External carotid artery pseudoaneurysm in an immunocompromised patient

Thomas L. Forbes, MD; Michael Tong, MD

Aneurysms of the extracranial carotid arteries are rare and are generally atherosclerotic in nature. In adults, infected or mycotic aneurysms are unusual and are most often due to systemic sepsis and bacteremia. The commonest infecting organisms are *Staphylococcus, Streptococcus, Klebsiella, Escherichia* and *Salmonella*.[1] In children, tonsillitis and pharyngeal abscesses are more common and can cause vascular complications including thrombophlebitis of the internal jugular vein (Lemierre's syndrome) and extracranial carotid pseudoaneurysms.[2]

We report a soft tissue infection of the neck in an immunocompromised adult secondary to a rare complication of dermatitis herpetiformis treated with diamino-diphenyl sulphone (dapsone).

Case report

A 33-year-old, previously healthy woman presented with an acute dermatologic disorder diagnosed as dermatitis herpetiformis. She was treated effectively with dapsone. Unfortunately, she suffered a rare complication of dapsone therapy, namely, agranulocytosis. Six weeks after the initiation of dapsone she presented with a decreased leukocyte count (0.5×10^9/L) and an upper respiratory tract infection. Even though she was admitted to hospital and treated with filgrastim and antibiotics, the infection progressed, causing upper airway obstruction and necessitating intubation and a subsequent tracheostomy. Subcutaneous abscesses subsequently developed in the anterior neck bilaterally. They were incised and drained. Culture specimens of the abscesses grew *Staphylococcus aureus*. Ten days later, a left submandibular pulsatile neck mass was noted, which enlarged over a couple of days. Contrast-enhanced CT revealed an inflamed area surrounding a 3-cm pseudoaneurysm adjacent to the left external carotid artery (Fig. 1). Three-dimensional reconstruction confirmed that the pseudoaneurysm was situated between the external carotid artery and the hyoid bone and likely originated from the lingual artery (Fig. 2). She was transferred to our centre for further treatment.

The patient's condition was essentially stable, so repair options were considered. An open approach was chosen rather than an interventional one because of the concern of ongoing infection and the need for appropriate drainage and débridement. Under general anesthesia, an incision was made along the anterior border of the left sternocleidomastoid muscle, and the platysma muscle was divided. Tissues deep to the platysma were markedly inflamed and indurated. The internal jugular vein was reflected posterolaterally and the common carotid artery was identified. Dissection continued in a cranial direction until the carotid bifurcation was encountered. Overlying the external carotid artery was significant inflammation surrounding the readily palpable pseudoaneurysm, which made identification of the cranial nerve difficult. The external carotid artery was ligated and the pseudoaneurysm was opened, allowing identification of the feeding lingual artery that was suture ligated from within. Thrombus and surrounding tissues were débrided as much as safely possible. The patient recovered quickly and was transferred to her referring institution on postoperative day 2.

Discussion

Dermatitis herpetiformis is a pruritic bullous dermatologic disorder primarily affecting the extensor aspects of the elbows, knees, back and scalp. It is part of the spectrum of gluten-sensitive disorders that includes celiac disease. Although patients with dermatitis herpetiformis frequently lack gastrointestinal manifestations, it is a direct consequence of an intestinal intolerance to gluten. It affects

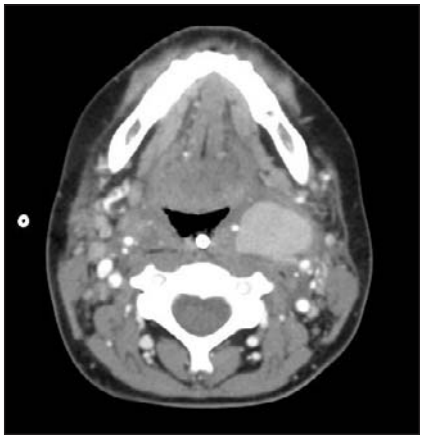

FIG. 1. Axial section from a CT scan showing the contrast-filled pseudoaneurysm just medial to the left external carotid artery.

Division of Vascular Surgery, London Health Sciences Centre, and the University of Western Ontario, London, Ont.

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Correspondence to: *Dr. Thomas L. Forbes, Division of Vascular Surgery, London Health Sciences Centre, 800 Commissioners Rd. E, E2-119, London ON N6A 5W9; fax 519 667-6549; tom.forbes@lhsc.on.ca*

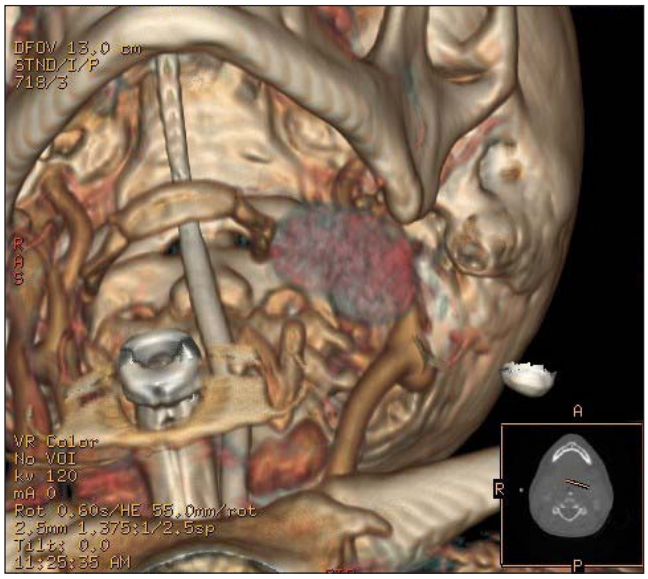

FIG. 2. Three-dimensional reconstruction of the CT scan showing the pseudoaneurysm situated between the left external carotid artery and the hyoid bone.

men more commonly than women and most frequently occurs between the second and fourth decades of life. Dapsone is the medication most commonly used to treat this disorder. Severe agranulocytosis is an idiosyncratic reaction to dapsone that usually occurs 1–3 months after the onset of treatment. The leukocyte count should be monitored during therapy, and dapsone should be discontinued if the leukocyte count falls below $4 \times 10^9/L$.[3]

In our case, the soft tissue neck infection with *S. aureus* caused localized inflammation and arterial destruction resulting in a pseudoaneurysm of the adjacent lingual artery. To prevent hemorrhage and further infectious complications, an open surgical approach was chosen to ligate the external carotid and lingual arteries and débride the surrounding inflamed and infected tissue. It is ill advised to place synthetic grafts in such infected fields. In similar cases, both ligation and reconstruction with autologous conduits have been employed with good results.[4] In this case, we considered that the external carotid artery was expendable. Successful endovascular treatment of a similarly infected pseudoaneurysm of an extracranial carotid artery has been reported,[5] but this procedure has the disadvantage of introducing a foreign material into an infected field, and it does not provide débridement and drainage.

Although infected pseudoaneurysms of the extracranial carotid arteries are rare, our case illustrates that this condition should be considered when neck masses develop in immunocompromised patients, especially in the presence of upper respiratory tract or soft tissue infection of the neck.

Competing interests: None declared.

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