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Carotico-cavernous fistula following septorhinoplasty

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Abstract
Septorhinoplasty is a very common operation in otolaryngological practice. We report the second case of a carotico-cavernous fistula following septorhinoplasty. This case presented with very severe epistaxis before the appearance of the typical pulsating exophthalmos, ophthalmoplegia, headache and engorged veins on the right side of the face. Our case was treated by endovascular thrombosis with electrolytically detachable coils.

Key words: Carotid artery, internal; Cavernous sinus; Fistula; Rhinoplasty

Introduction

Traumatic carotico-cavernous fistula (CCF) following maxillo-facial trauma has been reported before (Zachariades and Papavassiliou, 1988; Keiser *et al.*, 1991; Nocini *et al.*, 1995). CCF has been reported following a rhinoplasty (Song and Bromberg, 1975), Le Fort I osteotomy (Lanigan and Tubman, 1987), and after external ethmoid and sphenoid surgery (Pederson *et al.*, 1981). We report the second case of CCF as a complication of septorhinoplasty. CCF should be diagnosed and treated as soon as possible to prevent blindness, CSF rhinorrhoea, stroke, severe epistaxis or even death.

Case report

A 34-year-old male with nasal deformity and blockage, electively underwent septorhinoplasty involving septoplasty, removal of the bony hump, and medial and lateral osteotomies with infracture of the nasal bones. One week post-operatively the plaster of Paris was removed, at which time he was asymptomatic and satisfied with the nasal profile and airway.

Three weeks post-operatively he presented to the outpatient department with severe bouts of epistaxis, reported to be lasting 10 minutes, mainly from the right nostril. A bleeding point was found on the anterior part of the nasal septum in the right nostril, which was cauterized with silver nitrate. He was symptom free for a further two weeks but then presented to the casualty department with another bout of severe epistaxis. He was admitted to hospital and had the nose packed. The bleeding continued despite conservative measures. The only other symptom the patient had developed was pressure on the right side of the nose.

As the conservative measures failed to stop the bleeding, he was taken to the operating theatre. Using a Lynch Howarth incision, the periosteum was raised from the medial wall of the orbit, the anterior ethmoid artery was identified and coagulated by diathermy. This procedure did not involve entering the ethmoid or sphenoid sinuses. His epistaxis ceased but the following week he developed proptosis, dilated veins on the side of the forehead, headache and increased intra-orbital pressure. An ophthalmologist was consulted and a clinical diagnosis of CCF was reached. The patient underwent a carotid artery angiogram, which confirmed the CCF (Figure 1).

He was treated by trans-arterial embolization using Guglielmi electrolytically detachable coils which closed the fistula (Figure 2) and his proptosis, and pressure symptoms disappeared instantly and six months after treatment he has remained totally symptom free.

Discussion

Rupture of the carotid artery within the cavernous sinus results in a CCF. Trauma is responsible for almost every case in individuals under 40 years of age, but in older age groups spontaneous communications sometimes develop. In the spontaneous variety, small aneurysms or a weakness of the muscular layer may cause the rupture of the carotid wall and in the traumatic variety, the disruption presumably occurs secondarily to bone fracture and laceration of the artery within the sinus. Arterial blood enters into this channel, thus gaining entry to the superior ophthalmic veins. These veins ultimately exit in the superior eyelid, communicating with the facial vein and draining into the jugular system (Grossman *et al.*, 1991).

The traditional concept of cavernous sinus has been that of a trabeculated network of venous channels contained in two layers of dura, with the internal carotid artery (ICA) and the sixth cranial nerve running within the trabeculation and with the third, fourth, and the first and second division of the fifth cranial nerves enclosed in the dura of the lateral wall of the sinus. More recent studies have shown that the cavernous sinus is, on the contrary, a plexus of veins of various sizes dividing and anastamosing with each other, that incompletely surrounds the internal carotid artery (Isamat, 1987). This description was confirmed surgically by Parkinson (1973), who was able to open the lateral wall of the cavernous sinus and to remain outside the intracavernous arterial and venous components while occluding an arteriovenous fistula.

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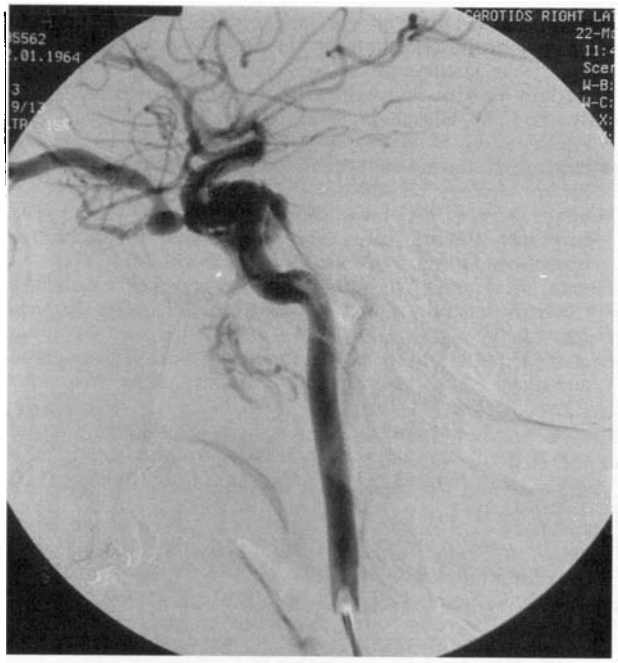

FIG. 1
Carotid angiogram showing carotico-cavernous fistula.

With this later concept, a direct shunt from the intracavernous ICA system theoretically connects with two different structures: the veins of the plexus or the perivascular bare spaces between the veins. The former connection will generate a CCF, while the later will create a false aneurysm. A combination of both anomalies can also be present in the same patient (Isamat, 1987).

Clinical signs which are commonly associated with CCF include proptosis in 94 per cent of cases; pulsating exophthalmos in 40 per cent; bruit in 75 per cent; orbital frontal headache and orbital pain in 40 per cent; chemosis in 71 per cent; extra ocular palsy and diplopia in 60 per cent; loss of visual acuity in 46 per cent, and fifth cranial

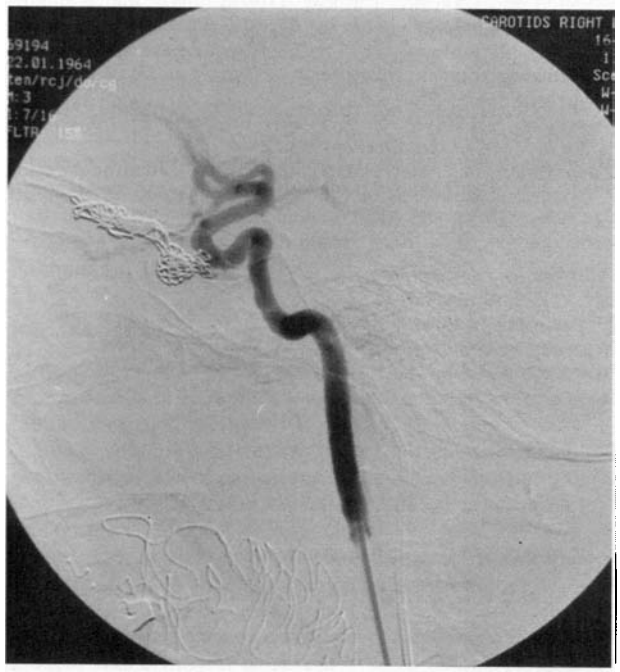

FIG. 2
Closure of the fistula after trans-arterial embolization by Guglielmi electrolytically detachable coils.

nerve involvement in 24.6 per cent of cases (Garland *et al.*, 1977). Massive epistaxis is a life-threatening, although rare manifestation of post-traumatic high flow arterial rents into the cavernous sinus. This has been shown in relation to the existence of false aneurysm (Handa and Handa, 1976). The triad of extensive fronto-basal fracture, unilateral blindness and severe epistaxis has been considered specific of such a condition (Handa and Handa, 1976). Recurrent severe epistaxis, stemming from neovascularity in the nasal mucosa, may lead to anaemia and even in death in CCF (Grossman *et al.*, 1991). Zacharaides and Papavassiliou (1988) observed that it is possible that the frequent nasal bleedings averted the establishment of definite clinical signs of the fistula, as the increase in the pressure was released through the nose. This was true in our case, in which definite symptoms of CCF developed following cessation of epistaxis by cauterization of the anterior ethmoid artery. Venous distension occurs over the face and forehead and very occasionally the nasal mucosa is involved and epistaxis may result (Macfarlane, 1983). In our case the presenting symptom was recurrent severe epistaxis, that occurred prior to the other symptoms. It is difficult to know whether the venous distension over the face and forehead, and the ocular symptoms were precipitated by anterior ethmoid artery cauterization or would have occurred themselves in a matter of time.

The symptoms in the traumatic type of CCF, may develop almost immediately from the time of trauma or may be delayed for a period of several days to a week or two. In this delayed group it is likely that the fracture or transorbital penetrating injury had produced a tear in the wall of the carotid artery with the rupture of the traumatic aneurysm into the cavernous sinus days after injury producing a delayed-type traumatic fistula (Macfarlane, 1983).

The relevant clinical test confirming the diagnosis of a CCF is that the bruit should cease with digital compression of the ipsilateral carotid artery. If pulsating exophthalmos is present, the pulsatile component to exophthalmos will also cease with compression of the involved carotid. Where the fistula is contra-lateral to the involved eye, compression of the contra-lateral carotid should have the same effect. However, the definitive diagnosis is radiological in the form of carotid and occasionally vertebral angiography. Both internal carotid arteries should be injected. A rough idea of the size of the fistula can be gained by the rapidity and volume of venous filling and drainage, and by the degree of reduction of ipsilateral hemisphere blood flow (Macfarlane, 1983).

The history of the treatment of this lesion parallels that of the development of neurosurgery. During the earliest period, Sugar and Meyer (1940) suggested occlusion of the venous outflow. Shortly thereafter, however, ligation of internal and external branches of the carotid artery became standard treatment. Dandy (1935) recognized that carotid ligation allowed the fistula to continue siphoning blood from the intracranial system and advised a trapping procedure by ligation of the internal carotid artery in the neck and intra-cranially above the fistula. Mason *et al.* (1954) further defined treatment by adding ophthalmic artery occlusion to the regimen. Recurrent proptosis and return of the bruit were commonly noted even after the most complete of the procedures, and visual loss was still the primary complication. Hamby (1964) outlined the procedure utilizing intracranial carotid and ophthalmic clipping followed by embolization of the internal carotid artery. The common internal and external carotid arteries were ligated after muscle had been embolized into the fistula. Using cardiopulmonary arrest, Parkinson (1967) was the first to successfully execute a

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direct attack on the fistula. In the final portion of this operation, the cavernous portion of the carotid artery was opened and packed with muscle.

The shortcomings of all these operative approaches is that in each case, the retinal artery pressure and circulation to the eye are further reduced leading to retinal ischaemia and blindness. Debrun *et al.* (1978) developed an ingenious approach utilizing detachable balloons to fill the cavernous sinus and block the fistula. This technique is the first, to allow the carotid and retinal circulation to be spared. The intra-cavernous carotid artery (ICCA) could be preserved in 60–88 per cent of the cases; in the remaining cases it had to be sacrificed to accomplish angiographic cure (Halbach *et al.*, 1991; Debrun *et al.*, 1992). The reasons for this undesired ICCA occlusion were too small a fistulous orifice for the balloon, a venous component too small to allow a balloon inflation, inability to guide the balloon into the involved ICCA segment due to intimal flaps, and repeated balloon puncture by sharp bone fragments. It is sometimes impossible to navigate an additional balloon into a fistula after subtotal occlusion without causing stenosis or occlusion of the parent artery (Halbach *et al.*, 1991; Lewis *et al.*, 1995).

Guglielmi *et al.* (1992) described endovascular treatment by electrothrombosis with detachable coils for CCF. These new coils can be placed and replaced several times until an ideal position is achieved. They are then detached by electrolysis, which further enhances the process of thrombosis at the fistula site.

Conclusion

CCF is an uncommon condition, that can occur after trauma, in young people. CCF after septorhinoplastic surgery is distinguished by its rarity, as ours is only the second reported case in the literature. Recurrent severe epistaxis could be a presenting symptom prior to the appearance of ocular complications, as in our case. Treatment of post-traumatic CCF using electrolytically detachable coils was very effective.

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