CASE REPORT

Angioleiomyoma of the larynx: beware the subglottic lesion

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Accepted 28 November 2015

SUMMARY

Angioleiomyomas are a benign group of tumours rarely documented in the head and neck region. We present a case of a 47-year-old man attending the ear, nose and throat outpatient clinic, with a history of hoarseness over many years. Investigation revealed a left-sided subglottic lesion. He underwent microlaryngoscopy and biopsy, and the lesion was sent for histological analysis.

Postoperatively, the patient began to expectorate fresh red blood that continued for several hours, requiring surgical intervention. Haemostatic control was achieved and a tracheostomy was sited for airway protection. Histopathology identified angioleiomyoma as the cause of his symptoms. Following multidisciplinary team discussion, this was managed with injection of bleomycin and subsequent surgical excision. This case is a reminder that subglottic lesions are often vascular in nature, and should be investigated with imaging and angiography prior to surgery, to avoid complications.

BACKGROUND

Angioleiomyomas are a benign group of tumours arising from the smooth muscle of vascular tissue. Although relatively rare, they are more frequently found within the uterus, extremities and gastrointestinal tract. Within the specialty of ear, nose and throat (ENT) they are particularly rare with few reports available in the literature. Given the relative number of microlaryngoscopy and biopsy procedures undertaken within ENT, it is important to remember that unexpected pathologies can and do occur with risk of complication.

CASE PRESENTATION

A 47-year-old man presented to the ENT outpatient clinic, with constant hoarseness for many years. In the weeks leading up to consultation, his voice quality had deteriorated further and he also reported of new odynophagia. Medical history was unremarkable. He was a non-smoker and there was no family history of head and neck tumours. A full ENT examination was carried out. Flexible nasolaryngoscopy revealed a left-sided subglottic lesion.

INVESTIGATIONS

Microlaryngoscopy was the first-line investigation. This again revealed a left-sided subglottic lesion, which was smooth and red in appearance. The lesion was felt to be benign and biopsy was taken for histopathology, with only minor bleeding perioperatively.

Figure 1 Coronal view MRI showing projection of tumour below the left vocal cord.

DIFFERENTIAL DIAGNOSIS

- Benign lesions—haemangioma, papilloma and granulation
- Malignancy—squamous cell carcinoma.

TREATMENT

Postoperatively, the patient began to expectorate fresh blood, which failed to settle with conservative management. He therefore returned to theatre for arrest of haemorrhage with diathermy to the biopsy site. He remained intubated postprocedure, for airway protection and was transferred to intensive care unit for overnight stay. Owing to intermittent bleeding, the decision was taken to site a tracheostomy and wean off sedation, which were achieved without complication.

CT and MRI of the head and neck were performed to identify the nature, vascularity and

Figure 2 Sagittal view MRI showing extent of tumour below the left vocal cord.

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To cite: Hope N, Smith CP, McCluney N. *BMJ Case Rep* Published online: [please include Day Month Year] doi:10.1136/bcr-2015-213469

Figure 3 Cerebral angiography showing tumour blush.

extent of the lesion. This revealed a $15 \times 15 \times 16$ mm lesion extending into the lumen of the trachea (figures 1 and 2). Following this, a dedicated cerebral angiogram was performed, which identified tumour blush while dye was injected into the left superior thyroid artery and superior laryngeal artery (figure 3). Histopathology confirmed angioleiomyoma (figure 4).

Following discussion at the head and neck multidisciplinary team, it was felt that the lesion would not be amenable to radiological embolisation as it may compromise the blood supply to the superior laryngeal vasa nervosa. Instead, the patient was

Figure 4 H&E stain of angioleiomyoma.

managed with sclerotherapy treatment to the area, with bleomycin injection and subsequent successful surgical excision.

OUTCOME AND FOLLOW-UP

Following discharge, the patient made an uneventful recovery. He remains under follow-up.

DISCUSSION

Following an extensive Medline search of angioleiomyoma and angioleiomyoma, with respect to the larynx, there were few case reports described within the literature. Of particular interest is the paucity of case reports discussing subglottic angioleiomyoma in the English language.

Xu *et al*¹ reviewed the literature in 2008 and identified 15 cases within the Chinese literature, and a further nine in the English language. Three individual cases over a 27-year period were reported in this publication, each with different presenting features. One was of a 53-year-old man with hoarseness, who, on examination, had a 2×3 mm polypoid mass of the left vocal cord, requiring laser excision. The second case was of a 47-year-old man who had a 6-year history of foreign body sensation within his throat and over the 2 months prior to admission had increasing dysphagia. This patient had a submucosal lesion of the aryepiglottic fold and required preoperative tracheostomy and lateral neck incision for removal of the mass. The final patient in the report was a 62-year-old man who presented with a globus sensation. On examination, he had a lesion on the epiglottis. Biopsy of this lesion was complicated by significant blood loss and he required emergency tracheostomy and local haemostasis, followed by transfer to a major head and neck unit. This patient's postoperative course was complicated by a spontaneous bleed on the ward 3 days postoperatively, where he lost 500 mL, requiring emergency lateral neck incision and tumour excision. The final patient in this case report highlights that although the initial complication of bleeding had been dealt with, it remained a significant risk until formal tumour excision had taken place.

In 1976, Shibata and Komune² reported a case of subglottic angioleiomyoma in a 52-year-old Japanese man. He presented with a gradual onset hoarseness and dyspnoea over the course of 2 years. He underwent excision of the lesion in theatre and was noted to bleed profusely at the time of operation. Bleeding settled and on discharge to the ward he began to bleed profusely a second time, and on this occasion, lost 1000 mL of blood. He underwent emergency tracheostomy for airway

Patient's perspective

- The high quality of care I received in my time of need, at a time when you all get negative press, I would just like to let you know how grateful I am to the whole team.
- You don't realise how scary it is to be a patient until you are sitting in the bed worrying about what is to come. The friendly staff and smiling faces were such a comfort during those tough weeks, and I made a few friends out of the other patients who were facing equally difficult times. Every day is a blessing.
- It was amazing, I hadn't talked properly in years, but following the removal of the tumour I could talk away without my voice getting tired, all I can say is thanks to you all.

Learning points

- ▶ Laryngeal tumours in comparatively low-risk individuals should alert the clinician to the possibility of an unusual diagnosis.
- ▶ Subglottic lesions may be vascular in nature and should be investigated with radiological imaging and angiography prior to biopsy, to avoid complication.
- ▶ Tracheostomy is an effective intervention to protect the airway in a patient with major airway haemorrhage.
- ▶ This case highlights the importance of the multidisciplinary team in the optimal management of unusual or complex cases, to ensure a satisfactory outcome.

protection. Tracheostomy was removed 2 weeks later and no further bleeding was experienced.

Anderson and Weinstein³ describe the case of a patient who presented with a cherry red mass of the vocal cord on a background of a 3-year history of hoarseness. This was excised via laser, however, the patient returned 13 years later with a 6-week history of similar symptoms and a recurrence of the disease was noted. Jun *et al*, in 2008, described the presentation of a

52-year-old-man with progressive hoarseness, pain, difficulty swallowing and breathlessness on lying flat.⁴ Subsequent examination and treatment identified an unusual 2×4 cm angioleiomyoma. Wang *et al*⁵ identified 21 patients diagnosed with vascular leiomyomas of the head and neck region over a 13-year period. Of these, none were involved within the larynx but were found in other regions of the head and neck.

Acknowledgements The authors would like to thank Dr Cromie and his team in Antrim.

Competing interests None declared.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

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