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Primary Pleural Leiomuscular Tumor of Uncertain Malignant Potential: The Twenty-Third Reported Case

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ABSTRACT

We report the case of a 64-year-old woman with a tumor mass adherent to the right visceral pleura. The histological findings and immunohistochemical pattern are consistent with the diagnosis of a Leiomuscular Tumor of Uncertain Malignant Potential (UMP). To the best of our knowledge, this case represents the twenty-third reported primary pleural leiomuscular tumor in the literature.

Keywords: *Primary pleural leiomuscular tumor, Leiomuscular tumor of uncertain malignant potential, Pleural spindle cell neoplasm, Immunohistochemistry, Differential diagnosis, Case report*

Introduction

Primary leiomuscular neoplasms of the pleura are an extremely rare event and pose challenging problems of differential diagnosis with other more frequent neoplasms of that district. For these reasons, we believe it is appropriate to report a case of our own observation.

Case Report

A 64-year-old woman, asymptomatic. A routine X-ray revealed a mass in the right pleural cavity. A subsequent CT scan showed extensive opacity of the right hemithorax due to a mass adherent to the visceral pleura and occupying a large portion of the pleural cavity. The patient underwent thoracotomy, and a neoplastic mass adherent to the visceral pleura was removed in a piecemeal manner.

Materials and Methods

The surgical specimen consists of some large fragments, the largest of which is approximately 8 cm in diameter, of fibrous consistency, in which some small cystic cavities open (Figure 1).

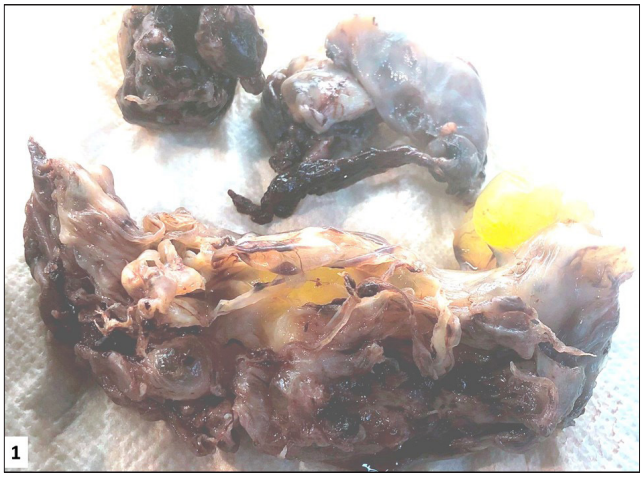

Figure 1. *Macroscopically, the fragment of the surgical specimen has a multilobed appearance and a brownish color. A small cystic formation can also be recognized.*

Numerous fragments of the surgical material are collected, fixed in buffered formalin, and embedded in paraffin. Sections are stained with hematoxylin–eosin. Other sections are subjected to immunohistochemical investigation with a panel of antibodies (Table 1).

Table 1.

VIM	SMACT	DESM	BCL2	CD99	CK AE1-AE3	HBME1	CALR	S100	CD34

VIM = vimentin, SMACT = specific muscle actin, DESM = desmin, CK = cytokeratin, HBME1 = Hector Battifora mesothelin 1

Histology

The tissue in the various fragments examined shows a uniform structure. It consists of a proliferation of spindle-shaped elements (Figures 2a–b) with a lanceolate appearance and acidophilic cytoplasm (Figures 2c, 3a). The nucleus of these elements is voluminous, hyperchromatic, and sometimes atypical. The tissue is affected by extensive edematous imbibition, sometimes giving rise to microcystic formations (Figure 3b). Sporadic mitotic activity is observed (Figure 3c, arrow). The intervening stroma is very scarce, as is the vascularization.

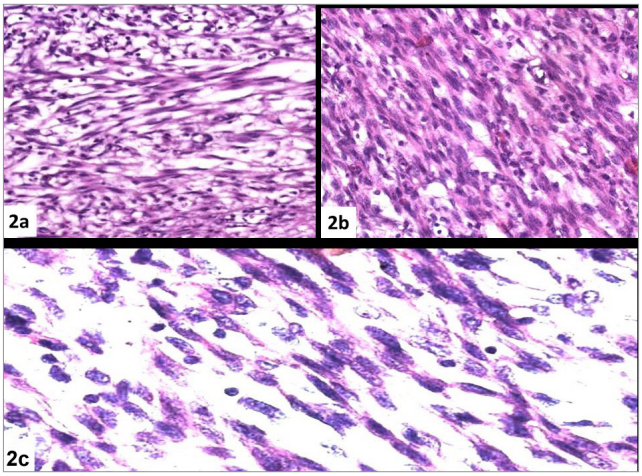

Figure 2. *(a–c) Spindle-shaped elements with hyperchromatic and voluminous nuclei. Note the lanceolate appearance of the elements and the scarcity of stroma. H.E., 150×.*

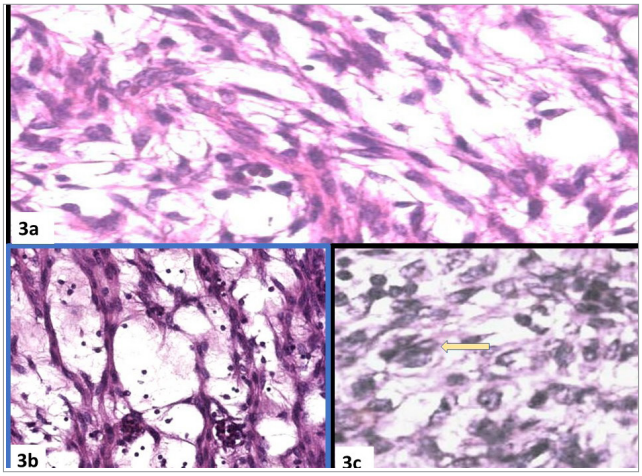

Figure 3. *(a) Evident dissociation of the elements due to edematous imbibition; (b) microcystic degeneration; (c) an atypical mitosis (yellow arrow). H.E., 150×.*

Immunohistochemistry

The results of immunohistochemical investigations are reported in Table 2.

Table 2.

VIM	SMACT	DESM	Bcl2	CD99	CK AE1-AE3	HBME1	CALR	S100	CD34
+++	+++	+++	-+	-+	-	-	-	-	-
Figure 4a	Figure 4(b-c)	Figure 4(d-e)							

VIM = vimentin, SMACT = specific muscle actin, DESM = desmin, CK = cytokeratin, HBME1 = Hector Battifora mesothelin 1

They demonstrate the mesenchymal nature of the proliferation (VIM +++) and its differentiation in a leiomuscular direction (SMACT +++, DESM +++).

Diagnostic Processing

A spindle cell pleural neoplasm first raises the question of whether it is a primary or secondary neoplasm. In this case, no other neoplastic location was reported. The architectural pattern is essentially characterized by a monotonous proliferation of spindle-shaped elements tending to arrange in a fascicled fashion, with little intervening stroma. The elements are tapered, with a pointed end and acidophilic, finely fibrillar cytoplasm, with a large and hyperchromatic nucleus, sometimes atypical. Mitoses are rare.

From a differential diagnostic point of view, also taking into account the location of the lesion, the possibility of Solitary Fibrous Tumor (SFT), Leiomyosarcoma (LMS), Monophasic Synovial Sarcoma (MSS), Mesothelioma (Mes), and Schwannoma (Schw) must be considered. The morphology, although providing some consistent suggestions, does not provide conclusive elements.

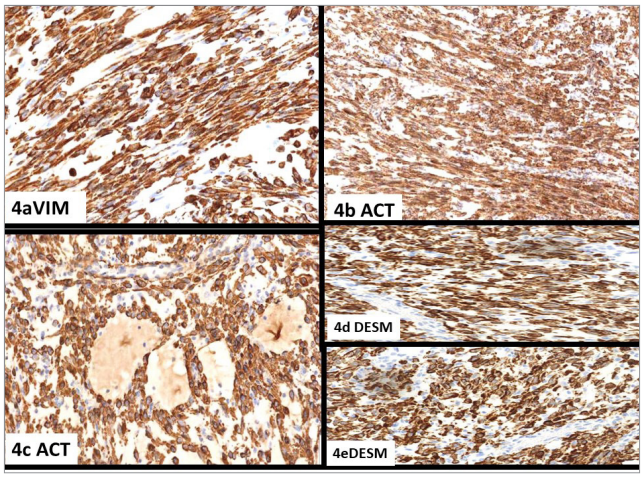

Figure 4. *Immunohistochemistry: (a) vimentin; (b–c) specific muscle actin; (d–e) desmin. 150×.*

Comparing the data from the immunohistochemical investigation (Table 2) with those reported in Table 3.

Table 3.

NEOPLASMS	Stat 6	CD 34	CD99	Bcl2	C K	Vim	Desm	Smact	Calr	HBME1	S100
SFT	+++	+++	+	+	-	+	-	- + foc	-	-	-
LMT	-	-	-	-	-	+	+++	+++	-	-	-
Mon. Syn S	-	-	++	++	++	+	-	-	-	-	-
Mesot	-	-	-	-	+	-	-	-	+++	+++	
SCHW	-	-	-	-	-	+	-	-	-+	-	+++

SFT = Solitary Fibrous Tumor, LMT = Leiomuscular Tumors, Mon. Sin. S. = Monophasic Synovial Sarcoma, Mesot. = Mesothelioma, SCHW = Schwannoma.
Note: These immunohistochemical profiles are taken from Pathology Outlines.

Relating to the immunohistochemical patterns of the neoplasms placed in differential diagnosis, the opinion to classify the neoplasm as of leiomuscular origin is consistent. The mild nuclear atypia and scarce mitotic activity suggest a Leiomuscular Tumor of UMP (Uncertain Malignant Potential). The immunophenotypic profile deserves comment. Alongside the widespread and intense expression of muscle antibodies, which gives unquestionable significance to the tumor cells, the expression, albeit modest, of BCL2 and CD99 does not fit into the immunophenotypic pattern of a muscle neoplasia. In the two cases of literature in which these two antibodies were tested, they were always negative [2–3]. In the given context, the expressiveness of these two antibodies at present finds no satisfactory explanation, if not that of an aberrant expression for technical reasons.

Discussion

The first report of primary leiomuscular malignant neoplasms of the pleura dates back to the seminal article by Moran et al. in 1995 [1]. The study involves five cases, three women and two men, aged between 21 and 69 years. In 3/5 cases, the finding was accidental because the patients were completely asymptomatic; the other two presented right empyema and right chest pain, respectively.

A subsequent report (2016) retrieves from the literature 18 cases of primary leiomuscular tumors of the pleura, including those previously reported by Moran. In total, 6 males and 12 females, between the ages of 28 and 73 years, were described. Eight were completely asymptomatic, 9 had chest pain, and 1 had empyema. Eight cases were classified as leiomyomas, 6 as UMPs, and 4 as leiomyosarcomas. In only one case, diagnosed as leiomyoma, was a recurrence reported after one year. In all other cases, in the reported follow-up, the outcome was favorable regardless of the histopathological diagnosis [2].

After 2016, four further cases of primary pleural leiomuscular tumors have been reported in the literature. The first concerns a leiomyoma in a 40-year-old woman [3], the second a leiomyosarcoma in a 40-year-old woman [4], the third a leiomyoma in a 72-year-old man [5], and the fourth a leiomyosarcoma in a 51-year-old man [6].

Conclusion

In conclusion, this case represents the twenty-third report of this rare localization of leiomuscular neoplasia. From the literature review, it emerges that the prognosis, substantially favorable, is not correlated with the histopathological picture. In the case in question, an unexpected expression of some antibodies was recorded, which finds no confirmation in the current literature.

Conflicts of Interest

The authors declare no conflict of interest and received no specific funding for this work.

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