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An unique tumour of the geniohyoid muscle: an intramuscular haemangioma

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Abstract
We present the first case report in the English literature of an intramuscular haemangioma of the geniohyoid muscle. This occurred in a 24-year-old female and the diagnosis was not made prior to resection. Haemangiomas are uncommon tumours of the head and neck and intramuscular haemangiomas account for fewer than one per cent of the total. Diagnosis of the vascular nature of the tumour is often missed. Recurrence is common and usually due to incomplete excision. A review of the literature and a case report of these locally destructive lesions is presented.

Key words: Haemangioma; Muscle

Introduction

Haemangiomas are uncommon tumours of the head and neck and those arising within skeletal muscle, intramuscular haemangiomas, account for fewer than one per cent of the total (Watson and McCarthy, 1940). The latter is most frequently described in the upper or lower limbs with the head and neck being the third most common site (MacDermott, 1935; Scott, 1957; Welsh and Hengerer, 1980). When such lesions do occur in the neck then the masseter muscle, followed by the trapezius, are those that are typically involved (Wolf *et al.*, 1985).

We describe the first case of an intramuscular haemangioma arising in the left geniohyoid muscle. This lesion was not diagnosed prior to surgical resection and removal was difficult due to a widely dispersed collateral blood supply. Intra-operative blood loss was at times considerable and complete detachment of the mass from the surrounding muscles was facilitated by a new method of unipolar diathermy dissection. The origin, presentation and natural history of these unusual lesions is reviewed.

Case report

A 24-year-old woman presented with a four-year history of a submental mass which had been gradually increasing in size. By the time of presentation the lesion had produced a considerable dewlap with cosmetic embarrassment. There had been no pain and no symptoms to suggest thyroid abnormality or primary pathology in the upper aerodigestive tract.

On examination the patient was euthyroid but there was a 4 by 3.5 cm mass in the submental region of the upper neck. This was discrete and non-tender. It appeared to be slightly fluctuant but was not transilluminable. The lesion was not pulsatile and there were no bruits audible. The overlying skin was not discoloured and was mobile over the mass. There was no intra-oral extension and the upper aerodigestive tract was unremarkable on examination.

Thyroid function tests were within the normal range and a fine needle aspiration biopsy showed only blood cells and no epithelium. A thyroid scan showed some generalized enlargement but the gland was homogenous with no evidence of ectopic tissue uptake. An ultrasound scan

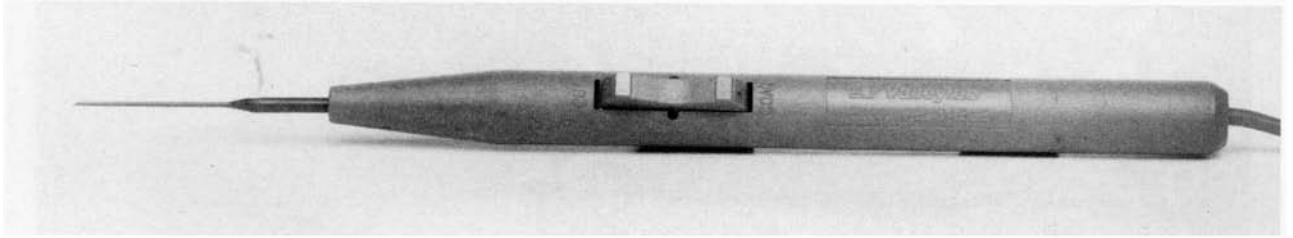

FIG. 1
Colorado microdissection needle.

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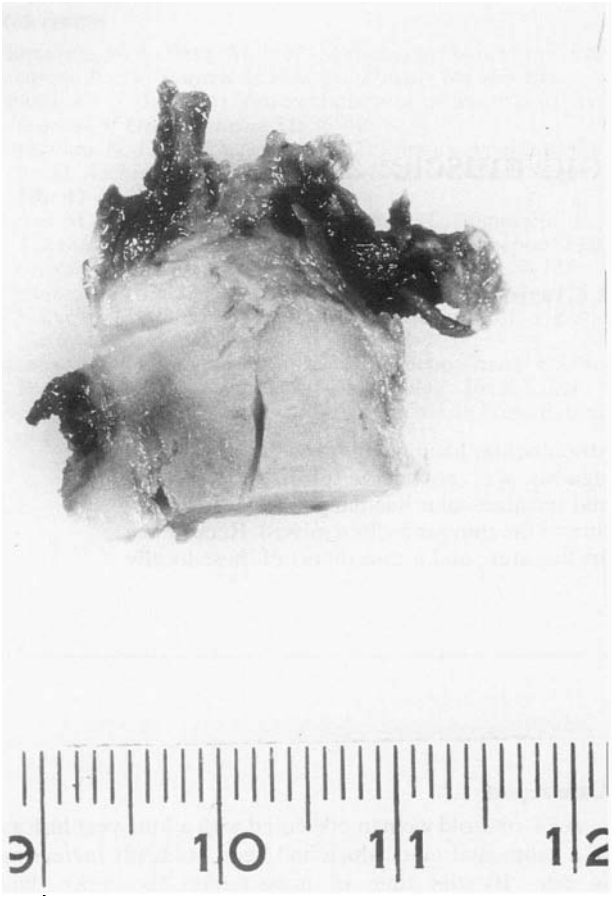

FIG. 2
Gross surgical specimen.

showed echogenicity which was similar to the thyroid with a generally homogenous pattern and with a slightly denser area to the left of the midline.

When the skin and platysma were opened it was readily apparent that the lesion was pulsatile. The overlying geniohyoid was thinned and was clearly an integral part of the mass but, while other margins were not easily identified, there appeared to be no other muscles ultimately involved. Resection was undertaken with loupe magnification (× 2) and using a small tungsten alloy dissecting needle with unipolar diathermy (Colorado Biomedical Inc.) (Figure 1). The resected specimen measured 3.5 by 2.5 by 1.6 cm (Figure 2) and histology showed it to be an intramuscular haemangioma of the mixed capillary and cavernous type (Figure 3).

Discussion

Intramuscular haemangiomas mostly occur before the age of 30 years. One large scale survey found that 21.6 per cent occurred before the first decade of life, 56 per cent arose before the age of 20 years and 85 per cent were apparent by the age of 30 years (Shallow *et al.*, 1944) and these findings have been mirrored in more recent studies (Welsh and Hengerer, 1980). As a result a congenital origin has been presumed. However, considerable confusion exists concerning the histogenesis of haemangiomas in general. Some experts consider haemangiomas as benign true neoplasms, congenital in nature, while others consider them as hamartomatous in origin (Ryan and Cherry, 1987). Histologically the lesions are either capillary, cavernous or mixed (Allen and Enzinger, 1972). The sex incidence is equal (Shallow *et al.*, 1944). Spontaneous regression does not occur and tissue remnants left following surgical resection may result in recurrence (Beham and Fletcher, 1991; Rossiter *et al.*, 1993).

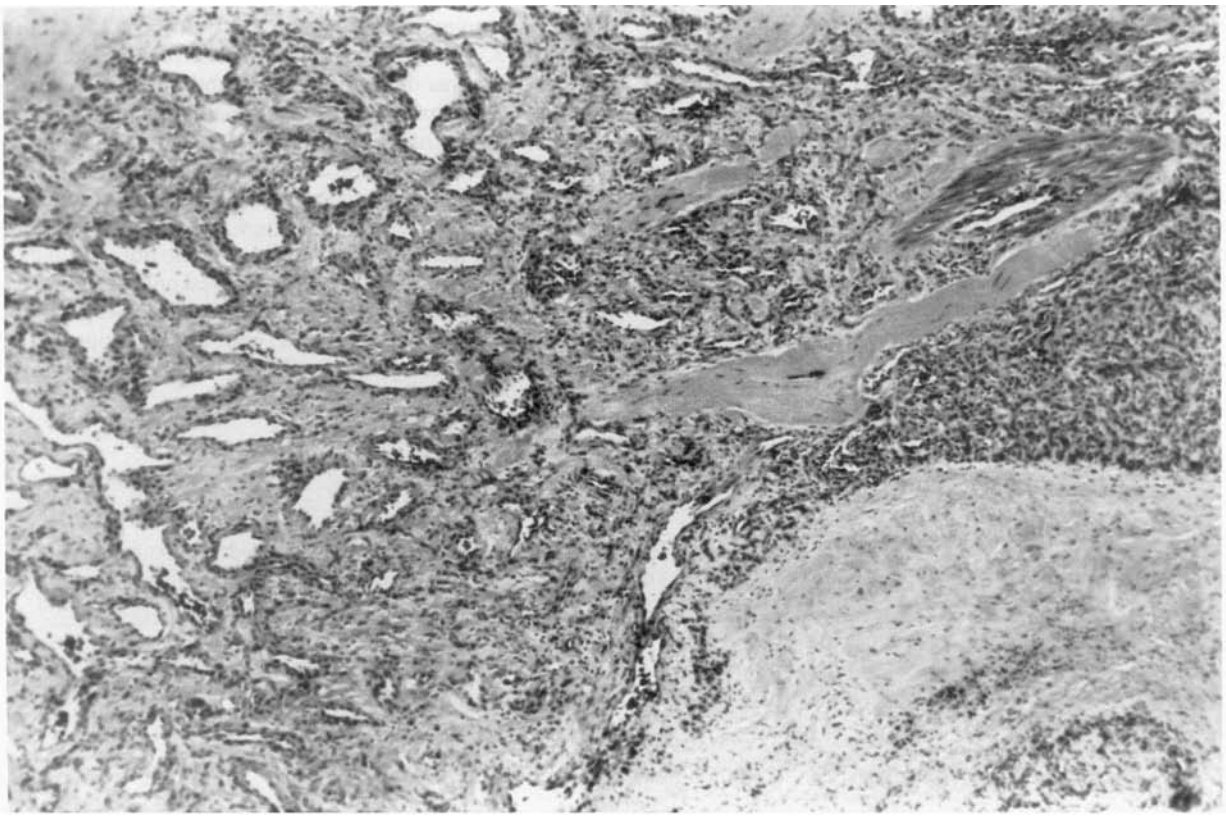

FIG. 3
Microscopic appearance of haemangioma.

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An intramuscular haemangioma in the geniohyoid has not been previously described and was not suspected in this instance on clinical grounds. However, in most large series accurate pre-operative diagnosis is reported in fewer than eight per cent of cases due to the rarity of the lesions and a lack of accurate diagnostic criteria (Allen and Enzinger, 1972). As was confirmed in this case fine needle aspiration is widely held to be unhelpful and bruits, thrills and pulsations are infrequently found due to the surrounding muscle bulk which conceals the vascular nature of the lesion. Skin discoloration is also rare – although it is reported that a cavernous lesion may occasionally produce a reddish-blue discoloration (Rossiter *et al.*, 1993): no skin changes were observed in this patient. An MRI scan, which shows good tissue delineation, is probably the radiological investigation of choice (Buetow *et al.*, 1990).

It is generally agreed that management should be surgical resection. Embolization has been used in the face of torrential haemorrhage and, if the lesion is diagnosed pre-operatively, can facilitate excision (Cohen *et al.*, 1983). Sclerosants cause post-injection scarring and recurrence is common after their use. While radiotherapy has been used alone, and following incomplete excision, its use cannot be recommended in young patients – especially since malignant transformation has been reported (Stofman *et al.*, 1989). The results of radiotherapy are, in any case, said to be disappointing.

Surgical resection in this case was undoubtedly facilitated by the use of diathermy excision with a microdissection tungsten needle (Colorado Biomedical Inc.). As with a laser fibre, which we believe would have produced a similar operative field, this needle produces high heat density at the cutting tip and has been shown in other clinical situations to produce minimal collateral damage while reducing intra-operative blood loss (Monro, 1995). Using this technique a meticulous and complete surgical resection was achieved with minimal blood loss and almost no post-operative oedema. While bleeding during surgery was often considerable for short periods, the overall blood loss during the procedure was minimal and no transfusion was required. Of greater importance is that complete excision was also achieved. We believe that such an outcome would almost certainly have been difficult or impossible to achieve with conventional surgical techniques.

In conclusion, we present an intramuscular haemangioma of the geniohyoid which is a site of origin which has not been previously reported for these tumours. The mass was widely and, apparently, completely excised. This is the treatment of choice for these rare but locally destructive tumours which appear to be of congenital origin. Reliable pre-operative diagnosis will often prove difficult and it seems likely that when these lesions are discovered accurate resection will be facilitated, and the risk of recurrence lessened, by the use of modern excisional techniques.

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