Case Report

True occipital artery aneurysm: A surgical case report and literature review

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A B S T R A C T

True occipital artery aneurysms are exceptionally rare. To our knowledge only five reports of true occipital artery aneurysm have been previously described.1–5 We present a rare case of a 70-year-old gentleman with a true occipital artery aneurysm associated with alopecia areata. This case report adds to the current lack of literature on true occipital artery aneurysms and summarises their presentation, investigation and management.

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Introduction

A true aneurysm is defined as an abnormal, localised and permanent dilatation, of all three layers of an arterial wall. Dilatation is typically greater than 150% of its original size. True occipital artery aneurysms are exceptionally rare. To our knowledge only five cases have been previously reported. Occipital artery pseudoaneurysms are slightly more common with 13 cases that were previously described. Pseudoaneurysms have been strongly associated with trauma, other causes include congenital occipital bone malformation, autoimmune disease and post-operative complications from direct iatrogenic trauma.

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Figure 1. Ultrasonographic imaging of the occipital aneurysm demonstrating normal calibre artery on the left and dilated aneurysmal arterial wall to the right.

The presentation of occipital artery aneurysms is variable and will depend upon their location, size and the presence of local compression of neurovascular structures. In this article we describe the presentation, investigation and surgical management of a patient with a true occipital artery aneurysm.

Case report

A 70-year-old man was referred with a three-month history of an enlarging, non-tender, subcutaneous swelling overlying the right occipital protuberance. A larger well-defined area of alopecia areata encircled the swelling and predated it by two years.

His past medical history included a tissue mitral valve repair, hypercholesterolaemia, hypertension and atrial fibrillation. His regular medication included Edoxaban. There was no previous history of trauma, autoimmune disease or infection. He was a retired hotelier and an ex-smoker with a 40-pack-year history. There was no family history of aneurysms.

On examination a pulsatile 2 × 2 cm mobile subcutaneous mass was revealed. Surrounding the mass was a 15 × 12 cm focal area of hair loss in keeping with alopecia areata. There were no other associated skin changes or visible punctum.

Initially, the mass was considered to be a sebaceous cyst or lipoma however the pulsatile nature of the mass prompted an ultrasound scan. This demonstrated an aneurysmal dilatation of the right occipital artery measuring 11 mm × 20 mm, as shown in Figure 1.

Autoimmune serology consisting of a lupus screen, anti-cardiolipin anti-bodies, anti-neutrophil cytoplasmic antibodies and erythrocyte sedimentation rate were normal. Large vessel imaging of the aorta and thoracic vasculature was also normal.

Following discussion with the patient a decision was made to proceed with the excision of the aneurysm. Edoxaban was stopped 48 h pre-operatively. The patient underwent a general anaesthetic in the left lateral decubitus position. A curvilinear incision was made (Figure 2).

Surrounding central and right-sided occipital hair loss is illustrated. The aneurysm was meticulously dissected (Figure 3), and the normal calibre occipital artery was identified and ligated (Figure 4) proximally and distally.

The aneurysm was then excised and sent for histopathological analysis (Figure 5).
Figure 2. Pre-operative skin markings demonstrating the planned incision and the aneurysm itself within the dotted lines. Surrounding central and right sided occipital hair loss is illustrated.

Figure 3. Skin and subcutaneous tissue retracted revealing the right occipital artery aneurysm. A tortuous distal segment of the occipital artery can be seen to the left of the aneurysm.

The wound was closed with 3–0 Monocryl and 4–0 Prolene sutures. He remained as an inpatient overnight before being discharged on the following day. His post-operative recovery was complicated by a local wound infection that was successfully treated with oral antibiotics.

Histological analysis reported a medium-sized 50 × 20 mm muscular artery with segmental dilatation and true aneurysmal formation. Organised thrombus was seen within the lumen of the dilated segment. The arterial wall showed prominent intramural and perivascular granulomatous inflammation with many giant cells, fibrointimal thickening, and destruction of the tunica media muscle and elastic fibres.
Figure 4. Normal calibre occipital artery proximal to the aneurysm.

Figure 5. The excised occipital artery specimen.

Discussion

Literature review

Five cases of true occipital artery aneurysms have been previously reported, as illustrated in Table 1. Three of these were idiopathic, one secondary to blunt trauma from a basketball and one case of a ruptured true occipital artery aneurysm secondary to neurofibromatosis type 1 (NF1). All previously reported cases of non-ruptured true occipital artery aneurysm have been successfully managed by surgical excision.

The first case of an idiopathic true occipital artery aneurysm was described in 2010. A 51-year-old man presented with a one-month history of a painless pulsatile mass overlying the right occiput. It was investigated by three-dimensional computerised tomography (CT) imaging and excised surgically.\textsuperscript{1}

In 2012, Rao et al. described a true occipital artery aneurysm that was presented four months following a traumatic blow to the occiput from a basketball in a 14-year-old male patient. Although the initial strike was painful, the aneurysm itself was non-tender and asymptomatic. The aneurysm was
mobile, non-pulsatile and had remained in its original size since impact. The mass was investigated with x-ray and magnetic resonance imaging (MRI). The aneurysm was later surgically excised due to concerns related to its cosmetic appearance.\(^2\)

In 2017, Chaudhry et al. described the third case of a true occipital artery aneurysm. An 83-year-old female presented with an eight-week history of headache, transient gait disturbance and right-sided pulsatile tinnitus. Physical examination revealed a tender pulsatile mass over the right occipital protuberance. This aneurysm was investigated with MRI and CT angiography (CTA) and then surgically excised.\(^3\)

The following year, Bissacco et al. described a case of multiple true occipital artery aneurysms secondary to NF1. A 53-year-old woman presented in an emergency setting following the rupture of the right occipital artery aneurysm. The patient developed a right laterocervical haematoma causing left tracheal deviation. This case was successfully managed with endovascular embolisation of the occipital artery and open surgical evacuation of haematoma. Complete occlusion of the right occipital artery was later confirmed by MRI.\(^4\) Illuminati et al. described the most recent case of true occipital artery aneurysm. An 83-year-old female patient presented with pulsating of her earring and a mild right sided tongue deviation. The pulsatile mass was situated over her right mastoid region. The patient underwent CTA, which confirmed a right sided occipital artery aneurysm. This was later managed with surgical resection.\(^5\)

Investigations including ultrasonography, CTA and MRI are useful modalities that can aid diagnosis and surgical planning of occipital aneurysms. Options for managing occipital artery aneurysms include percutaneous ultrasound-guided thrombosis and coil occlusion. However, the current adopted standard approach to managing true occipital artery aneurysms is through surgical excision.\(^3\)–\(^7\)

We present the first case of a true occipital artery aneurysm associated with preceding localised alopecia areata. We postulate this may be a pressure effect leading to local tissue hypoxia and hair loss.\(^8\) Pressure alopecia has been previously described following endovascular procedures and arterial embolisation.\(^9\)

The variability of presenting symptoms can make clinical diagnosis difficult, particularly when more common scalp lesions such as epidermoid cysts or lipomas are suspected. Careful clinical examination by assessing whether a subcutaneous mass is pulsatile can help to avoid misdiagnosis and mismanagement.\(^6\)

### Table 1
Summary table of all previously reported true occipital artery aneurysms.

| Case | Age (Years) | Gender (M/F) | Side (R/L) | Aetiology | Investigation | Management |
|------|-------------|--------------|------------|-----------|---------------|------------|
| 1. Kim et al. [1] | 51 | M | R | Idiopathic | CTA | Surgical excision |
| 2. Rao et al. [2] | 14 | M | L | Trauma | XR + MRI | Surgical excision |
| 3. Chaudhry et al. [3] | 83 | F | R | Idiopathic | CTA + MRI | Surgical excision |
| 4. Bissacco et al. [4] | 53 | F | R | NF1 | CTA | Embolisation |
| 5. Illuminati et al. [5] | 83 | F | R | Idiopathic | CTA | Surgical excision |

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### Ethical approval

Not required.

### Declaration of Competing Interest

None.
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