAVF. DAVF, dural arteriovenous fistula.

Table 1 Causes of proximal vagal nerve paralysis

Location	Aetiology	Examples
Carotid space	Trauma	Transection, carotid artery dissection or pseudoaneurysm
	Tumour	Schwannoma, neurofibroma, cervical adenopathy
	Mononeuritis	Viral, diabetic, idiopathic
	Iatrogenic	Surgery
Jugular foramen	Tumour	Glomus jugulare, schwannoma, meningioma, metastasis
	Trauma	Base of skull fracture
	Infarction	
Brainstem	Tumour	
	Demyelination	
	Haemorrhage	
	Infection	

Reported symptoms associated with ACC DAVF are ipsilateral pulsatile tinnitus,⁵ unilateral hypoglossal nerve palsy³ from the vascular compression of the hypoglossal nerve in the hypoglossal canal as well as chemosis/proptosis⁴ secondary to retrograde flow from the ACC into the ophthalmic vein via the cavernous sinus and the inferior petrosal vein. Our patient presented with symptoms of high vagal nerve paralysis without ocular symptoms or hypoglossal nerve palsy but reported a 1-month history of pulsatile tinnitus which spontaneously resolved.

Unilateral paralysis of the vocal cord with involvement of the ipsilateral soft palate as depicted in this case suggests a proximal disruption of the vagal nerve.⁷ This is because the muscles of the soft palate, except for the tensor veli palatini, are supplied by the pharyngeal plexus arising from the pharyngeal branches of the proximal vagal nerve. On CT, this may be identified by asymmetry of the soft palate, ipsilateral atrophy of the pharyngeal constrictor muscles or contralateral deviation of the uvula.⁷ Imaging features of unilateral vocal cord paralysis include ipsilateral pyriform sinus dilation, medial positioning and thickening of ipsilateral aryepiglottic fold and ipsilateral laryngeal ventricle dilation.⁸

Many causes of central-proximal vagal nerve paralysis have been described (table 1). In the case presented, where symptoms of proximal vagal nerve paralysis have not been previously described, we postulate two possible causes for this. The first is ischaemic neuropathy due to shunting of blood away from the vasa nervosum of the vagal nerve (supplied by the jugular branch of the neuromeningeal trunk). The second possibility is neuropathy secondary to the venous congestion because of the arteriovenous shunting. No infarct or fluid-attenuated inversion recovery (FLAIR) signal abnormality is seen in the medulla and the cranial nerve X nucleus to suggest a central cause.

Several classification systems for DAVFs are available such as the Borden *et al*⁹ or Cognard *et al*¹⁰ classifications (table 2) which provide a means for risk stratification, guiding treatment decision. Presence of cortical venous reflux (Borden II/Cognard IIb and above) with an annual mortality rate of 10.4%, annual risk of intracranial haemorrhage of 8.1% and annual risk of non-haemorrhagic neurological deficit of 6.9%¹¹ would necessitate prompt treatment. Treatment is also indicated in patient's presenting with aggressive symptoms such as intracranial haemorrhage, neurological/cranial nerve deficits, orbital symptoms, severe headache or seizures. Spontaneous regression of DAVF has been reported which may explain the spontaneous resolution of

Table 2 Borden and Cognard classification of DAVFs

	Venous drainage	Drainage pattern
Borden classification		
I	Venous sinus/meningeal vein	Antegrade
II	Venous sinus/meningeal vein	Cortical venous reflux (CVR)
III	Cortical vein	CVR only
Cognard classification		
I	Venous sinus	Antegrade
IIa	Venous sinus	Retrograde
IIb	Venous sinus	Antegrade with CVR
IIa+b	Venous sinus	Retrograde with CVR
III	Cortical vein	CVR only
IV	Cortical vein with venous ectasia	CVR only
V	Spinal perimedullary veins	

DAVF, dural arteriovenous fistula.

our patient's tinnitus. In the absence of aggressive symptoms, patients (Borden I/Cognard I/IIa) may improve with conservative treatment. However, the eventual decision to treat should involve discussions within a multidisciplinary team of neurosurgeons, neurologists and interventional neuroradiologists. Our patient's DAVF was classified as a Cognard IIa. Our team's decision to treat was due to the presence of cranial nerve deficit (vagal nerve) which is the likely cause of the patient's hoarseness.

Successful treatment of the ACC DAVF requires endovascular embolisation or surgical disconnection of the fistulous point at the anterior condylar vein. Onyx-18,¹² glue (n-butyl-2-cyanoacrylate) or coil packing can be employed in endovascular embolisation. Transarterial approach for embolisation should be avoided due to possible intracranial and vertebral artery communication via the branches of neuromeningeal trunk.¹³ The combined transarterial and transvenous approach with superselective catheterisation of the arterial feeders can be used to control the inflow for concurrent transvenous embolisation. The transvenous approach for embolisation of ACC DAVF has been established to be safe and effective but care should be taken to avoid overpacking in the anterior condylar vein as this may result in hypoglossal nerve palsy from mass effect.

In conclusion, we describe a rare case of unilateral vocal cord and proximal vagal nerve palsy caused by a DAVF which resolved after successful transvenous embolisation of the DAVF.

Learning points

- ▶ Anterior condylar confluence (ACC) dural arteriovenous fistula (DAVF) is a rare cause of ipsilateral proximal vagal nerve and vocal cord palsy.
- ▶ Proximal vagal nerve palsy can be differentiated from recurrent laryngeal nerve palsy by the additional involvement of the soft palate, pharyngeal constrictor muscles and uvula.
- ▶ Transvenous coil embolisation is a safe and effective treatment for ACC DAVF.

Contributors GSC prepared and revised the manuscript. WEHL performed the treatment, edited and approved the final version of the manuscript.

Competing interests None declared.

Patient consent Obtained.

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