Congenital Facial Arteriovenous Fistula Successfully Treated by Tranarterial Embolization: a case report

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It is difficult to manage arteriovenous malformations or fistulas of the face because of the abundant vascular network in this region. They are commonly manifest in childhood or adolescence and often present with significant hemorrhage or cosmetic defects. We report a one-year-old infant with a facial arteriovenous fistula. It is successfully treated by transarterial embolization with metallic coils, resulting in clinical and angiographical improvement.

Key words: **Arteriovenous malformation, therapeutic embolization; Face**

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Arteriovenous malformations (AVMs) or arteriovenous fistulas (AVFs) of the head and neck are quite rare in contrast to low-flow vascular malformations. They are 20 times more common in the intracranial vasculature than in that served by the external carotid arteries [1]. Cervicofacial involvement is most common in the cheeks, ears, nose, and forehead, in descending order of prevalence. The primary pathology occurs at the capillary level, where arteriovenous shunting occurs. Engorged feeding arteries and drainage veins result from uncontrolled shunting through these channels. They are rarely symptomatic in the neonatal or infant period, but most commonly discovered in late childhood, adolescence, or early adulthood. AVMs / AVFs manifest as warm, firm masses with thrills, bruits, and pulsatility.

Treatment of these high-flow vascular anomalies is complex and has a predictably high incidence of recurrence if not managed correctly. Intervention is indicated for complications such as pain, haemorrhage, pressure symptoms, ischaemic ulceration and even congestive cardiac failure.

CASE REPORT

A 1-year-old infant had suffered from a pulsatile lesion over right preauricular area for several months. There was an erythematous blush of the skin overlying the lesion. Physical examination revealed a remarkablely palpable thrill with continuous murmur at right preauricular area. Digital subtraction angiography with selective right external carotid arteriography revealed arteriovenous fistula between right internal maxillary artery and right external jugular vein (Fig. 1). After the feeding arteries were identified, transarterial embolization with two 5mm spring coils was performed to embolize the fistula (Fig. 2). The thrill in preauricular region disappeared immediately. Post-embolization angiogram showed complete disappearance of the

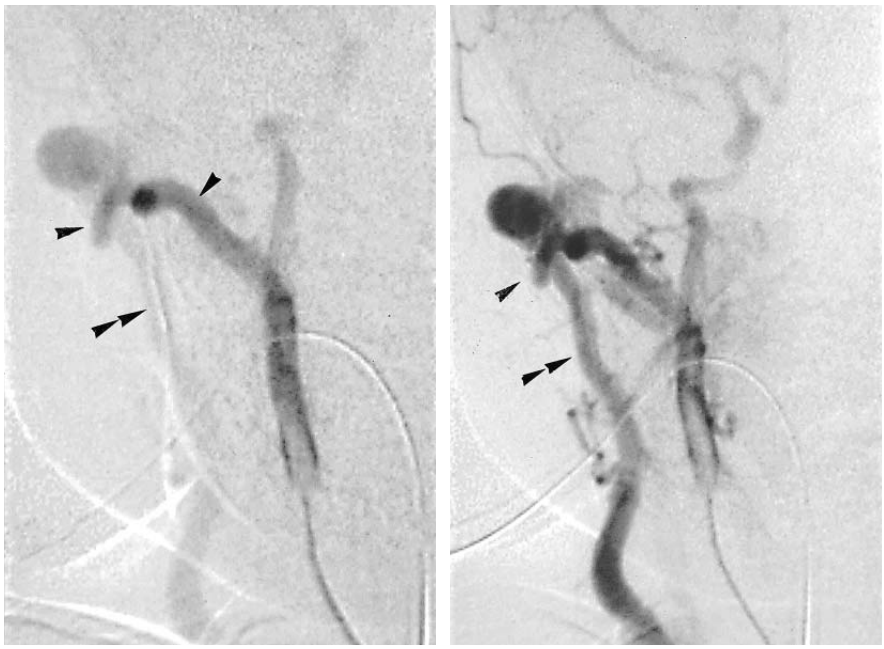

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Figure 1. a. Arterial, and **b.** capillary phase of lateral projection digital subtraction images from right external carotid angiogram show arteriovenous fistula (white arrow) between the internal maxillary artery (black arrow) and external jugular vein (double black arrow).

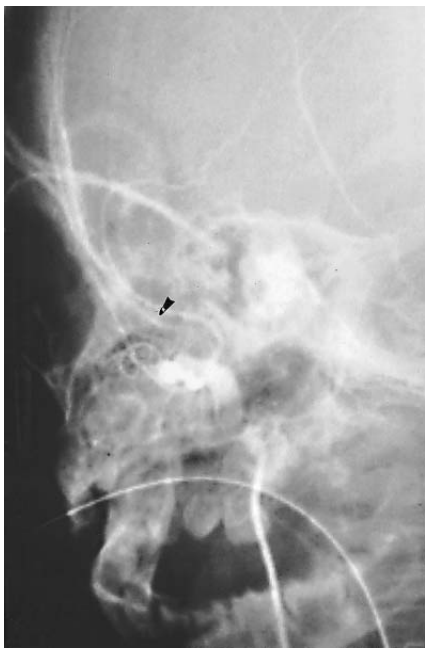

Figure 2. Unsubtracted lateral radiograph obtained after embolization of the feeding artery with two pieces of 5mm spring coils (arrow).

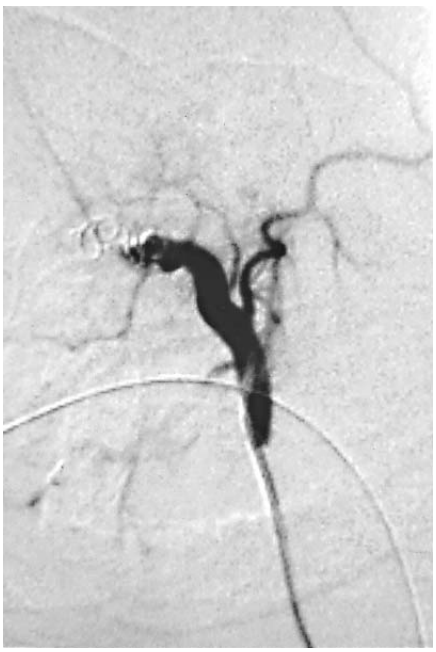

Figure 3. Lateral projection of right external carotid angiogram after embolization shows complete occlusion of the fistula.

fistula (Fig. 3). The patient recovered without complication. No recurrence of the lesion was seen on subsequent clinical examination.

DISCUSSION

AVFs occur most often in the carotid cavernous fistula and vertebral fistula, rarely in external carotid fistula. AVMs / AVFs of the face or neck are high-flow vascular malformations and have a rich arterial network fed by branches of the external carotid artery. There are two types of arteriovenous fistulas, congenital and acquired. A congenital arteriovenous fistula is uncommon and one that formed during fetal develop-

ment. In congenital fistulas, blood vessels of the lower extremity are more frequently involved than other areas of the body. Spontaneous development of AVF occurs in neurofibromatosis 1 and fibromuscular disease, as well as without specific underlying disease [2, 3]. An acquired arteriovenous fistula may cause by iatrogenic reasons or more commonly, traumatic in origin. It is rare of our case, a congenital arteriovenous fistula of the external carotid artery, which is symptomatic in such a little baby.

Our Case is a stage II arteriovenous fistula, which displays bruit and thrill. In early stage, AVM may manifest only cutaneous warmth or blush. When the extensive lesion causes increased shunting of blood and decreased nutritive blood flow to the skin, the symptoms and signs include pain, ulceration, bleeding, audible bruits, palpable thrills, and pulsatility [4]. Factors associated with expansion include trauma, infection, incomplete resection, and hormonal changes occurring during pregnancy, puberty.

Symptomatic or complicated lesions may necessitate an attempt at surgical excision or selective embolization. In the past, treatment of AVM/ AVF of the face was primarily reliant on surgical excision or ligation of the feeding arteries [5, 6, 7]. For small lesions, excision alone may be possible. For more extensive lesions, the treatment of choice is combination therapy that consists of preoperative angiography with selective embolization followed by definitive resection within 24- 48 hours. Excision was often associated with extensive blood loss and the need for skin reconstruction procedures. Surgical ligations of the feeding vessels proximal to the fistula are sometimes ineffective because of the recruitment of a collateral vessel supply and the loss of access to the fistula for further embolization [8].

Embolization as the sole treatment of AVM/ AVF can be either curative or palliative for symptoms such as pain or mass effect [9, 10, 11]. When used embolization immediately prior to resection, it reduces intraoperative blood loss, shortens surgical time, and decreases surgical morbidity and mortality [9]. Embolization may use PVA, balloons, coils or NBCA [12]. Small diameter coils are especially useful when it is impossible to pass a balloon through the narrow orifice of the fistula, or when the venous portion of the fistula is not large enough to inflate the balloon.

Congenital arteriovenous fistula of the internal maxillary artery in a infant is rare. Early recognition and proper treatment are essential for an acceptable long term outcome. The case we present here demonstrates a quick and successful method of closing an AVF by the transvascular embolization with coils. ◆

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成功栓塞治療先天性臉部動靜脈瘻管

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臉部動靜脈畸型或瘻管因為內含大量血管網，導致很難處理。通常在兒童時期或青少年期開始表現明顯，時常伴隨有出血或臉部美觀的問題。我們提出一個病例報告有關一個有先天性臉部動靜脈瘻管的一歲嬰兒，成功用金屬線圈栓塞治療。治療後不論臨床症狀與血管攝影方面都有顯著改善。

關鍵詞：動靜脈瘻管，栓塞；臉部