

Hemorrhagic Shock due to Intrathoracic Rupture of an Osteosarcoma of the Rib

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A 13-year-old girl presented with dyspnea and chest pain. Chest radiography showed a massive left pleural effusion. Computed tomography revealed a tumor of the fourth rib. A large bloody effusion was drained. Her anemia worsened (hemoglobin: 4.8 g/dl), and hemorrhagic shock ensued. An emergency thoracotomy was performed. Bleeding from the ruptured tumor was identified. The fourth rib, the tumor, and the adjacent tissues were resected. Histopathologic examination revealed a ruptured primary osteosarcoma of the rib with pleural dissemination. (Ann Thorac Cardiovasc Surg 2001; 7: 232–4)

Key words: osteosarcoma of the rib, hemothorax, hemorrhagic shock

Introduction

Osteosarcomas, which commonly originate in the long bones, rarely occur in the chest wall.¹⁾ The sudden onset of hemorrhagic shock due to intrathoracic rupture of a rib osteosarcoma, has not been reported.

Case Report

An otherwise normal 13-year-old girl complained of slight chest pain on the left side without a history of injury. She was admitted with severe dyspnea 3 days after the initial onset of pain. Chest radiography showed a massive pleural effusion on the left (Fig. 1a). Diagnostic thoracentesis revealed bloody fluid. Chest sonography showed a massive pleural effusion and a mass adherent to the chest wall. Computed tomography revealed a tumor of the fourth rib with calcifications (Fig. 1b).

Laboratory investigation revealed anemia (hemoglobin: 9.4 g/dl) and hypoxia (PO₂: 68 mmHg). The serum alkaline phosphatase concentration was increased (458

IU/l), but the serum calcium concentration was not elevated. Therapeutic thoracentesis yielded 1400 ml of bloody fluid, and the drainage persisted. The hemothorax did not improve on chest radiography, and the anemia worsened (hemoglobin 4.8 g/dl). The blood pressure continued to drop until the patient was in hemorrhagic shock.

In an emergency operation via a left anterolateral thoracotomy, 1900 ml of coagulated blood was removed from the left pleural cavity. A tumor of the fourth rib was identified. Active bleeding continued from the ruptured tumor. A subpleural hematoma adherent to the tumor measured 4 cm in diameter and coincided with the mass found on sonography.

Active bleeding continued from the ruptured tumor, which was controlled by ligation of the intercostal vessels. The fourth rib was resected with the intercostal muscles around the tumor. There was macroscopic evidence of invasion of the rib (Fig. 2a). The serratus anterior muscle, which was adherent to the fourth rib which contained the tumor, was partially resected. A degenerative lesion in the lung corresponding to the site of the tumor was also partially resected. Several pleural lesions were resected with adequate margins. A total of 1600 ml of blood was transfused.

The postoperative course was uneventful. Histopathologic examination of the tumor revealed primary osteosarcoma of the fourth rib (Fig. 2b). The pleural lesions also

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Received December 20, 1999; accepted for publication May 15, 2001.

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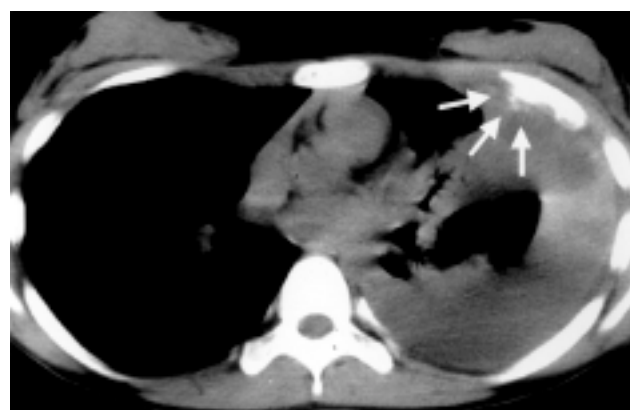


Fig. 1. a: Chest radiography on emergency admission showing a massive left pleural effusion.
b: Computed tomography showing a tumor of the left fourth rib with calcifications (arrows) and a massive pleural effusion.

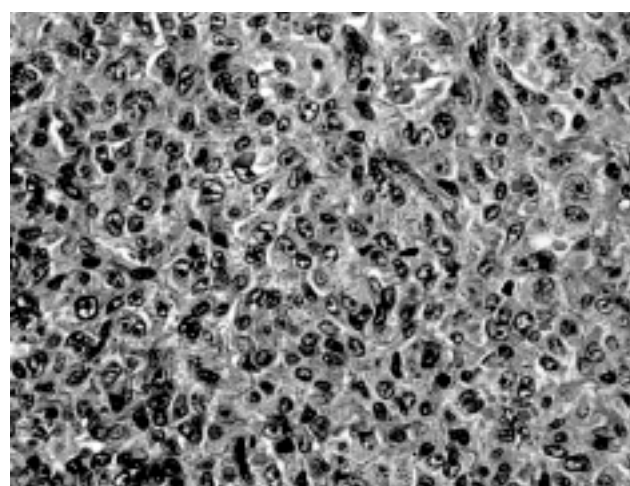
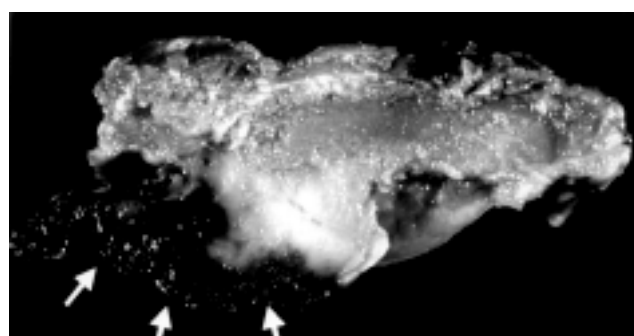


Fig. 2. a: Cut surface of the resected specimen showing tumor invasion into the rib and a hematoma (arrows).
b: Photomicrograph showing pallisading spindle cells and giant cells with bone formation. (Hematoxylin and eosin stain, magnification, $\times 400$)

contained osteosarcoma. Specimens from the left lung and the serratus anterior muscle contained no tumor. Post-operative bone scintigraphy showed no other metastatic lesions. The patient underwent chemotherapy. At 2 years and 9 months after surgery, there is no evidence of recurrence.

Comment

Primary chest wall tumors are uncommon. These lesions have been reported to originate from a variety of cell types. But more than half of primary chest wall tumors are malignant. Although osteosarcoma is the most common malignant bone tumor, osteosarcoma of the rib is

rare. The most common tumors of the ribs are chondrosarcoma and Ewing's sarcoma.¹⁻⁵⁾ Only two reports of osteosarcoma of the ribs associated with a bloody pleural effusion are found in the literature.^{6,7)} Hemorrhagic shock due to rupture of an osteosarcoma of the rib has not been reported. Usually, osteosarcoma presents with chest pain. In our case, chest pain and rupture occurred simultaneously. It is therefore postulated that the tumor grew rapidly. Since the chest wall is rich in blood vessels and osteosarcomas are usually surrounded by a hypervascular capsule, a massive pleural effusion with hemorrhagic fluid may occur.⁸⁾ In our case, massive bleeding occurred due to rupture of the tumor. Histopathologic examination showed invasion of the tumor into the intercostal vessels, which may have precipitated the rupture.

Treatment of osteosarcoma of the rib is radical excision sometimes with chest wall reconstruction.^{3,4,9,10)} In an emergency operation as in this case, if malignancy is suspected, thorough resection should be performed.

Another nontraumatic cause of massive intrapleural bleeding and hemorrhagic shock is the intrathoracic rupture of a pulmonary arterio-venous fistula, we have also experienced such a case. Considering the importance of intervention to outcome in these cases, prompt diagnosis and treatment of the underlying cause of a bloody pleural effusion is recommended, even if emergency thoracotomy is required.

References

1. Dahlin DC, Coventry MB. Osteogenic sarcoma: a study of six hundred cases. *J Bone Joint Surg* 1967; **49**: 101-10.
2. Ochsner A Jr, Lucas GL, McFarland GB. Tumors of the thoracic skeleton: review of 134 cases. *J Thorac Cardiovasc Surg* 1966; **52**: 311-21.
3. Groff DB, Adkins PC. Chest wall tumors. *Ann Thorac Surg* 1967; **4**: 260-81.
4. Stelzer P, Gay WA Jr. Tumors of chest wall. *Surg Clin North Am* 1980; **60**: 779-91.
5. Eskenasy A. Histopathology of tumorous ribs: a review of 130 cases. *Morphologie et Embryologie* 1985; **31**: 35-50.
6. Arbib F, Thevenet F, Gamondes JP, Heyrand JD, Gontier C, Loire R. Primary tumor of the thoracic wall unusual in aged patients: costal osteosarcoma. *Revue de Pneumologie Clinique* 1991; **47**: 220-4. (in French)
7. Nakamura A, Yamada Y, Yamamoto T, Yamamoto K, Takeuchi T. A case of osteosarcoma of the rib with bloody pleural effusions. *Nihon Kyoubu Shikkann Gakkai Zasshi* 1993; **31**: 394-8. (in Jpse.)
8. Maher GG, Berger HW. Massive pleural effusion: malignant and nonmalignant causes in 46 patients. *American Review of Respiratory Disease* 1972; **105**: 458-60.
9. Pairolero PC, Arnold PG. Thoracic wall defect: surgical management of 205 consecutive patients. *Mayo Clin Proc* 1986; **61**: 557-63.
10. Grosfeld JL, Rescorla FJ, West KW, et al. Chest wall resection and reconstruction for malignant conditions in childhood. *J Pediatr Surg* 1988; **23**: 667-73.