

**Case
Report**

Mediastinal Ancient Schwannoma Causing Intrathoracic Bleeding

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Spontaneous hemothorax caused by the rupture of a benign schwannoma has rarely been reported. Herein, we present the successful excision of an extremely rare case of mediastinal ancient schwannoma causing intrathoracic bleeding. A 27-year-old man was admitted to our emergency department because of back pain and dyspnea. Computed tomography revealed massive pleural effusion with a posterior mediastinal tumor. We performed a resection of the tumor which had ruptured, and the tumor was diagnosed as an ancient schwannoma.

Keywords: ancient schwannoma, mediastinal tumor, intrathoracic bleeding, hemothorax

Introduction

Schwannomas are benign neurogenic tumors that may occur in any region of the body. When these tumors exhibit unusual growth accompanied by regressive changes, they are called ancient schwannomas. We report a rare case of intrathoracic bleeding due to a mediastinal ancient schwannoma and detail its successful resection.

Case Report

A 27-year-old man presented to a local clinic because of sudden back pain with dyspnea. He was referred to our

emergency department after a chest X-ray that revealed left pleural effusion. He had no significant medical, social, or family history. Contrast-enhanced computed tomography (CT) showed a left intrathoracic tumor measuring $8 \times 5 \times 10$ cm located beside the vertebra with bloody pleural effusion, which compressed the diaphragm below (**Fig. 1**). The mass was enhanced heterogeneously. To reduce intrathoracic bleeding, we initially considered transcatheter arterial embolization; however, the CT did not show extravasation of the enhancing agent from the chest wall.

We performed emergency surgery with a posterior lateral incision, an additional small incision and a thoracoscopic port. The ruptured and bleeding tumor was located in the thoracic cavity with severe adhesions to the posterior chest wall and diaphragm. Since we were forced to resect a part of the chest wall with the tumor, the origin of the tumor was not conclusively identified, but it was considered to be the intercostal or sympathetic nerve. The tumor was entirely resected. The diaphragm was partially deflected and needed repair by suturing. The blood loss volume was 1090 g including the bloody pleural effusion. Active bleeding was not seen at the start of the operation, and no specific vessels were identified as the source of hemorrhage. We presumed that the tumor destruction resulted in hemothorax. The resected tumor measured $9.8 \text{ cm} \times 8.5 \text{ cm} \times 4.6 \text{ cm}$ and weighed 235 g. It consisted of a solid component with necrotic

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Received: May 27, 2019; Accepted: August 16, 2019
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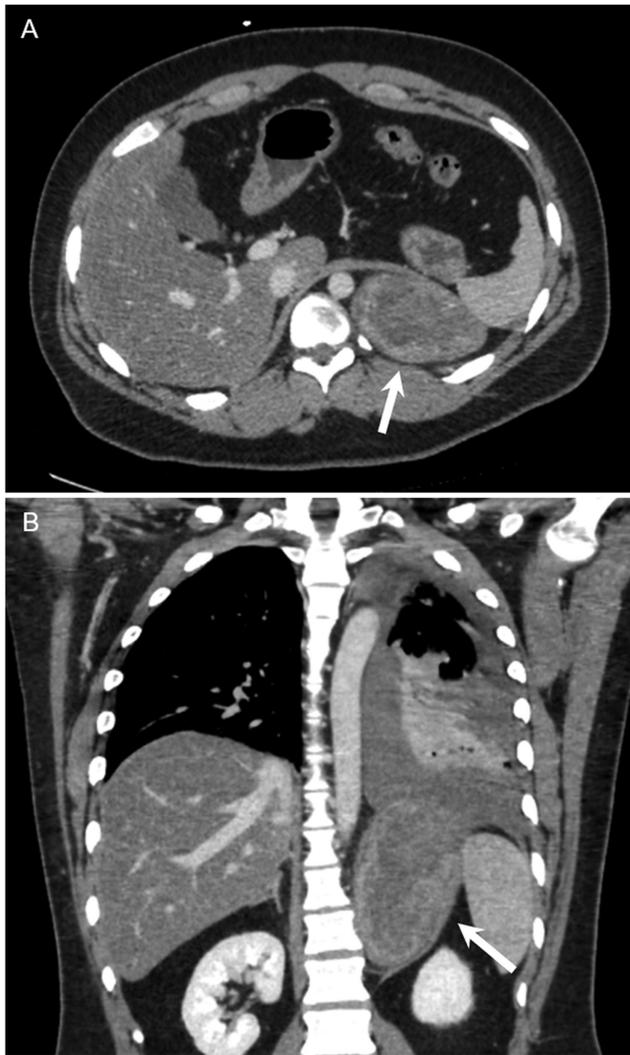


Fig. 1 Contrast-enhanced CT showing an intrathoracic tumor (white arrow) with left bloody pleural effusion, compressing the diaphragm below. CT: computed tomography

changes and cystic degeneration with rupture (**Fig. 2A**). The histological examination revealed that the tumor was comprised of two regions. One was filled with spindle cells showing palisading, and the other was less cellular with mucinous changes. Mitotic cells were absent. The tumor cells were noted to be immunoreactive to S-100 protein and vimentin, and collagen fiber was stained blue with Masson trichrome stain (**Figs. 2B–F**). Accordingly, the tumor was diagnosed as a benign schwannoma with extensive degenerative changes (ancient schwannoma).

The postoperative period was uneventful, and the patient was discharged without complications.

Discussion and Conclusion

Schwannoma is a neurogenic tumor which can occur in any region of the body, such as the head, neck, and extremities. It also arises in the mediastinum, which is the most common location of intrathoracic neurogenic tumors, with some cases specifically originating from the intercostal or sympathetic nerve.¹⁾ Some of these tumors grow to unusually large sizes and have a high frequency of regressive changes, such as fatty degeneration, hemorrhage, and cystic formation. Tumors exhibiting these characteristics are described as ancient schwannomas.²⁾

We rarely encounter a case of intrathoracic bleeding caused by neurogenic tumors. Although there are some reports of malignant peripheral nerve sheath tumors, multiple neurofibroma associated with Von Recklinghausen's disease, or trauma associated with bloody pleural effusion,³⁾ there are only five published cases that reported bleeding due to a benign schwannoma (**Table 1**,^{4–8)}). In previous cases, the tumors occurred more often in women than in men, at a relatively early age. Patients complained of symptoms associated with hemothorax caused by destructive tumors 60–100 mm in size.

Resection of an ancient schwannoma in the thoracic cavity is reported to be challenging due to its typically large size and severe adhesions.⁹⁾ It is necessary to remove the tumor from the pleura with caution, as the risk of bleeding is high due to nutrient vessels from the chest wall. In our case, we had difficulties in separating the tumor from the bottom of the posterior thoracic wall, and moreover, the diaphragm was partially deflected by the tumor and needed repair by suturing. The thoracoscopic port was useful to ensure the field of view, which was hidden behind the huge tumor mass.

We suggest that the preoperative imaging should be reviewed carefully in planning the surgical approach. First, the necessity of arterial embolization of the intercostal artery should be evaluated by searching for extravasation of contrast agent from the chest wall. Second, the shape and extent of the tumor should be evaluated. For example, a dumbbell-shaped tumor extending to the spinal canal may make the resection even more complicated. Furthermore, non-resected cases require periodical follow-up, as there is the potential for growth and rupture in asymptomatic benign schwannoma.

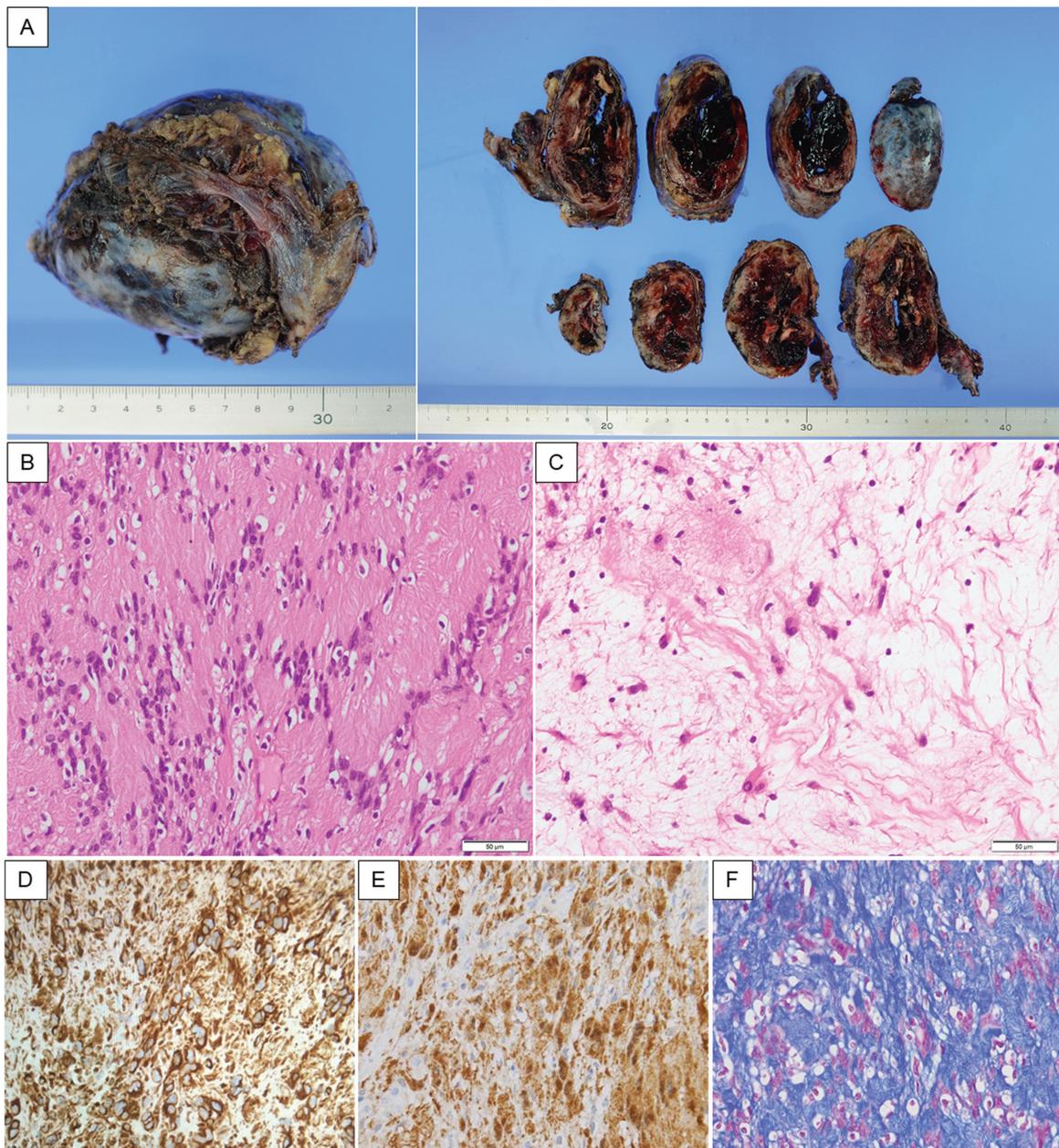


Fig. 2 (A) Resected tumor consisting of a solid component with necrotic changes and cystic degeneration with rupture, (B) HE stain in a region with spindle cells showing palisading, (C) HE stain in a less cellular region with mucinous changes, (D) S-100 protein (+), (E) Vimentin (+), and (F) collagen fiber stained blue for Masson trichrome stain. HE: hematoxylin and eosin

Table 1 Cases of benign schwannoma presenting as hemothorax

Author	Year	Age (years)	Sex	Size (mm)	Side	Main complaint
Reyburn ⁴⁾	1996	40	M	70	Left	Chest pain
Lee ⁵⁾	1998	27	M	60 × 50 × 40	Left	Loin pain
Eröglu ⁶⁾	2002	37	F	100 × 80 × 50	Right	Dyspnea, chest pain
Morimoto ⁷⁾	2011	37	M	73 × 50 × 47	Left	Back pain
Ishibashi ⁸⁾	2016	73	M	80 × 50 × 40	Left	Chest pain, dyspnea
Our case	2018	27	M	98 × 85 × 46	Left	Back pain

Acknowledgment

We would like to thank Editage (www.editage.jp) for English language editing.

Disclosure Statement

The authors report no conflicts of interest.

References

- 1) Ribet ME, Cardot GR. Neurogenic tumors of the thorax. *Ann Thorac Surg* 1994; **58**: 1091-5.
- 2) Rosai J. Rosai and Ackerman's Surgical Pathology. Volume 2. Ninth edition. St. Louis, MO: Mosby; 2004: pp 495.
- 3) Vaziri M, Mehrazma M. Massive spontaneous hemothorax associated with Von Recklinghausen's disease. *Ann Thorac Surg* 2006; **82**: 1500-1.
- 4) Reyburn TA, Morse WS, Solomon NJ. Schwannoma: an unusual case of sudden chest pain and hemothorax. *Emergency Radiol* 1996; **3**: 293-5.
- 5) Lee MH, Graham AN, Nicholson AG, et al. Solitary cellular schwannoma presenting with haemothorax. *J R Soc Med* 1998; **91**: 596-7.
- 6) Eröglu A, Kürkcüoğlu IC, Karaoglanoglu N, et al. Spontaneous hemothorax due to cellular schwannoma. *Case Rep Clin Pract Rev* 2002; **3**: 141-3.
- 7) Morimoto J, Nakajima T, Iizasa T, et al. Successful resection of schwannoma from an intercostal nerve causing bloody pleural effusion: report of a case. *Surg Today* 2011; **41**: 989-91.
- 8) Ishibashi H, Takasaki C, Okubo K. Successful excision of a massive bleeding schwannoma by thoracoscopic surgery. *Asian Cardiovasc Thorac Ann* 2016; **24**: 484-6.
- 9) Wu Y, Zhang J, Chai Y. Giant mediastinal schwannoma located in the lower right side of the chest. *Nigerian J Clin Pract* 2016; **19**: 678-80.