A Rare Case of Primary Intercostal Leiomyoma: Complete Resection Followed by Reconstruction Using a Gore-Tex® Dual Mesh

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We report the case of a 28-year-old woman with primary intercostal leiomyoma who presented with a complaint of right chest pain. Contrast-enhanced computed tomography (CT) demonstrated a slightly enhanced solid mass in the right anterior fifth intercostal space. Percutaneous needle biopsy revealed spindle cells without evidence of malignancy. Wide en bloc excision of the chest wall, including the anterior fifth and sixth ribs and the upper costal arch, was performed. This way, the mass was completely resected without exposure, and the chest wall defect was reconstructed using a Gore-Tex® dual mesh. Histopathological analysis confirmed localized primary intercostal leiomyoma. The patient has been disease-free for more than 2 months since surgery.

Primary leiomyomas of the chest wall are extremely rare. To the best of our knowledge, 9 cases of leiomyoma of the pleura have been reported till date, but this is the first case report of an intercostal leiomyoma of the chest wall. This case report describes the clinical course of this case and presents a review of the relevant literature.

Keywords: chest wall, leiomyoma, intercostal space, dual mesh

Introduction

Several histological types of chest wall neoplasms have been reported.1) Because tumors of the pleura or chest wall cannot be differentiated radiologically, a final diagnosis can only be established by histological examination. Leiomyomas originating from the chest wall are extremely rare benign tumors. To the best of our knowledge, only 9 cases with the pleura as the primary site of involvement have been reported till date.2–9) Here we describe the first case, as per our knowledge, of an extremely rare leiomyoma originating in the intercostal space in a 28-year-old woman.

Case Report

A 28-year-old woman without any significant past history was presented at a local hospital with a complaint of right-sided chest pain since 6 months ago. Contrast-enhanced computed tomography (CT) demonstrated a slightly enhanced solid mass in the right anterior fifth intercostal space. Ultrasound echo-guided percutaneous fine-needle aspiration was performed and cytology revealed spindle cells without any evidence of malignancy. The patient was subsequently referred and admitted to our hospital for further treatment. Physical examination and routine blood biochemistry findings were normal. Chest roentgenography revealed a mass in the right lower lung field, and CT and magnetic resonance imaging (MRI) revealed a well-circumscribed solitary tumor...
measuring 43 × 30 × 37 mm in the right anterior fifth intercostal space near the costal arch. The mass was slightly enhanced homogeneously with no infiltration to the lungs or adjacent ribs (Fig. 1A and 1B). A benign tumor was suspected, and differential diagnoses included neurogenic tumor, desmoid tumor, solitary fibrous tumor, or osteochondroma. Surgical intervention was scheduled for the purpose of both diagnosis and treatment. Chest wall resection was performed through a transverse incision over the tumor. The tumor was resected without exposure by combined resection of the overlying anterior serratus muscle, the rectus abdominis as a lateral margin, the costal arch, and the fifth and sixth costal bones. Tumor-free margins of at least 2 cm were achieved. Then, a Gore-Tex® dual mesh, an expanded polytetrafluoroethylene prosthesis (ePTFE) (Japan Gore-Tex Inc., Tokyo, Japan), was cut to fit the defect of the bony chest wall and tightly sutured tense as a drum using 2–0 vicryl (Ethicon, Tokyo, Japan) through drilled holes in the ribs and costal cartilage along the chest wall defect. The dual mesh patch was placed with the patterned indented surface upward and the medial sutured side facing the posterior layer of the rectus abdominis sheath. No chest tube was inserted into the pleural space, but a suction drain was placed over the patch. The total surgical duration was 155 minutes, and the total blood loss was 30 ml. The patient’s postoperative course was uneventful. The drain was removed on the third postoperative day and the patient was discharged on the fifth postoperative day. Gross pathological examination revealed a tumor measuring 42 × 33 × 32 mm without infiltrative growth and a pale yellow cut surface. Histopathological examination of hematoxylin and eosin (H&E)-stained specimens revealed monotonous proliferation of interlaced fascicles of spindle cells without malignant characteristics such as mitosis or cellular dysplasia. Immunohistochemistry revealed positive staining of the tumor cells for smooth muscle actin, desmin, and...
caldesmon (Fig. 2). Tumor tissue stained negative for CD34, bcl2, CD117, S-100, and progesterone receptor. Specimens were <1% positive for estrogen receptor. Gynecological examination showed no evidence of uterine leiomyoma; therefore, the chest wall was diagnosed as the primary site. On the basis of these findings, a diagnosis of benign leiomyoma arising from the chest wall was made. At 2 months after surgery, three-dimensional (3-D) CT images revealed no evidence of recurrence, and the bony chest wall defect appeared to be perfectly reconstructed with the dual mesh (Fig. 1C and 1D).

Discussion

Leiomyomas may occur in those parts of the body that constitute smooth muscle. They usually include the uterus, small intestine, or esophagus. Tumors of this type rarely originate in the chest wall. Till date, only 9 cases of leiomyomas originating from the pleura have been reported worldwide. Table 1 summarizes the patient and tumor characteristics of the nine previously described cases and our case. Of the total 10 cases including ours, five were asymptomatic and five had nonspecific chest pain. From these reports, it appears that this type of tumor appears to occur more frequently in young to middle-aged females (8 of 10 cases). Routine blood biochemistry provides no contribution to diagnosis, while radiologic findings also cannot provide a definitive diagnosis of tumors of the chest wall. The origin of the previously reported tumors was the pleura. Primary leiomyomas of the pleura involve neither the ribs nor the skeletal muscle. In the case reported by Proca, et al., it was necessary to resect the chest wall in combination with resection of the third costal bone because of a secondary lesion in the pectoral muscle, which was implanted by preoperative CT-guided percutaneous biopsy. The other cases were reportedly resected with relative ease. Pleural tumors grow toward the intrathoracic cavity without progression into the intercostal space. Our case report is the first, as per our knowledge, to document a primary leiomyoma that grew mainly in the intercostal space.

Differential diagnoses for chest wall spindle cell neoplasms include smooth muscle tumors, malignant mesothelioma, spindle cell carcinoma, and other soft tissue tumors arising in adjacent organs. They also include solitary fibrous tumors in cases of circumscribed masses.
A definitive diagnosis of leiomyoma always requires confirmation of smooth muscle fibers without signs of malignancy (pleomorphism, mitotic figures, and poor differentiation) by H&E and immunohistochemical staining.\(^1,^{10}\) The histological features of H&E sections plus positive staining for smooth muscle actin, desmin, and caldesmon provide unambiguous evidence for the diagnosis of leiomyoma.

The leiomyoma is classified as angioleiomyoma, piloleiomyoma, or genital leiomyoma based on its origin. The cells of origin in six of the nine previous cases were reported to be vascular smooth muscle cells on the basis of clinicopathological findings, whereas it was unclear for the remaining 3 cases (Table 1). These tumors may arise from mesodermal cells that have acquired the capacity to differentiate along smooth muscle lines. It is reasonable to deduce tumorigenic origin on the basis of anatomic position, relationship with adjacent structures, macroscopic findings, histopathological manifestations, and occurrence time. In this case, the tumor was too large at the intercostal space to detect specifically-connected vascular wall. Furthermore, we could not find vascular channels among tumor cells microscopically.\(^5,^{8}\)

The original tumor lesion was obviously not involved with myoepithelial cells. Thus, we could not determine the exact tumorigenic origin of the tumor because it showed no positional relationship with adjacent structures and there was no additional information except for histological data.

The presence of estrogen and progesterone receptors supports the origin of benign metastasizing leiomyoma from uterine smooth muscle.\(^11\) However, in our case, gynecological examination revealed no evidence of uterine leiomyoma; moreover, estrogen receptor positivity was <1% according to immunohistopathological investigation. Therefore, the chest wall was identified as the primary tumor site.

Moran, et al. reported a case wherein tumors invaded adjacent tissue and were not respectable.\(^4\) Despite a benign histological appearance, tumors may increase in size with local invasion if not resected completely. The clinical course of leiomyomas of the chest wall is not always concordant with histological findings, but these tumors also have low malignant potential. However, surgical excision is justified and recommended because these tumors can grow very large, causing serious symptoms or signs, and degenerate into malignant tumors. The prognosis is fairly good for patients in whom tumors have been completely resected. Because it is difficult to
determine oncological predisposition in cases of chest wall leiomyomas, we recommend complete resection and long-term follow-up for all such cases.

When the bony chest wall is resected, reconstruction is performed to prevent postoperative complications such as paradoxical respiration, pneumonia, and atelectasis. In our case, the chest wall deformity was perfectly reconstructed with Gore-Tex® dual mesh, which is a pure and unique ePTFE comprising 2 functionally distinct surfaces. The smoother surface is designed for minimal tissue attachment, whereas the patterned, indented surface is designed for active tissue attachment. This material has been successfully used for the repair and reconstruction of the abdominal wall in cases of ventral incisional hernia or large hiatal hernia; however, only a few surgeons have reported lateral chest wall reconstruction using Gore-Tex® dual mesh. The advantages of this material are its ability to stabilize the chest wall and the development of fewer adhesions to inner organs. Conversely, seroma is the most common complication that occurs after prosthetic reconstruction. Use of a chest drain under negative pressure may prevent complications of seroma and infection; however, it also facilitates a high degree of adhesion between the dual mesh and adjacent tissues. In this case, the postoperative course was uneventful, and the chest wall deformity was perfectly reconstructed by the dual mesh as evidenced by 3-D CT. On the basis of our experience, we propose chest wall reconstruction using a dual mesh as a safe and effective procedure.

**Conclusion**

We report the first case, as per our knowledge, of primary intercostal leiomyoma, which is an extremely rare tumor with a benign histological appearance. Leiomyomas of the chest wall are not always concordant with histological findings and unrecteable growths usually have low malignant potential. However, it is difficult to determine the oncological predisposition of these tumors on the basis of their atypical infestation site. Therefore, we recommend complete resection and long-term follow-up for such cases. In addition, chest wall reconstruction using a dual mesh can be used as a safe and effective procedure in such cases.

**Disclosure Statement**

The authors report no conflicts of interest.

**References**