CASE REPORT

Development of syndrome of inappropriate antidiuretic hormone secretion (SIADH) after Onyx embolisation of a cavernous carotid fistula

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SUMMARY

Patients with cavernous carotid fistulas (CCFs) can present with pituitary hypoperfusion and hypopituitarism; however, there are no previous reports of pituitary or hormonal abnormalities developing after CCF embolisation in an asymptomatic patient. We describe a patient with no hormonal abnormalities who developed syndrome of inappropriate antidiuretic hormone (SIADH) secretion after CCF embolisation. The patient had bilateral indirect CCFs, which were completely embolised via a transvenous approach, and was neurologically stable postoperatively and discharged. In the subsequent 2 weeks the patient was readmitted twice for acute hyponatraemia and a tonic-clonic seizure. Laboratory studies revealed severe SIADH. Clinical status and sodium levels improved after treatment. One year later the patient was weaned off all medications and remained neurologically stable. SIADH may be a delayed phenomenon after CCF embolisation. Given the proximity of embolised vessels to the pituitary's vascular supply, CCF treatment may result in flow disturbance, ischaemia and hormonal abnormalities.

BACKGROUND

Endovascular treatment for cavernous carotid fistulas (CCFs)¹ is indicated when patients present with proptosis, visual loss, cranial nerve palsies, elevated intraocular pressure or increased angiographic cortical venous filling.² Primary CCFs may cause pituitary enlargement,³ hypoperfusion or hypopituitarism.^{4,5} No previous cases of pituitary hormonal disturbance—specifically, syndrome of inappropriate antidiuretic hormone (SIADH)—have been reported after endovascular CCF treatment.

CASE PRESENTATION

History and examination

A 51-year-old woman with no significant medical history presented with 5 months of headaches and diplopia. Neuro-ophthalmological examination revealed a partial right sixth nerve palsy, bilateral injected sclera and proptosis. Initial CT angiogram of the head demonstrated an enlarged ophthalmic vein and right-sided cavernous sinus hypervascularity, concerning for an indirect CCF. Formal angiography was performed to delineate lesion vascularity and recommend treatment.

Treatment

Cerebral angiography demonstrated bilateral high-risk indirect CCFs (figures 1 and 2). An initial attempt was made to establish bilateral venous access to embolise each fistula separately. Microcatheter positioning could not be secured within the left-sided fistula draining pouch; thus embolisation was performed through the right-sided catheter. This catheter crossed over but did not fully reach the site of the left-sided pouch, so

Figure 1 (A) Right and (B) left external carotid artery anteroposterior angiography shows bilateral cavernous carotid fistulas with retrograde venous drainage into the right sphenoparietal sinus. Used with permission from Barrow Neurological Institute, Phoenix, Arizona.

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Figure 2 (A) Right internal carotid artery anteroposterior view angiography and (B) left common carotid artery anteroposterior view angiography further demonstrate the fistulae. Used with permission from Barrow Neurological Institute, Phoenix, Arizona.

coils could not be used for fistula occlusion. Instead, slow Onyx-18 infusion (Covidien, Mansfield, Massachusetts, USA) was used to reflux the pouch and cure the fistula. No steroids were infused during or after the case. There was no immediate suggestion of iatrogenic injury, and steroids are not routinely administered at our institution in the setting of CCF embolisation without evidence of new symptoms. Final angiograms demonstrated no residual shunting ([figure 3](#)).

Outcome and follow-up

The patient remained at neurological baseline after the procedure and was discharged on post-procedure day 1 without complication. Three days later she presented to an outside hospital after a generalised tonic-clonic seizure. No structural abnormalities were found on MRI with and without contrast. She was hyponatraemic (serum sodium 114 mEq/L) and treated with hypertonic saline, a 1500 mL/day fluid restriction and a high

salt diet. Full endocrine investigation was not completed and the patient was discharged 5 days later (serum sodium 133 mEq/L).

Nineteen days after CCF embolisation the patient presented to our institution with dizziness, nausea and malaise. Laboratory studies were consistent with SIADH: 123 mEq/L serum sodium, 256 mOsm/kg serum osmolality and 163 mEq/day urine sodium. Other endocrine test results were normal, indicating no concern of hypopituitarism. The serum sodium was gradually corrected by the endocrinology team with fluid restriction and hypertonic saline, eventually converting to oral salt supplementation, desmopressin antagonists (demeclocycline) and glucocorticoids (fludrocortisone). The patient never developed hypernatraemia or any sign of diabetes insipidus concerning for triphasic response. She was gradually weaned off all medications as an outpatient, and these were eventually discontinued at 12-month follow-up (serum sodium 140 mEq/L).

Figure 3 (A) Right and (B) left internal carotid artery post-embolisation anteroposterior angiography demonstrates no residual shunting. (C) Anteroposterior and (D) lateral images demonstrate the liquid embolic cast. Used with permission from Barrow Neurological Institute, Phoenix, Arizona.

DISCUSSION

To our knowledge, this is the first report of SIADH occurring after CCF embolisation. Known complications after Onyx embolisation of CCFs include cranial nerve neuropathies, embolic internal carotid artery events and transient haemodynamic instability.^{6 7} In our patient, we suspect that CCF embolisation caused thrombosis of small adjacent arterial fistula branches, resulting in devascularisation of the posterior pituitary gland. Disruption of the pituitary blood supply can cause dysregulation of hormones such as antidiuretic hormone, modulated by the hypothalamic-pituitary axis. The patient presented solely with SIADH and no other hormonal abnormality; thus, vascular disruption was most likely isolated to the posterior pituitary, without hypothalamic or anterior pituitary involvement.

Treatment was performed from the right-sided microcatheter due to inaccessibility on the left. We relied on the ability of Onyx to penetrate small arterioles and venules to achieve fistula occlusion. These properties, as well as the inflammatory

characteristics of liquid embolics, probably resulted in penetration of the vasculature supplying the posterior pituitary and the complication described. Alternatively, alterations in flow may have caused venous congestion and ischaemia, or dimethyl sulphoxide toxicity may have caused temporary hormonal axis alterations. The patient's rapid improvement and eventual medication independence suggests the pituitary injury was transient and incomplete.

All patients experiencing severe hyponatremia (sodium <125 mEq/mL) after endovascular procedures should undergo formal endocrine evaluation. Patients with symptomatic hyponatraemia secondary to SIADH, refractory to initial therapies, can be treated with vasopressin receptor agonists. Conivaptan is most commonly used.

Contributors Conception and design: TC, AD, CGM; acquisition of data: TC, MYSK; interpretation of data: TC, MYSK, AD; drafting the article: TC, MYSK, AD; critically revising the article: AD, FCA, CGM; reviewing final version of the manuscript and approving submission: CGM.

Competing interests None declared.

Patient consent Obtained.

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Learning points

- ▶ Although cavernous carotid fistulas (CCFs) are typically treated safely and effectively with endovascular embolisation, given their close proximity to the pituitary gland's vascular supply and venous drainage, endovascular embolisation of high-risk CCFs may result in devascularisation of the pituitary and hormonal dysregulation.
- ▶ Patients experiencing post-embolisation hormonal dysregulation should be first evaluated for structural abnormalities and subsequently treated by an endocrinologist. In our experience, the hormonal dysregulation phenomenon is temporary and can resolve with appropriate outpatient management.

REFERENCES

- 1 Barrow DL, Spector RH, Braun IF, *et al.* Classification and treatment of spontaneous carotid-cavernous sinus fistulas. *J Neurosurg* 1985;62:248–56.
- 2 Ducruet AF, Albuquerque FC, Crowley RW, *et al.* The evolution of endovascular treatment of carotid cavernous fistulas: a single-center experience. *World Neurosurg* 2013;80:538–48.
- 3 Sato N, Putman CM, Chaloupka JC, *et al.* Pituitary gland enlargement secondary to dural arteriovenous fistula in the cavernous sinus: appearance at MR imaging. *Radiology* 1997;203:263–7.
- 4 Jamieson KG. Bilateral carotico-cavernous fistulae. hypopituitarism from bilateral carotid ligation for surgical cure. *Aust NZ J Surg* 1964;34:1–10.
- 5 Yarandi SS, Khoshnoodi MA, Chandra P. Hypopituitarism caused by carotid cavernous fistula. *Intern Med* 2011;50:1105–8.
- 6 Hassan T, Rashad S, Aziz W, *et al.* Endovascular modalities for the treatment of cavernous sinus arteriovenous fistulas: a single-center experience. *J Stroke Cerebrovasc Dis* 2015;24:2824–38.
- 7 Amiridze N, Darwish R. Hemodynamic instability during treatment of intracranial dural arteriovenous fistula and carotid cavernous fistula with Onyx: preliminary results and anesthesia considerations. *J Neurointerv Surg* 2009;1:146–50.

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