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Carotid Cavernous Fistula Associated with Persistent Trigeminal Artery: Endovascular Treatment Using Coil Embolization

ABSTRACT–Carotid-cavernous fistula (CCF) associated with persistent trigeminal artery (PTA) is a rare but important clinical entity. We present a case treated by microcoil embolization with preservation of internal carotid, PTA, and basilar artery flow following embolization. A 62-year-old female developed pulsatile tinnitus followed by left eye proptosis and diplopia. Examination revealed a cranial nerve VI palsy and an objective bruit over the left orbit. Angiographic evaluation revealed a carotid cavernous fistula originating from a persistent trigeminal artery. Placement of a detachable balloon across the fistula site while preserving the PTA proved impossible, and the fistula was treated with microcoils following placement of a microcatheter across the fistula into the cavernous sinus. Complete closure of the fistula was followed by resolution of the patient's symptoms. Preservation of all major vessels including the PTA was accomplished through the use of coil embolization. Careful evaluation of the angiogram is necessary to identify PTA associated with a CCF. Previous reports have described treatment of CCF with PTA by surgical or balloon occlusion, some involving sacrifice of the PTA. Examination of the relevant embryology and anatomy reveals, however, that occlusion of the PTA must be approached with caution due to potential supply to the posterior circulation.

Persistent trigeminal artery (PTA) is the most common of the persistent fetal arterial communications between the carotid and vertebrobasilar systems. The anomaly has been noted to have an incidence of 0.1 to 0.3% on adult angiograms.[1,2] Carotid-cavernous fistulas (CCFs) associated with PTA are uncommon entities. The potentially complex anatomy and hemodynamics associated with PTA requires recognition and understanding of possible complicating factors to treat associated CCFs safely and successfully.

CASE REPORT

A 62-year-old female developed left sided pulsatile tinnitus following strenuous physical activity. She denied head trauma or associated symptoms. Approximately 1 month later, she developed left eye pain, redness, and diplopia. Neurological examination was remarkable for a left VI nerve palsy. An objective bruit was present over the left orbit.

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Angiographic examination of the left internal carotid artery demonstrated a PTA on the left with arteriovenous shunting into the cavernous sinus. The fistula was a single arteriovenous communication located at the junction of the left internal carotid artery and the PTA, which filled primarily from the left internal carotid artery. No aneurysm of the PTA was identified. Although the fistula filled poorly from the vertebral artery injection initially, opacification of the fistula was well demonstrated by left vertebral artery injection during compression of the left carotid artery. No brainstem or cerebellar arteries originated directly from the PTA, and the proximal basilar artery was of normal caliber.

Multiple attempts to place a detachable balloon across the fistula site were unsuccessful. A Tracker 18 microcatheter was successfully placed through the fistula and into the cavernous sinus via the left internal carotid artery. Contrast injection through the microcatheter confirmed placement of the distal tip within the cavernous sinus with retrograde venous drainage via the superior ophthalmic veins, pterygoid plexus, and inferior petrosal sinuses bilaterally. Five 10-mm platinum microcoils were placed into the left cavernous sinus, occluding the fistula. Postembolization angiography demonstrated preservation of the left internal carotid artery, PTA, and basilar artery flow. No residual arteriovenous shunting was present. Filling of both posterior cerebral and superior cerebellar arteries was noted to occur via the PTA (Fig. 1). At 24 hours postembolization, the patient's pain and bruit had resolved. Mild improvement in the CN VI palsy was also noted. At 6 months following embolization, examination demonstrated normal ocular motility with complete resolution of the CN VI palsy. After 3 years no signs or symptoms to suggest recurrence of the fistula have been noted.

DISCUSSION

CCFs associated with PTA are uncommon lesions. They have been described as resulting from trauma and from presumed rupture of PTA aneurysm.[1–6] Potential pitfalls in treatment of patients with these combined lesions may arise from additional clinically significant vascular abnormalities that have been associated with up to 15% of PTA. These include aneurysms involving the PTA itself or located more distally on the internal carotid artery or circle of Willis.[3,7] In addition, retained features of fetal vascular anatomy may have implications for treatment. Review of the relevant embryology of the PTA highlights the need for understanding potential associated anomalies prior to endovascular or neurosurgical treatment.

The PTA represents retention of an embryologic arterial connection between the internal carotid artery and the developing posterior fossa circulation. The PTA arises in conjunction with the development of the trigeminal sensory system at about the 4-mm stage of fetal development. The vessel initially connects the petrous portion of the internal carotid artery with the cranial end of the paired plexiform neural arteries, which run along the ventral aspect of the brainstem. The trigeminal artery supplies blood flow to developing structures of the upper brainstem in a craniofugal direction, whereas the artery of C1 supplies craniopedal flow from below.[8,9]

By the 5- to 6-mm stage the caudal division of the internal carotid artery, which will later form the posterior communicating artery, anastomoses with the longitudinal neural arteries cranial to their junction with the trigeminal artery, and regression of the trigeminal artery begins. Coalesence of the neural arteries in the midline occurs by the 9-mm stage and results in formation of the basilar artery. Development of the vertebral arteries by fusion of the cervical intersegmental anastomoses permits establishment of basilar artery flow in the craniopedal direction seen in the adult. By the 9-mm stage, regression of the PTA is normally complete. Persistence of the vessel has been reported in 0.1 to 0.3% of cerebral angiograms in adults.[1,2,10]

Additional vascular anomalies may accompany a PTA, which reflect its embryologic role as supply to the upper brainstem and which may have significant implications for surgical or endovascular treatment.[11] Ohshiro et al reported pontine branches as well as supply to the trigeminal nerve root in an adult patient with PTA.[13] Cerebellar arteries originating from the internal carotid artery have been described as a manifestation of PTA.[12] Hypoplasia as well as complete interruption of the basilar artery proximal to the PTA have also been reported as a result of failure of the trigeminal artery to regress normally.[10,13]

Retention of fetal trigeminal branches may result in PTA supply to significant amounts of brain normally supplied by the vertebrobasilar circulation in the adult. Compromise of a PTA may therefore cause life-threatening ischemia, depending on the persistence of embryological branches, the anatomy of the posterior communicating arteries, and the presence or absence of other potential sources of collateral flow. Complete evaluation of vascular anatomy is therefore particularly important prior to planning treatment of vascular lesions associated with PTA.

CCF associated with PTA is an uncommon lesion. Most reported cases of CCF associated with PTA have been located at the junction of the PTA and internal carotid artery. Several reported cases have required permanent occlusion of major vessels, including the PTA and internal carotid artery for obliteration of the fistula. More recently, reports in which balloon occlusion the PTA was accomplished with preservation of the internal carotid flow have appeared.[2] Despite the increasing availability of endovascular techniques, technical features associated with the lesion may make treatment dif-

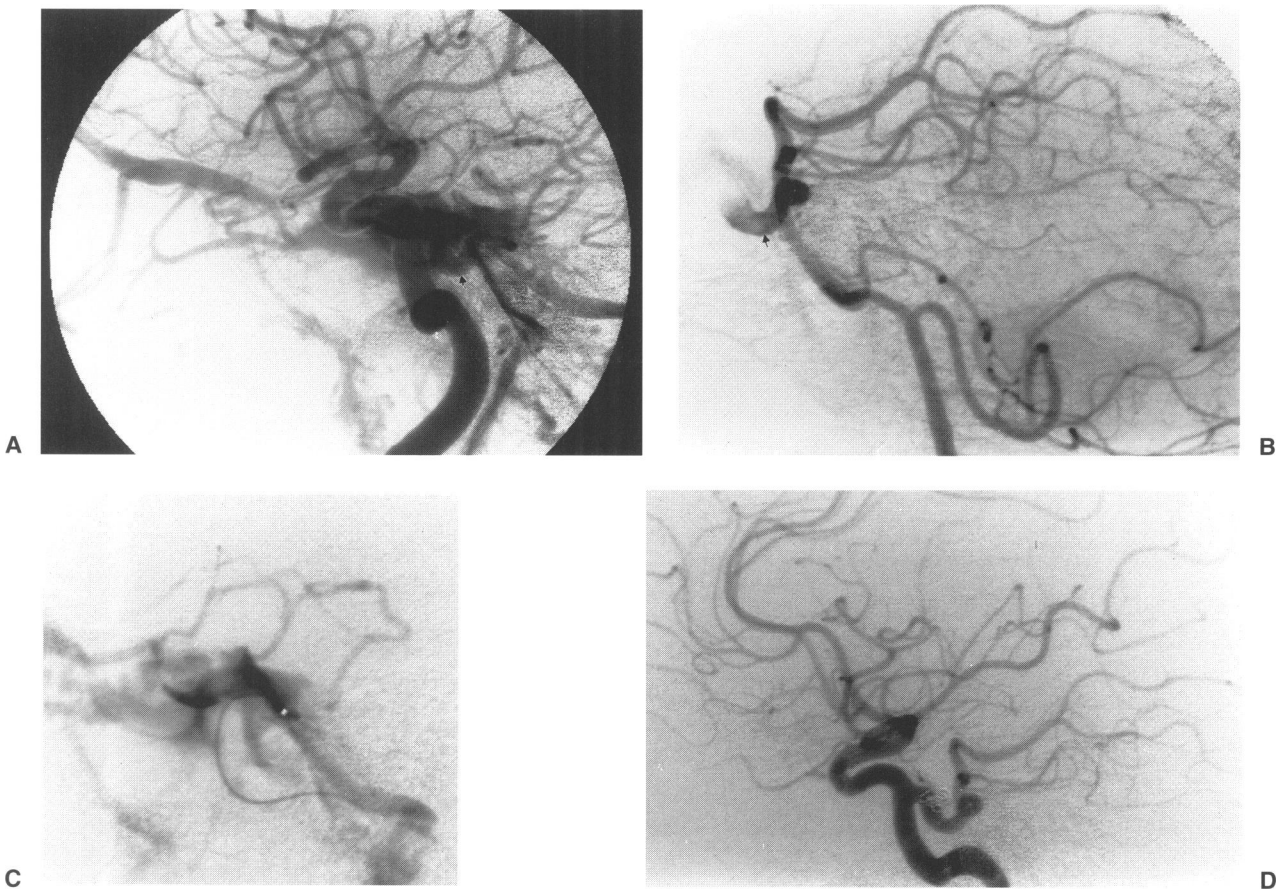

Figure 1. (**A**) Injection of the left internal carotid artery shows filling of the cavernous sinus as well as anterior portion of the persistent trigeminal artery (PTA, **arrow**). (**B**) Vertebral artery injection with left carotid compression demonstrates filling of the PTA (**arrow**) and filling of the fistula (**C**) Microcatheter tip within the cavernous sinus prior to coil placement. (**D**) Postembolization angiogram with no arteriovenous shunting, preservation of the internal carotid artery, and PTA.

ficult. Tortuosity of the internal carotid artery and PTA may make successful placement of a balloon impossible, as was the case in our patient. The use of microcoils system allows more flexibility to navigate tortuous vessels, as demonstrated in this case.

The presence of a PTA in the patient with CCF is a critical observation that should be sought in all cases of CCF. A PTA may provide an additional route for endovascular treatment if approach through the internal carotid artery is not possible. Identification of a PTA is particularly important if trapping of the fistula with occlusion of the carotid artery is being considered as a therapeutic option. If the persistent fetal connection is not identified, therapeutic failure due to filling of the fistula via vertebrobasilar flow is likely. Therefore, rapid sequence filming of both carotid and vertebrobasilar injections is mandatory. Spontaneous flow through the PTA is usually from carotid to basilar reflecting the hemodynamic situation that exists in utero. This was the case in our patient despite the presence of the fistula. The direction of flow may prevent opacification of the fistula on a standard vertebral artery angiographic injection. In such cases, vertebral injection during carotid compression is of benefit in evaluating a possible posterior circulation connection to the fistula.

In conclusion, identification of a PTA in association with a CCF is an important clinical observation with major treatment implications. The potentially complex anatomy of the lesion may create unique difficulties in its management. This case demonstrates coil embolization of the CCF associated with PTA to be an effective therapy that should be considered in cases where anatomic or technical factors prevent selective closure with a detachable balloon.

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