Case Report
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Chest Wall Leiomyomas: A Case Report and Review of the Literature

Göğüs Duvarı Leiomyomları: Olgu Sunumu ve Literature Özeti

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Özet

Leiomyomlar düz kaslardan köken alan benign yumuşak doku tümörleridir. Plevra ve göğüs duvarı bu tümörler için nadir lokalizasyonlardır. Göğüs duvarında yerleşmiş leiomyomalar, parietal plevranın subplevral bağ dokusundan başlayıp cilt altı dokuya kadar olan göğüs duvarı bölgesinde bulunan düz kaslardan köken alabilir. Literatürde bu tümörler "plevral leiyomyom" veya "chest wall leiyomyom" şeklinde farklı iki isimle tanımlanmışlardır. İster subplevral bağ dokusundan köken alsın, isterse göğüs duvarındaki diğer yapılardan köken alsın her iki grupta benzer radyolojik bulgulara sahiptir ve bu tümörlerin tam olarak nereden köken aldığını tespit edebilmek zordur. Parietal plevranında göğüs duvarının bir parçası olduğu kabul edildiğine göre, bu tümörlerin sadece "göğüs duvarı leiomyomu" olarak adlandırılmasının her iki grubu da kapsayacağını düşünmekteyiz. Olgu sunumunda, 56 yaşında erkek hastada sol hemitoraksta yer kaplayan kitle lezyonu tespit edildi. Rezeksiyon uygulanan hastanın patolojik incelemesinde kitlenin göğüs duvarı leiomyomu olduğu tespit edildi. Olgu, literatür özeti ile birlikte sunulmuştur.

Anahtar Kelimeler

Leiomyom; Göğüs Duvarı; Plevra; Benign

Abstract

Leiomyomas are benign soft tissue tumors that originate in the smooth muscles. Pleura and chest wall are uncommon location for such tumors. Leiomyomas located in the chest wall may originate from smooth muscles of the chest wall region beginning from the subpleural connective tissue of the parietal pleura, to the subdermal tissue. These tumors have been defined in literature using two different terms: "pleural leiomyoma" and "chest wall leiomyoma". Both possess similar radiological signs independent from the tissue from which they originate, which are either subpleural connective tissue or other structures in the chest wall. It is therefore difficult to make a clear determination of the tissue from which they originate. Parietal pleura is also accepted as a part of the chest wall, and thus, in our opinion, the term "chest wall leiomyoma" can be described as comprising both groups. The current study presents a 56-year-old male patient who had a space-occupying mass in the left hemi-thorax. After resection of the mass, pathological examination diagnosed it as chest wall leiomyoma.

Keywords

Leiomyoma; Chest wall; Pleura; Benign

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Introduction

Leiomyomas are benign soft tissue tumors that originate from the smooth muscles and are classified in the mesenchymal tumor family [1]. These benign tumors commonly originate from the urogenital tract, occasionally from the gastrointestinal tract, and rarely from the respiratory tract [2]. Pleural or chest wall leiomyomas are uncommon and atypical tumors, and only 12 cases have been published in the English literature. There have been six cases of pleural leiomyoma and six cases of chest wall leiomyoma presented to date. However, it is difficult to determinate the exact origin of these tumors. As reported below, we present a chest wall leiomyoma case and a review of pleural and chest wall leiomyomas, along with a proposal to combine both groups under one term, "chest wall leiomyomas".

Case Report

A 56-year-old man without any significant past medical history was admitted to the hospital with a complaint of backache for six months. His physical examination and routine laboratory examinations were normal. On radiologic examination, a chest x-ray revealed a well-circumscribed mass in the left hemi-thorax upper zone (Figure-1a). Computed tomography of the thorax revealed a 6 x 5.5 cm, solid, round, well-circumscribed mass lesion that had cystic components and compressed the lung parenchyma without any sign of infiltration (Figure -1b).

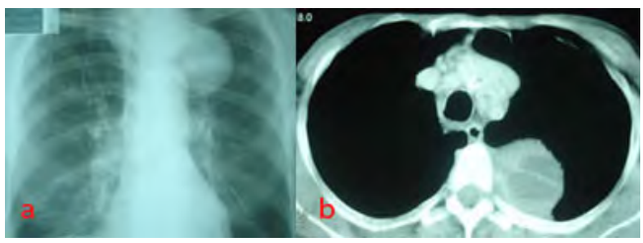


Figure 1. A chest x-ray revealed a well-circumscribed mass(a); Computed tomography confirmed solid, well-circumscribed mass lesion(b).

Our investigations did not reveal an extrathoracic leiomyoma that might lead to a metastasis to the chest wall. A decision was made for surgical excision with left muscle-sparing minithoracotomy. During surgical exploration, an extrapleural, encapsulated, smooth-surfaced chest wall mass, originating from the chest wall and in close association with the fifth intercostal space was observed (Figure-2a). The tumor was covered with the parietal pleura; after dissection of the pleura, it was easily removed en-bloc and excised without difficulty.

The macroscopic examination revealed that the tumor was soft, had a smooth surface, and measured 6 x 6x 5.5 cm. When the mass was dissected with all layers in the midline in two parts, solid, dirty, white, cystic cavities were observed (Figure-2b). The histopathological examination revealed benign spindle cells without mitosis, necrosis, or signs of cellular atypia (Figure 2c). There were randomly arranged, interlacing bundles of smooth muscle differentiation, with fascicles of spindle cells with oblong, bland nuclei without pleomorphism. Immunohistochemical staining revealed diffuse and strong positivity for both smooth muscle actin and desmin. With these findings, the diagnosis of leiomyoma was established without difficulty. The post-operative course was uneventful and the patient was discharged on post-operative sixth day. He was followed-up regularly and there were no signs of recurrence nine years after surgery.

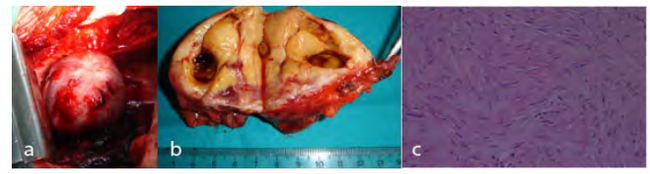


Figure 2. Intraoperative exposure of the mass(a); After the mass was dissected with all layers in the midline in two parts(b); Microscopic examination revealed benign spindle cells without mitosis, necrosis, or signs of cellular atypia(c).

Discussion

Leiomyomas were first described by Virchow in 1854 and can occur in any part of the body where smooth muscles are present [1]. However, leiomyomas originating primarily from the chest wall or pleura are extremely rare. On the chest wall, smooth muscles are located in the connective tissues, commonly in the walls of blood vessels.

The layers of the chest wall from the parietal pleura to the skin are ordered as mesothelial layer of the parietal pleura, superficial elastic layer, subpleural connective tissue layer of the parietal pleura, endothoracic fascia, cartilage and bony skeleton, straight muscles, and finally skin. Subpleural connective tissue is a part of the parietal pleura and this layer contains collagen fibers, elastic fibers, lymphatic network, nerve fibers, and small blood vessels. Leiomyomas located in the chest wall may originate from smooth muscles of the chest wall region beginning from the subpleural connective tissue of the parietal pleura, to the subdermal tissue. In the literature two different terms have been used: "pleural leiomyoma" and "chest wall leiomyoma". However, both groups exhibit a similar radiological appearance independent from the tissue from which they originate.

The origin of pleural leiomyomas are smooth muscles which are located in the wall of small blood vessels. It is known that straight muscles, bony skeleton, and other structures located on the chest wall also have blood vessels, arteries, and veins, which have smooth muscles in their vessel wall. The origins of chest wall leiomyomas are smooth muscles from those feeding or draining vessels. Therefore, the determination of the exact origin of a leiomyoma in the thoracic wall is quite difficult. It can either originate from the subpleural connective tissue layer of the parietal pleura or from straight muscles or other blood-stained tissues of the chest wall. Parietal pleura is also accepted as a part of the chest wall, and thus, in our opinion, the term "chest wall leiomyoma" can be used to comprise both groups. The leiomyomas originating from the subpleural connective tissue located under the parietal pleura are, in fact, leiomyomas of the chest wall.

In the English literature, there have been 12 cases reported. Including the current case, a total number of 13 cases are summarized in Table -1. Of these 13 cases, nine (69.2%) were female and four (30.8%) were male, with the mean age of 38.8 (range: 21-56) years. Four patients (30.7%) were asymptomatic. The most common symptom was chest pain (n=7; 53.9%) followed by backache (n=2; 15.4%). Tumor origins were described as subpleural connective tissue layer in six cases (46.2%) and chest wall structures in seven cases (53.8%).

Both groups had similar radiological findings. Both those tumors, whether originating from the subpleural connective tissue layer of the parietal pleura or from straight muscles or

Table 1. Chest wall or pleural leiomyoma cases, published in the literature.

Author	Year	Age	Gender	Symptom	Origin	Surgery	Survive
Moran et al. [10]	1995	23	Female	Asymptomatic	Pleura	Incompletely resected, surgery type not mentioned	6 months without (w/o) recurrence
Moran et al.	1995	21	Female	Asymptomatic	Pleura	Incompletely resected, surgery type not mentioned	4 months w/o recurrence
Proca et al.	2000	32	Male	Asymptomatic	Pleura	Thoracotomy, chest wall resection	12 months w/o recurrence
Nose et al.	2006	55	Female	Asymptomatic	5th intercostal space	Video assisted thoracic surgery	26 months w/o recurrence
Turhan et al.	2007	50	Female	Chest pain	Pleura	Thoracotomy	53 months w/o recurrence
Ziyade et al.	2009	33	Female	Chest pain and heartburn	2nd rib	Thoracotomy	14 months w/o recurrence
Rodriquez et al.	2009	48	Female	Pleuritic pain and dyspnea	Pleura	Thoracotomy	18 months w/o recurrence
Tuncer et al.	2011	38	Female	Backache	6th rib	Thoracotomy	16 months w/o recurrence
Qiu et al.	2011	45	Male	Chest pain	Pleura	Thoracotomy	15 months w/o recurrence
Nakada et al.	2013	28	Female	Chest pain	5th intercostal space	Thoracotomy, chest wall resection and reconstruction	2 months w/o recurrence
Kanlioglu et al.	2014	32	Female	Chest pain	Chest wall (2 separate masses)	1) Thoracotomy 2) Re-thoracotomy, chest wall resection and reconstruction	1) Recurrence after 12 months follow-up after first operation 2) 45 months follow-up w/o recurrence
Kanlioglu et al.	2014	43	Male	Chest pain	7th rib	Thoracotomy, chest wall resection and reconstruction	40 months follow-up w/o recurrence
Yazicioglu et al.	2016	56	Male	Backache	5th intercostal space	Muscle-sparing, mini-thoracotomy	115 months follow-up w/o recurrence

other blood stained tissues of the chest wall, will extend to the location where they are subjected to minimal tissue resistance. As the intrathoracic space provides the least tissue resistance, the tumor growth into the thoracic space is normal for all types of chest wall leiomyomas. In the literature, only two cases reported tumor growth into the thoracic wall. Kanlioglu et al. mentioned a chest wall leiomyoma case who had a mass which destroyed the seventh rib [3]. Nakada et al. presented a case report in which a leiomyoma originated from the chest wall and grew into the chest wall [4]. Other case presentations had radiological findings similar to our case and all of them, regardless of origin reported a mass growth into the intrathoracic space.

On the other hand, benign tumors of the pleura or chest wall cannot be differentiated by radiological methods and the final diagnosis can only be established by histological examination. Complete surgical excision is recommended not only for establishing the diagnosis, but also to relieve and prevent symptoms and to eliminate the possibility of degeneration into a malignant tumor [5,6]. The preferred resection type was mentioned in 11 case reports, and thoracotomy was the type of approach in the majority of cases (n = 10; 90.9%). Only Nose et al. completely resected the tumor via minimally invasive surgery [7]. Chest wall resection is usually not required, but was added to surgery in four patients (36.4%). Kanlioglu et al. presented two cases, both of them requiring chest wall resection and reconstruction [3]. Additionally, in the case reports of both Nakada et al. and Proca et al., the authors completely resected the tumor with chest wall resection [4,8].

The prognosis is fairly good for patients in whom tumors have been completely resected. The recurrence of chest wall leiomyoma is uncommon. Only Kanlioglu et al. mentioned a case that had recurrence of the tumor at 12 months follow-up following the first operation [3]. The authors performed a second operation that included chest wall resection and reconstruction. They reported, disease-free survival of 45 months without regional or systemic recurrence in a published case study. Our case had the longest disease-free survival rate of 115 months, followed by Turhan et al.'s case with survival of 53 months [9].

None of the cases reviewed in the literature reported malignant degeneration. The chest wall consists of many layers that extend from the parietal pleura to skin. We consider that it is too difficult to determine the exact origin of a leiomyoma that originates from the subparietal pleura and grows into the chest wall or that of a tumor which originates from other layers of the chest wall and which grows into the subpleura. We also consider that a determination of the exact origin of the leiomyoma would not make any useful contribution to the diagnosis, treatment, and surgical approach of the case. Thus, these tumors called ''chest wall leiomyoma'' are generally benign, and are resected completely. We recommend complete resection to cure and long-term follow-up for all cases.

Competing interests

The authors declare that they have no competing interests.

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