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CLINICAL RECORD

Pyriform sinus haemangioma: an unusual presentation of an unusual condition

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Abstract

Objective: We present a rare case of an unusual presentation of a pyriform sinus haemangioma in a normally healthy, Caucasian woman, including our management and a review of the literature.

Case report: A 44-year-old woman presented complaining of bringing up fresh blood whilst brushing her teeth, dysphonia, food sticking in her throat, and epigastric pain for the preceding 12 months, accompanied by a 17.5 kg weight loss. She underwent pre-operative computed tomography and subsequent complete excision of a pyriform sinus haemangioma using CO_2 laser.

Discussion: Haemangiomas are congenital vascular malformations and can affect any part of the body. They are not prevalent in adults and are rarely found in the pyriform sinus. Their common presenting symptoms and management have been previously documented; however, the presented case is unusual in both its presentation and management.

Key words: Haemangioma; Pyriform Sinus; Dysphonia; Laser Therapy

Introduction

Haemangiomas are congenital vascular malformations and can affect any part of the body. They constitute the most common tumour of the head and neck in infancy,[1] but are less prevalent in adults. Although the mucosa of the upper aerodigestive tract is a common and recognised subsite for haemangiomas, there are very few published cases of haemangiomas occurring in the hypopharynx, and fewer still in the pyriform sinus. Ferguson[2] and Kleinsasser[3] have reported a difference in the symptoms experienced by adults compared with infants. In adults with supraglottic lesions, symptoms more commonly include hoarseness, dyspnoea, haemoptysis and dysphagia.[4]

Case report

A 44-year-old woman was urgently referred to the otolaryngology department. Her main complaint was that of bringing up fresh blood whilst brushing her teeth, for the preceding 12 months. This was accompanied by intermittent dysphonia, food sticking in her throat and epigastric pain. Over the same time period, she had lost 17.5 kg in weight. Significant past medical history included excision of a thyroglossal cyst via a Sistrunk's procedure, 26 months ago. She had stopped smoking 15 months prior to presentation.

The patient had initially been referred to the gastroenterologists, who had performed an oesophagogastroduodenoscopy which had identified a 'lesion', photographed at the level of the larynx, which had eventually resulted in referral to ENT.

On direct inspection with a flexible nasendoscope in the clinic, a vascular lesion was seen in the right pyriform fossa. The remaining ENT examination was unremarkable.

A computed tomography (CT) scan confirmed the presence of a soft tissue mass in the right aryepiglottic fold, effacing the right pyriform fossa. The nature of this lesion was not clear.

Thus, direct inspection under general anaesthetic, with tissue sampling, was planned.

Surgical procedure

The patient was counselled pre-operatively that, if there was excessive bleeding, only a biopsy of the lesion may be possible. Consent was also taken for a tracheostomy. However, we planned an en masse resection if at all possible, as we believed that incising the tumour would increase bleeding if it was indeed a haemangioma. Thus, the patient was prepared for two potential operations, but we aimed to perform one definitive procedure.

Microlaryngoscopy under general anaesthesia was performed using a Storz Weerda® distending diverticularscope (Storz, Tuttlingen, Germany), which allowed assessment of the lesion including its margins. A discrete, purple, vascular lesion was seen to involve the right aryepiglottic fold and the medial wall of the right pyriform fossa (Figure 1). Excision biopsy was performed using the CO_2 laser AcuBlade™ (Lumenis, registered in Dreieich-Dreieichenhain, Germany) on a 10 W setting. The lesion was removed en masse. The specimen (Figure 2) was sent for urgent histological diagnosis. Bleeding was minimal, making suction diathermy redundant; haemostasis was achieved with patties soaked in 1:1000 adrenaline, following complete excision (Figure 3).

The patient was discharged the following day with simple analgesia.

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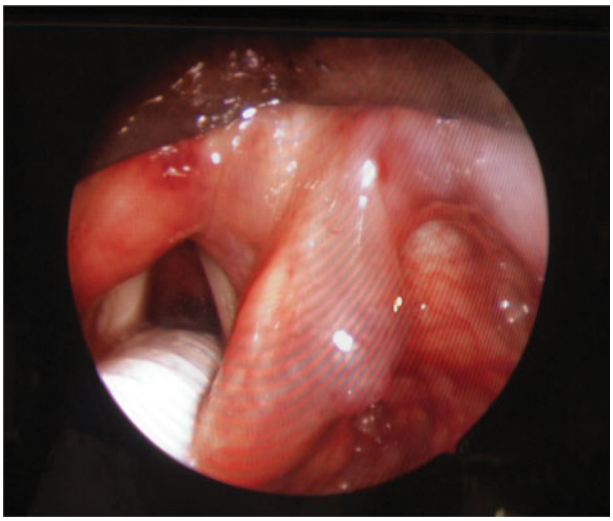

FIG. 1
Pre-operative endoscopic view of the haemangioma involving the right aryepiglottic fold and pyriform fossa.

Microscopically, the specimen showed dilated venous channels with muscularised walls, consistent with a cavernous haemangioma.

At the patient's first clinic review, two weeks post-operatively, she unfortunately complained of site-specific pain, dysphagia and intermittent dysphonia with haemoptysis. On examination, there was evidence of a healing pyriform fossa with no fresh bleeding.

Speech and language therapy assessment indicated a safe and normal swallow, with demonstrable impedance. However, the patient complained of some discomfort that was eliminated by turning her head to the right. Although she complained of her voice being subjectively deeper in pitch, it was in fact deemed to be of a high pitch.

At the second out-patient assessment, 10 weeks post-operatively, the patient reported resolution of her initial pain and no further haemoptysis or dysphonia. However, she described a sensation of 'something in her throat'. Despite this, she reported eating a normal diet.

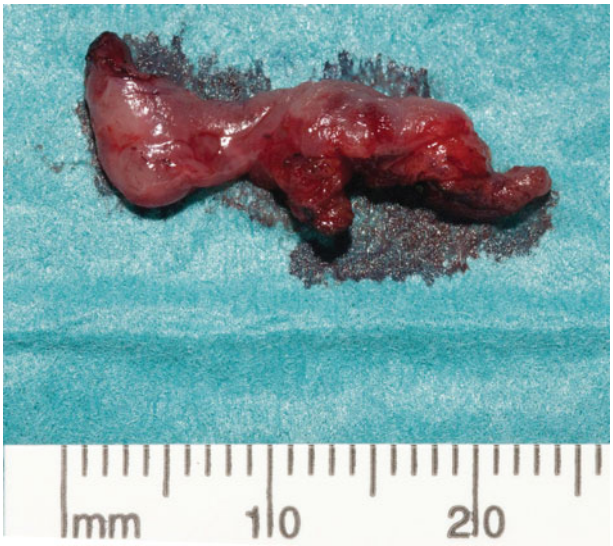

FIG. 2
The excised haemangioma.

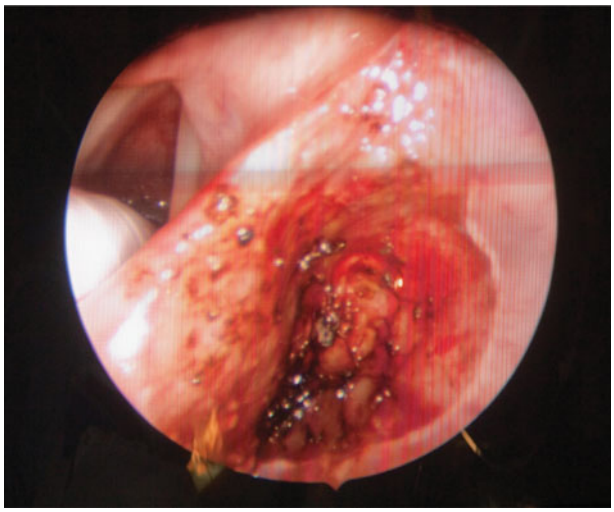

FIG. 3
Endoscopic view of the surgical bed following complete excision of the haemangioma and achievement of haemostasis.

Examination with a flexible nasendoscope revealed a healed pyriform fossa, confirmed by direct inspection under general anaesthesia. Subsequent video fluoroscopy was normal, and the patient was reassured.

Discussion

On review of the literature, we identified 15 cases of haemangiomas involving the hypopharynx. Of these cases, only six were confined to the hypopharynx,[5–10] and fewer still occurred in the pyriform fossa.[9,10]

Any lesion in the pyriform fossa must be treated with a high index of clinical suspicion, as the vast majority are malignant. Considering this fact, and our patient's complex array of symptoms, it was essential in her case to make a tissue diagnosis as a matter of urgency.

Haemangiomas are benign lesions but can cause symptoms ranging from annoying to life-threatening, depending on their location. Our patient's case was particularly unusual in its combination of presenting symptoms, which resulted in an inappropriate initial referral to the gastroenterologists. Although haemoptysis is a recognised symptom associated with haemangiomas, this particular patient described bringing up fresh blood whilst brushing her teeth, when a periodontal cause had been eliminated. This occurred in association with epigastric pain, along with other complaints; this combination has not previously been described.

There are currently no clear guidelines on how to treat hypopharyngeal haemangiomas, largely due to the rarity of the condition. The current literature describes mainly paediatric and subglottic cases, for which different treatment modalities have been documented, including Nd-YAG laser,[11] CO_2 laser,[12] interferon α-2a[13] and radiotherapeutic management.[14] There has been a decline in the use of some of these treatment options due to lack of efficacy or potential side effects.[15]

With respect to adult and hypopharyngeal haemangiomas, the literature is even more limited. The use of cryosurgery to treat a cavernous hypopharyngeal haemangioma was described in 1976.[16] In 1996, Yellin *et al.*[6] described the use of Nd-YAG laser in the treatment of laryngeal and

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hypopharyngeal haemangiomas, as a new technique.[6] In 2006, Hazarika *et al.*[10] used the potassium-titanyl-phosphate laser to excise a pyriform fossa haemangioma; this laser has great haemostatic properties.

- **Haemangiomas are congenital vascular malformations which constitute the most common tumour of the head and neck in infancy, but are less prevalent in adults**
- **Very few cases of haemangiomas in the hypopharynx have been described, and fewer still in the pyriform sinus**
- **A case is presented of a pyriform sinus haemangioma that resulted in initial inappropriate referral and investigation**
- **Use of a CO_2 laser enabled complete surgical excision of this non-pedunculated pyriform sinus haemangioma**

We opted to use a CO_2 laser in our case, due to diagnostic uncertainty regarding the lesion, as there was concern it could be malignant. The senior author ZGGM had extensive experience in oncological resection of laryngeal and hypopharyngeal cancers, as advocated by Steiner and Ambrosch.[17]

In our patient, the use of CO_2 laser enabled safe, complete and effective excision of a pyriform fossa haemangioma. This been described only once previously; that report stated that a pedunculated lesion was a prerequisite when using this modality.[9] However, experience in oncological resections in the hypopharynx proves that non-pedunculated pyriform fossa lesions can also be excised safely with the CO_2 laser.

Conclusion

Pyriform fossa haemangiomas are rare lesions which pose an exigent diagnostic and management challenge to the ENT surgeon. We report an atypical presentation of such a lesion.

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Miss K Hussain takes responsibility for the integrity of the content of the paper
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