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Case Report

Panhypopituitarism caused by an unruptured giant cavernous internal carotid artery aneurysm compressing the pituitary gland treated with a flow-diverting stent: A case report

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ABSTRACT

Background: Internal carotid artery (ICA) aneurysms extending into the intrasellar region that mimics pituitary tumors and leads to pituitary dysfunction are relatively rare. The treatment for aneurysms includes surgery and endovascular procedures. However, functional recovery of the pituitary gland is difficult.

Case Description: We report a case of a 43-year-old woman who presented with severe headaches and generalized malaise. Magnetic resonance imaging (MRI) revealed a giant unruptured cavernous ICA aneurysm that pushed the pituitary stalk contralaterally. A baseline endocrinological examination suggested panhypopituitarism. Hypopituitarism was treated with hormone replacement therapy, which improved the patient's symptoms of headaches and malaise after 4 days. The aneurysm was treated using a pipeline flow-diverting stent. Two years later, the aneurysm had reduced to half of its maximum diameter, and the pituitary stalk was visible on MRI. Hormone loading tests 1 week postoperatively showed almost no response. At postoperative 6 months, there was a trend toward improvement.

Conclusion: Flow-diverting stent deployment is useful for large or giant carotid artery aneurysms with pituitary gland compression.

Keywords: Cavernous internal carotid artery aneurysm, Flow-diverting stent, Panhypopituitarism

INTRODUCTION

It is relatively rare for internal carotid artery (ICA) aneurysms to extend into the sella and leads to pituitary dysfunction. However, it can occur with large or giant aneurysms. Mechanical compression and pulsation of aneurysms, and disruption of microvascular supply can cause hormone depletion.[10] Although functional improvements of the pituitary gland caused by an unruptured intrasellar aneurysm after open surgery or endovascular treatment have been reported,[2,4-7,9,11] there have been no reports showing hormonal improvement with a reduction in the aneurysmal mass effect on magnetic resonance imaging (MRI) in cases treated with flow-diverting stent. Here, we described a patient with a giant thrombotic ICA aneurysm causing pituitary impairment which was successfully treated with endovascular deployment using a flow-diverting stent, with subsequent reduction in the mass effect on MRI.

CASE DESCRIPTION

A 43-year-old woman presented with severe headaches and generalized malaise for 5 days. Endocrine investigation revealed hypopituitarism with markedly decreased levels of early morning cortisol, thyroid hormone with low thyroid-stimulating hormone, growth hormone, and insulin-like growth factor 1. Physical examination was unremarkable. There were no visual deficits on ophthalmological examination. MRI and magnetic resonance angiography showed a significant mass effect on the pituitary gland, with a giant right cavernous ICA thrombotic aneurysm measuring 26.4 mm at the largest diameter [Figures 1a and b]. Digital subtraction angiography revealed a right cavernous ICA aneurysm [Figures 1c and d]. Considering the possibility of pituitary apoplexy, hormone replacement therapy with hydrocortisone (40 mg/day) and levothyroxine (25 mg/day) was initiated without a hormone loading test. Bayaspirin tablets (100 mg/day) and clopidogrel tablets (75 mg/day) were also administered for 2 weeks before the treatment. One month after the beginning of hormone replacement therapy, the aneurysm was treated using a pipeline Flex device (size: 5.0 mm × 16 mm; Medtronic). Immediately after pipeline placement, the flow-diverting effect and good patency of the stent were confirmed [Figures 2a-c]. The patient tolerated the procedure well, without any complications. MRI at 1 week, 9 months, 16 months, and 24 months after endovascular surgery showed that the cerebral aneurysm thrombosed and shrank accordingly [Figures 3a-d]. One week after endovascular surgery, loading tests of the hypothalamic hormones (corticotropin-releasing hormone, thyrotropin-releasing hormone, and luteinizing hormone-releasing hormone) and growth hormone-releasing peptide-2 demonstrated panhypopituitarism. Although complete functional recovery of the pituitary gland was not achieved at 6 months postoperatively, the hormone loading test confirmed functional improvement of the pituitary gland [Figure 4]. The patient was clinically stable but still required hormone replacement therapy (Cortil tablets [15 mg/day], Thyradin-s tablets [25 mg/day], Minirin melt OD tablets [120 mg/day], Duphaston tablets [10 mg/day], and estradiol tape [0.72 mg once a day]).

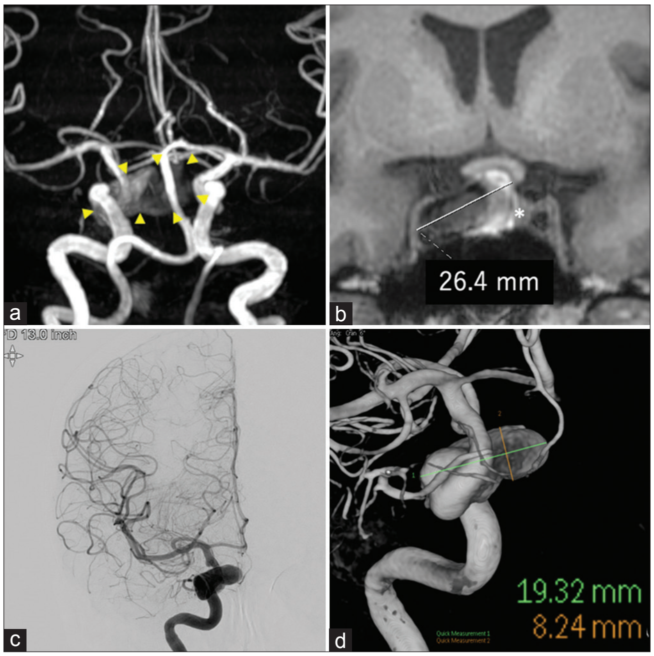

Figure 1: (a) Magnetic resonance angiography showing a giant right cavernous internal carotid artery (ICA) aneurysm. The aneurysm is surrounded by arrowheads. (b) T1-weighted coronal magnetic resonance imaging showing a thrombotic aneurysm measuring 26.4 mm at the largest diameter, as indicated by white line. The asterisk refers to the pituitary gland that is ablated by an aneurysm. (c) Digital subtraction angiography shows the right cavernous ICA aneurysm. (d) 3D rotational angiography shows aneurysm excluded thrombosis measuring at 19.3 mm at largest diameter, as indicated by green line. The height of the aneurysm is 8.2 mm, indicated by the orange line.

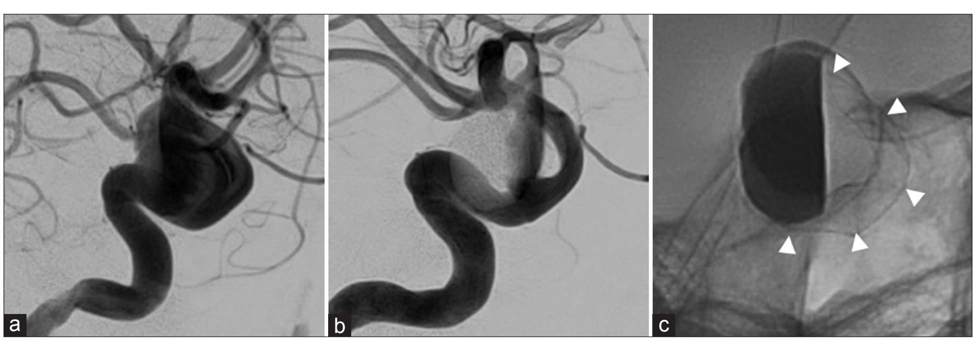

Figure 2: (a) The working angle of digital subtraction angiography (DSA) before flow diverter deployment is presented. (b) DSA immediately after deployment of the flow-diverting stent for the aneurysm showing an eclipse sign. (c) A flow-diverting stent was deployed and radiographs were taken. The flow diverter was confirmed to cover the neck of the aneurysm. Arrowheads indicate flow-diverting stents.

DISCUSSION

This is the first case to demonstrate that deployment of a flow-diverting stent for a cerebral aneurysm associated with hypopituitarism resulted in mass reduction on MRI and improved hormonal response over time. Pituitary dysfunction is estimated to have an annual incidence of 4.2 cases/100,000 and a prevalence of 45.5/100,000.[8] The most common cause of hypopituitarism is the mass effect of tumors such as pituitary adenomas, craniopharyngiomas, meningiomas, and gliomas. Large or giant ICA aneurysms may cause hypopituitarism by compressing the pituitary gland (a reported incidence of 0.17%).[3] There is a wide range of treatments for hypopituitarism caused by aneurysms. Open surgical procedures, such as clipping, proximal carotid ligation with high- or low-flow bypass, and endovascular therapy such as coiling and flow-diverting stent deployment, can improve hypopituitarism due to cerebral aneurysms [Table 1].[2,4,5,7,9,11] The time to functional improvement of the gland following treatment ranges from a minimum of 2 months to a maximum of 2 years.[5,7,9] In addition to our patient, two other cases of cerebral aneurysms with hypopituitarism treated with flow-diverting stent deployment have been reported. One showed functional improvement of the pituitary gland at 10 months after treatment, and the other showed no improvement at 1 month after treatment.[1,9] The former case did not include postoperative MRI or whether the pituitary gland displacement had improved. The latter case report also did not include their postoperative MRI findings. In the latter case, the pituitary function may have improved since the case only considered 1 month postoperatively. Factors that affect the recovery of pituitary function after treatment include the mass effect of the aneurysm, a decrease in pulsation, and ischemia of the pituitary gland. The present case demonstrated that hormonal function improved with a reduction in mass effect. Other factors may include the length of pituitary dysfunction, the degree of pituitary compression, and inflammation caused by thrombosis of the aneurysm spilled over into the pituitary gland. In our case, the mass effect improvement was important. The reduction of the aneurysm using a flow diverter played an important role in the functional improvement of the pituitary gland postoperatively.

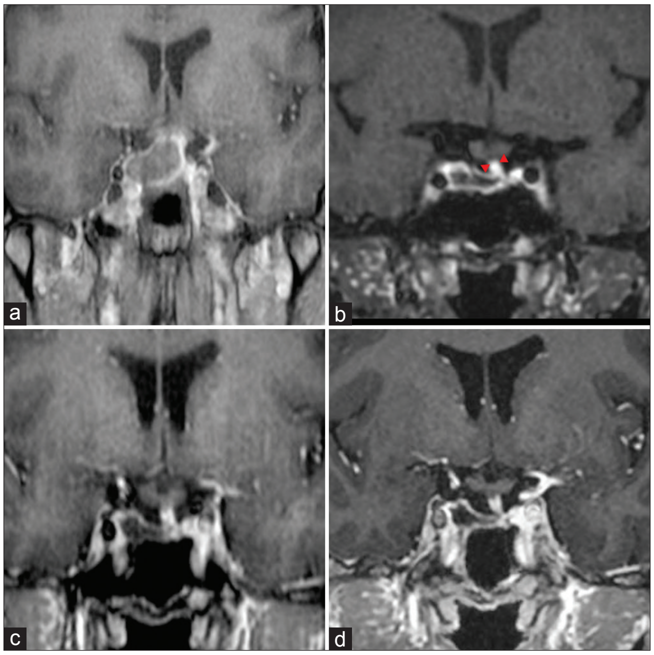

Figure 3: (a) Contrast-enhanced T1-weighted imaging 1 week after endovascular surgery is presented. It shows a tendency toward thrombosis, but the pituitary gland is compressed and cannot be seen clearly. (b) Contrast-enhanced T1-VISTA imaging performed 9 months after the endovascular surgery. The aneurysm shrank significantly. The pituitary stalk, circled by arrowheads, can now be observed. (c) Contrast-enhanced T1-weighted image 16 months after endovascular surgery the aneurysm was further reduced compared to 9 months after endovascular surgery. (d) Contrast-enhanced T1-weighted image 24 months after endovascular surgery. The aneurysm was generally unchanged in size 16 months after endovascular surgery.

Table 1: Review of literature on the improvement of pituitary function after surgical treatment of unruptured cerebral aneurysms with hypopituitarism.

Study	Age, Sex	Surgical procedure	Endocrinological evaluation/time
Verbalis *et al*., 1982	59, F	Clipping	Complete recovery/2 months
Krause *et al*., 1982	59, F	Clipping	Complete recovery/2 months
Kita *et al*., 1986	61, F	Clipping	Complete recovery/8 months
Fujii *et al*., 2008	81, M	Coiling	Partial recovery/1 year
Tan *et al*., 2015	56, M	FD	Complete recovery/10 months
Ono *et al*., 2017	56, M	PCO and high-flow bypass	Complete recovery/1 year
Qi *et al*., 2018	76, M	Coiling	Partial recovery/2 years
Our case	43, F	FD	Partial recovery/6 months

PCO: Proximal carotid artery occlusion, FD: Flow diverter

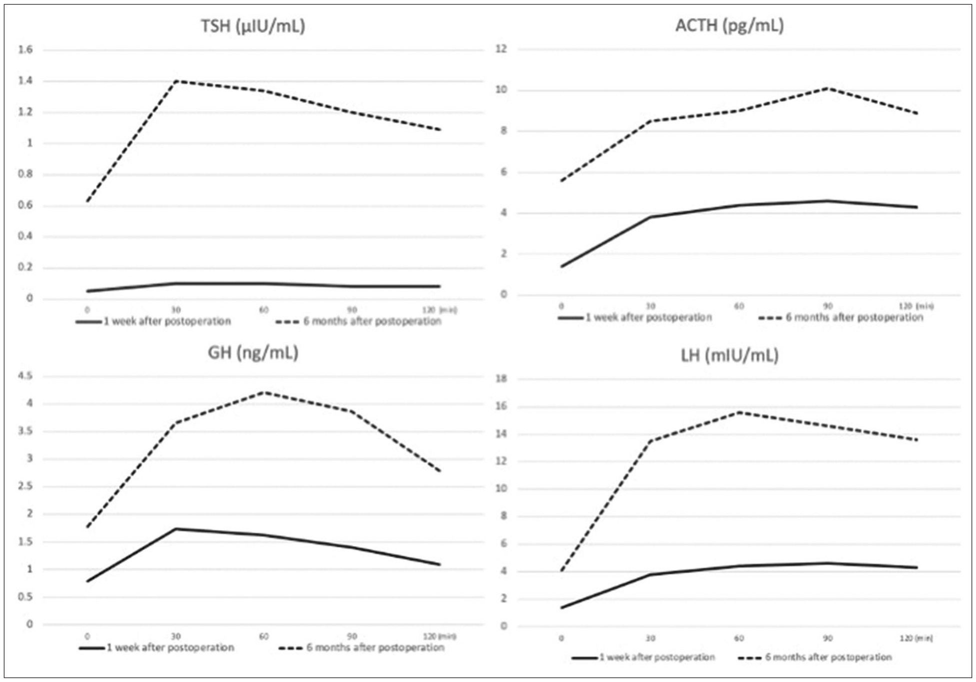

Figure 4: Time series graphs of hormone load tests. The solid and dotted lines indicate the date 1 week and 6 months after the operation, respectively. In the thyrotropin-releasing hormone loading test, the normal maximum thyroid-stimulating hormone level was more than 6 µIU/mL, but the measured value did not reach the normal value at 1 week and 6 months after surgery. In the corticotropin-releasing hormone loading test, the normal apex value of adrenocorticotropic hormone is more than twice the preloading value at more than 30 pg/mL, but the measured value did not reach the normal value at 1 week and 6 months after surgery. In the growth hormone-releasing peptide-2 loading test, the normal maximum growth hormone level is more than 9 ng/mL, but the measured value did not reach the normal value at 1 week and 6 months after surgery. In the follicle-stimulating hormone loading test, the normal maximum luteinizing hormone level is more than 5 times the preloading value, but the measured value did not reach the normal value at 1 week and 6 months after surgery. However, there is an improvement in responsiveness in all loading tests at 6 months compared to 1 week postoperatively.

CONCLUSION

We treated a panhypopituitarism-associated cerebral aneurysm with flow-diverting stent deployment. Postoperatively, the aneurysm was significantly reduced in size, and the response of pituitary function improved. Flow-diverting stent deployment may be an option for patients with large or giant ICA aneurysms which cause pituitary insufficiency.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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