



Bibby, A., & Maskell, N. (2017). Benign pleural schwannoma presenting with a large, blood-stained pleural effusion. *Thorax*.
<https://doi.org/10.1136/thoraxjnl-2017-211102>

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FRONT SHEET

Benign pleural schwannoma presenting with a large, blood-stained pleural effusion

Anna C Bibby^{1,2}, Richard Daly³, Eveline Internullo⁴, Anthony J Edey⁵ & Nick A Maskell^{1,2}

Affiliations:

1. University of Bristol, Bristol UK.
2. North Bristol Lung Centre, North Bristol NHS Trust, Bristol UK.
3. Department of histopathology, North Bristol NHS Trust, Bristol UK.
4. Department of Thoracic Surgery, University Hospitals Bristol NHS Foundation Trust, Bristol UK.
5. Department of radiology, North Bristol NHS Trust, Bristol UK

Corresponding author:

Anna Bibby

Address: Academic Respiratory Unit, University of Bristol, 2nd Floor L&R building, Southmead Hospital, Bristol BS10 5NB.

Email: Anna.Bibby@bristol.ac.uk

Tel: 0117 414 8049

Funding statement

This research received no specific grant from any funding agency in the public, commercial or not-for-profit sectors.

Conflicts of interest

All authors have completed the ICMJE uniform disclosure at www.icmje.org/coi_disclosure.pdf and declare: ACB is the holder of a National Institute for Health Research (NIHR) Doctoral Research Fellowship (DRF-2016-09-065). The views expressed are those of the author(s) and not necessarily those of the NHS, the NIHR or the Department of Health. NAM has received an unrestricted research grant from CareFusion (San Diego, US) and has sat on the advisory board for CareFusion (San Diego, US).

Author contributions

ACB, EI & NAM conceived the idea for the article. AJE provided the radiology images and wrote the corresponding text. EI supplied the photo in Figure 2 and wrote the text regarding the surgical procedure. RD supplied the histological pictures and wrote the corresponding text. ACB wrote the rest of the article, with input and guidance from NAM. All authors reviewed and approved the final manuscript.

Word count: 520

Figures: 2

References: 3

Appendices: 1

ARTICLE

A previously fit and well 43-year-old carpenter presented with 8 weeks of worsening breathlessness and a dry cough. He was an ex-smoker of 10 pack years with no history of asbestos exposure, thoracic radiation or recent trauma.

Chest radiograph and computed tomography thorax are shown in Figure 1. Pleural aspiration was undertaken and 1.5L of blood-stained fluid was removed, with symptomatic improvement. The fluid was exudative, with LDH 437_{U/L} (serum 374_{U/L}) and protein 47_{g/L} (serum 77_{g/L}). Infection was excluded by negative microbiological culture and normal inflammatory markers. Cytology revealed numerous red blood cells, scattered lymphocytes and eosinophils, but no malignant cells.

The patient underwent ultrasound-guided core biopsy, which revealed a spindle cell neoplasm with positive immunohistochemical staining for vimentin and AE1/AE3, and S100 positivity in many nuclei (Fig 2). Haemosiderin pigmentation was noted (Fig 2). The features were consistent with a low-grade nerve sheath tumour, specifically schwannoma.

PET-CT was undertaken to assess for metastatic disease (Fig 1). This confirmed a left paraspinal mass with heterogeneous SUV avidity (maximum 6.6) and a moderately avid internal mammary chain lymph node (maximum SUV 3.0).

He underwent left-sided single-port Video-Assisted Thoracic Surgery to exclude pleural malignancy. The procedure was converted to thoracotomy to allow complete resection of tumour. Macroscopically, the tumour was a heterogeneous, lobulated mass, measuring 105x111x98mms and encapsulated by a thin fibrous layer (Fig 1). Cysts were visible on the surface of the tumour, some of which appeared defective or ruptured. The histological features and immunohistochemical profile confirmed schwannoma, with no evidence of malignancy. The mass was completely excised (R0). The faintly PET-avid lymph nodes were also removed and were benign.

The patient had an uneventful recovery and was discharged 4 days postoperatively. He was asymptomatic when reviewed three months later.

Schwannomas are rare tumours arising from Schwann cells - specialised myelin producing cells of peripheral nerve sheaths.¹ They represent 1-2% of thoracic tumours, usually occurring in the third or fourth decade, with an equal sex distribution.¹ Schwannomas are typically benign, slow-growing and asymptomatic and are often diagnosed incidentally. Malignant schwannomas exist, but are significantly rarer, occurring at a ratio of 1:11 compared with benign schwannomas.²

Although malignant schwannomas frequently present with pleural effusions caused by invasion of local structures, effusions are uncommon in benign schwannomas.² In a recent series of 49 cases, only 1 presented with an effusion.³ Review of the literature revealed just four cases of benign schwannomas with pleural effusion since 1946 (see Appendix A). In all cases the effusion was blood-stained.

Combined with the frequent histological finding of haemosiderin, this suggests spontaneous tumour haemorrhage as the possible aetiology of effusions. The appearance of the tumour in this case suggests cyst rupture also plays a role in the development of pleural effusions.

An alternative explanation would be chronic pleural irritation by the tumour, causing fluid build-up and intermittent bleeding from capillary beds. This may explain the enlarged mammary chain lymph node seen in this case.

This case confirms that benign nerve sheath tumours such as schwannomas can present with pleural effusions. Effusions may be large and blood-stained, and malignancy should not be assumed on the basis of these factors.

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