Spontaneous Carotico-Cavernous Fistula Following Normal Labor

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INTRODUCTION

Carotico-cavernous fistula is an abnormal connection between the carotid artery and the cavernous sinus (1). The exact etiology of spontaneous carotico-cavernous fistula largely remains unknown. Of the various predisposing factors, pregnancy also has been associated with their development. There have been only few reported instances of carotico-cavernous developing immediately in the postpartum period following normal spontaneous vaginal delivery or abortion (2). The cases reported earlier resolved spontaneously and did not require active intervention.

Ours is probably the first case report of a spontaneously developing carotico-cavernous following labour. The fistula was not only confirmed angiographically but also successfully treated using the detachable balloon system.

CASE REPORT

A 28-year-old female patient weighing 47 kgs was admitted to the neurosurgical ward with presenting complaints of headache and vomiting since 2 months and diminished vision and swelling of both eyes since 1 month. Her symptoms started 5 days after her normal vaginal delivery, two months back.

On examination, she had bilateral proptosis, conjunctival prolapse and chemosis. Her extraocular muscle movement was restricted in all directions. Her visual acuity was 6/36 and 6/20 in the left and right eye, respectively. A Magnetic Resonance Imaging of the head showed multiple flow voids with left-sided cavernous sinus larger than the right. There was a prominent superior ophthalmic vein bilaterally, suggestive of carotico-cavernous fistula.

A transarterial cerebral angiogram was performed via femoral artery, which showed evidence of left carotico-cavernous from the ascending portion of the C4 segment of the left internal carotid artery. It drained anteriorly into the superior ophthalmic vein and posteriorly into the petrosal venous plexus, inferior petrosal sinus and through intercommunication to the opposite cavernous sinus.

Balloon embolization was planned to occlude the fistula. The procedure was done under general anesthesia, on patient's request. After premedication with Glycopyrrolate 0.2 mg intramuscularly, general anesthesia was induced with Fentanyl 100 mcg and Propofol 100 mg. Rocuronium 50 mg was given to facilitate endotracheal intubation. Anesthesia was maintained with isoflurane in O2 and N2O (1:2) mixture.

Monitoring was routine using the Datex-Ohmeda AS/3 (Helsinki, Finland) monitor. During the entire procedure, normocapnia was maintained.

Balloon embolization was done using a BAL 2 XRAY (BALT, extrusion, France) with a BALT guiding catheter of 8F. This is a 0.8-ml volume balloon, which on deflation has a length of 1.5 mm and when inflated measures 8 ±0.05 mm x 18 ±0.05 mm.

Following a successful embolisation, the neuromuscular blocked was reversed with neostigmine 2.5 mg and atropine 1.2 mg. The patient was kept under observation in the neurosurgical ICU for the next 24 hours.

The symptoms of the patient improved over the next 2 weeks and she was finally discharged with an improved vision.
DISCUSSION

Carotico-cavernous fistula has been a well-known entity since Travers first reported it in 1809 (4). Direct carotico-cavernous fistula represents direct connections between the internal carotid artery and the cavernous sinus, and may occur as a result of a ruptured aneurysm, trauma, collagen deficiency syndromes, fibro muscular dysplasia, arterial dissection or direct surgical trauma. Indirect carotico-cavernous fistula are usually supplied by dural branches of the external carotid artery but can be supplied by dural branches of the internal carotid artery. The exact cause of indirect carotico-cavernous fistula is unknown; however, factors associated with their development include pregnancy, sinusitis, trauma, surgical procedures and cavernous sinus thrombosis (8).

Pregnancy is an important precipitating factor in the occurrence of spontaneous carotico-cavernous fistula. Dandy and Follis reported the association of pregnancy with carotid cavernous fistula in 17 of 41 spontaneous cases, but Hamby had only 2 out of 27. Taniguchi has also reported carotico-cavernous fistula following a normal delivery and an abortion. Their diagnosis was confirmed by cerebral angiography (7). Newton et al reported 2 cases of fistula after delivery but no cerebral angiographic findings or detailed records of these patients exist (9). In the reported incidents, less emphasis has been laid on the management of these patients. Taniguchi has recommended conservative management, unless there is exacerbation of symptoms (10).

Ours is probably the first case of spontaneous carotico-cavernous fistula that was confirmed angiographically and treated with interventional neurovascular technique using a detachable balloon system.

Walker and Allegre in their report suggested arterial hypertension associated with gestation might rupture a pre-existing aneurysm in the cavernous sinus (11). Our patient was normotensive throughout her pregnancy and so this possibility is ruled out.

Pregnancy is associated with hemodynamic and hormonal changes. Hormone relaxin is known to induce changes in the fibrous tissue and leads to relaxation of the pelvic ligaments (12). These factors may have weakened her cerebral vessels, which along with the stress of labor could have possibly resulted in carotico-cavernous fistula.

The possibility of an occult is unlikely as the patient developed symptoms after the delivery. Moreover her gestational period was uneventful whence there was an increased possibility of development of symptoms and signs of carotico-cavernous fistula. Also this patient had her earlier two pregnancies uneventful.

Transarterial balloon embolization is the treatment of choice for carotico-cavernous fistula these days, (2m,4,11) which was done in our case. In most of the centers, balloon embolisation is usually done under local anesthesia. General anesthesia requires special consideration of maintaining hypercapnia during embolization. This is recommended to prevent the normal perfusion pressure breakthrough. The embolization in our case was done under general anesthesia maintaining normocapnia. There was a less concern in our minds regarding normal perfusion pressure breakthrough as the patient had symptoms of short duration. It is postulated that perfusion breakthrough occurs because of loss of normal auto-regulation from chronic arteriolar distension associated with vascular steal in patients with carotico-cavernous fistula (11).

The goal of therapy in carotico-cavernous fistula is to preserve and improve vision and return of the orbit and its contents to normal (3). Our patient was treated successfully as her signs and symptoms improved over the next two weeks.

To conclude, we present a rare case of symptomatic carotico-cavernous fistula developing as a result of stress of labor in a previously healthy woman.

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References

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