Case Report

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Giant Lymph Node Hyperplasia (Castleman's Disease) as a Rare Cause of Back Pain

Giant lymph node hyperplasia (Castleman's disease) is a nonneoplastic lymphoproliferative disorder of unknown etiology that usually occurs in the chest. Its morphological recognition is based on a composition of various histological features. The mass is often asymptomatic, but it can cause nonspecific thoracic symptoms, such as regional pain. This disease can be found wherever lymph nodes are present, but two-thirds of these tumors are found in the chest, along the tracheobronchial tree in the mediastinum or lung hilus. However, we experienced an unusual case of Castleman's disease as a cause of back pain that was localized in the posterior mediastinum bordering the chest wall.

KEY WORDS : Castleman's disease · Giant lymph node hyperplasia · Hyaline vascular type · Mediastinum · Back pain.

INTRODUCTION

Castleman's disease, also known as "angiofollicular lymph node hyperplasia", "giant lymph node hyperplasia"⁷⁾, and "angiomatous lymphoid hamartoma", is a rare nonneoplastic lymphoproliferative disorder of unknown etiology. Castleman's disease was first described in 1954 as a form of benign lymph node hyperplasia simulating thymoma²⁾. This disease can be found wherever lymph nodes are present, but two-thirds of cases are found in the chest, along the tracheobronchial tree in the mediastinum or lung hilus, but it can occur in other areas such as the pelvis, neck and retroperitoneum⁷⁾. However, chest-wall involvement is a rare manifestation of Castleman's disease. We report our experience of Castleman's disease as a cause of back pain extending from the border between the posterior mediastinum and intercostal space.

CASE REPORT

A 25-year-old man was referred to our department due to back pain. He complained of vague back pain without definitive localization. A simple chest X-ray revealed a mass-like lesion in the left paravertebral area of T10-12 (Fig. 1). We then examined the thoracic magnetic resonance imaging (MRI) and confirmed that there was a mass in the posterior mediastinum. The lesion presented as a homogeneous, well-marginated soft-tissue mass located in the left costovertebral sulcus. We presumed that the mass was a neurogenic tumor and performed a left lateral thoracotomy and exploration. The mass was well encapsulated and covered with parietal pleura in the left T10-12 paravertebral area of the posterior mediastinum. We then removed it completely along with the surrounding soft tissue. The resected mass measured 2 × 3 × 4 cm in diameter. It was soft and the surface of the tumor was whitish to yellowish in color (Fig. 2A). Histopathological evaluation confirmed the tumor to be Castleman's

Fig. 1. The chest X-ray showed a left paravertebral opacity (arrow) 2 × 4 cm in diameter at T10-12.

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Fig. 2. A : The surgical specimen was soft, whitish to yellowish in color and measured $2 \times 3 \times 4$ cm in size. B : Histopathologically, this mass had the characteristics of the hyaline-vascular type of Castleman's disease. Note how the germinal centers are surrounded by lymphocytes, thereby imparting a classic "onion skin" appearance to the specimen.

Fig. 3. Pre and postoperative magnetic resonance (MR) images demonstrating total removal of the lesion in the left T10-12 paravertebral area. A : Preoperative Gadolinium (Gd)-enhanced T1-weighted coronal and axial MR images. B : Postoperative Gd-enhanced T1-weighted coronal and axial MR images.

disease. Microscopic evaluation revealed the mass to be composed of lymphoid follicles that contained multiple capillaries surrounded by hyaline sheaths that were characteristic of the hyaline vascular type of Castleman's disease (Fig. 2B). Postoperative magnetic resonance imaging confirmed total removal of the mass in the left T10-12 paravertebral area of the posterior mediastinum (Fig. 3). The patient showed an uneventful postoperative recovery and discharged from the hospital with no postoperative symptoms after 9 days. No further treatment has been required to date.

DISCUSSION

Since Castleman et al. first reported a case of "localized mediastinal lymph node hyperplasia resembling thymoma"²⁾, similar cases have been reported as "Castleman's disease." Although most of these lesions occur within the chest, other regions, including the neck, pelvis, retroperitoneum, and axilla, may be involved⁷⁾. There is no remarkable sex predominance or identifiable risk factors for the development of Castleman's disease⁸⁾, which has been reported to arise as a tumor, inflammation or hamartoma^{5,11)}, but its cause remains uncertain.

Its morphological recognition is based on a combination of various histological features, but it is generally classified into two subtypes, the hyaline-vascular type and plasma cell type. However, Castleman's disease can be further classified into two types, solitary and multicentric. Chest-wall involvement by Castleman's disease is a rare manifestation of this pathology, and only seven cases had been reported in the international literature as of 2004^{4,12)}. These cases were reviewed by Erdogan et al., who reported that there was no specific symptom among the seven patients, and only one man was histologically confirmed to have the plasma cell type⁴⁾. There have been four reports of spinal Castleman's disease occurring as an extradural lesion in the cervical and thoracic region^{1,3,6,10)}.

Patients are usually either asymptomatic or have nonspecific symptoms, such as coughing, dyspnea, chest pain, respiratory infection, and back pain, mainly caused by tracheobronchial compression¹³⁾. The back pain experienced by our patient was probably caused by the compression of the parietal pleural nerves by the mass lesion⁹⁾. The resolution of the pain after the tumor had been removed confirmed that the lesion was the source of the patient's discomfort. No

other source for the patient's pain could be identified. The patient's back pain disappeared after surgery, and no perioperative complications were found.

CONCLUSION

In this case, chest-wall localization was a rare manifestation of Castleman's disease. Due to the unusual localization of this disease, nonspecific symptoms and lack of specific radiologic findings, many chest and neurosurgeons do not consider this disease preoperatively. We report an unusual case in which Castleman's disease caused back pain.

References

1. Alper G, Crumrine PK, Hamilton RL, Albright AL, Wald ER : Unusual case of inflammatory spinal epidural mass (Castleman syndrome). *Pediatr Neurol* 15 : 60-62, 1996
2. Castleman B, Iverson L, Menendez VP : Localized mediastinal lymphnode hyperplasia resembling thymoma. *Cancer* 9 : 822-830, 1956
3. Eisenstat RS, Price DB, Rosenthal AD, Schuss AL, Katz DS : Thoracic epidural Castleman's disease. *AJR Am J Roentgenol* 178 : 208-210, 2002
4. Erdogan A, Eser I, Ozbilim G : Posterior mediastinal localization of Castleman's disease : report of a case. *Surg Today* 34 : 772-773, 2004
5. Inada K, Hamazaki M : Localized mediastinal lymph-node hyperplasia resembling thymoma; a case report. *Ann Surg* 147 : 409-413, 1958
6. Kachur E, Ang LC, Megyesi JF : Castleman's disease and spinal cord compression : case report. *Neurosurgery* 50 : 399-402; discussion 402-393, 2002
7. Keller AR, Hochholzer L, Castleman B : Hyaline-vascular and plasmacytic types of giant lymph node hyperplasia of the mediastinum and other locations. *Cancer* 29 : 670-683, 1972
8. Kim JH, Jun TG, Sung SW, Shim YS, Han SK, Kim YW, et al : Giant lymph node hyperplasia (Castleman's disease) in the chest. *Ann Thorac Surg* 59 : 1162-1165, 1995
9. Lakadamyali H, Ergun T, Oguzkurt L : Alcohol ablation therapy of an atypically located symptomatic bronchogenic cyst : a case report. *Cardiovasc Intervent Radiol* 30 : 1274-1276, 2007
10. Lee HY, Lee SH, Kim SN : Castleman's disease : unusual case of inflammatory spinal dorsal epidural mass : case report. *Neurosurgery* 50 : 396-398, 2002
11. Patel U, Forte V, Taylor G, Sirkin W : Castleman's disease as a rare cause of a neck mass in a child. *J Otolaryngol* 27 : 171-173, 1998
12. Rena O, Casadio C, Maggi G : Castleman's disease : unusual intrathoracic localization. *Eur J Cardiothorac Surg* 19 : 519-521, 2001
13. Slotwiner A, Garwacki CP, Moll S : Castleman's disease. *Am J Hematol* 73 : 64-65, 2003