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Two Decades Of Dormancy: Pleural Metastasis From Extremity Synovial Sarcoma

R. Laamim, I. Achour, M. Beaouiss, A. Rafik, H. Asri, A. Zegmout, H. Souhi, H. El Ouazzani, I.A. Rhorfi

Department Of Pneumology, Mohammed V Military Training Hospital, University Hospital Center Of Rabat (CHU Rabat), Rabat, Morocco

Abstract:
- *Synovial sarcoma is a rare malignant soft tissue tumor that most commonly arises in the extremities and predominantly metastasizes to the lungs. Pleural involvement is exceptionally uncommon, and true pleural metastasis from an extremity synovial sarcoma is extremely rare.*
- *We report a 54-year-old man previously treated in 2004 for synovial sarcoma of the left leg with neoadjuvant chemotherapy followed by limb amputation. After a disease-free interval of nearly twenty years, he presented with persistent left basithoracic pain. Chest CT revealed a necrotic pleural mass with pleural effusion, and biopsy confirmed metastatic synovial sarcoma. The course was rapidly complicated by massive hemoptysis leading to death.*
- *This case highlights the possibility of very late pleural metastatic relapse, underscoring the need for long-term surveillance.*

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I. Introduction:

- Synovial sarcoma is a rare malignant soft tissue tumor accounting for approximately 5–10% of soft tissue sarcomas, most commonly arising in the deep soft tissues of the extremities. It is characterized by a marked propensity for distant metastasis, which occurs in nearly half of patients and predominantly involves the lungs.
- Thoracic involvement is therefore well recognized; however, pleural localization remains extremely uncommon and is most often related to the extension of pulmonary metastases or to primary pleuropulmonary synovial sarcoma rather than true metastatic disease.
- In addition, synovial sarcoma displays an unpredictable clinical course, with the possibility of metastatic relapse occurring after prolonged disease-free intervals.
- In this context, we report a case of isolated pleural metastasis occurring twenty years after treatment of a primary extremity synovial sarcoma, highlighting both the rarity of this metastatic site and the potential for very late recurrence.

II. Case Report:

- A 54-year-old man with a history of type 1 diabetes mellitus and Behçet's disease was referred to our department for evaluation of persistent left basithoracic pain for more than one year. He had previously been treated in 2004 for a synovial sarcoma of the left leg, managed with neoadjuvant chemotherapy followed by limb amputation, and had remained disease-free since then.
- At presentation, the patient was in good general condition without constitutional symptoms apart from persistent thoracic pain. Physical examination revealed a left lower limb amputation and findings suggestive of a left-sided pleural effusion.
- A chest CT performed eight months earlier had revealed an 11 × 8 mm left lower-lobe nodule, which on follow-up had progressed to a necrotic left mediobasal pleural mass measuring 54 × 37 mm, associated with moderate pleural effusion.

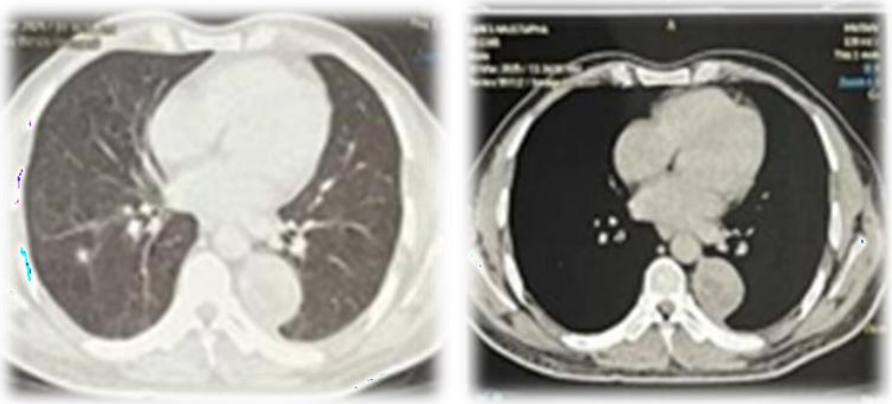

Figure 1: Axial chest computed tomography (CT) images
CT scan showing a necrotic left mediobasal pleural mass measuring 54 × 37 mm

- The patient underwent 18F-FDG PET-CT, which showed intense uptake of the pleural mass (SUVmax = 13) without other hypermetabolic lesions.
- A pleural biopsy was subsequently performed. Histopathological examination revealed a tumor proliferation composed of epithelial and spindle cell components. Immunohistochemical analysis demonstrated strong nuclear expression of TLE1 and positivity for cytokeratin AE1/AE3, findings consistent with pleural involvement by synovial sarcoma in a patient with a known oncologic history.

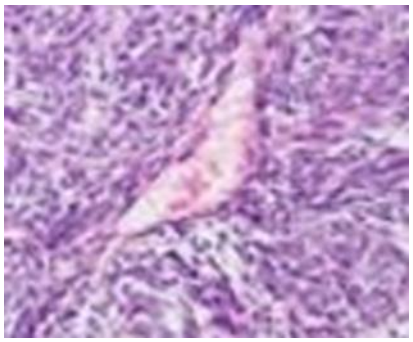

Figure 2: Histopathological examination showing biphasic synovial sarcoma in the pleura

- Following the histological diagnosis, the patient was referred for multidisciplinary evaluation to determine the most appropriate therapeutic strategy. However, the clinical course was abruptly complicated by massive hemoptysis, leading to rapid respiratory and hemodynamic collapse. Despite immediate resuscitative measures, the patient died shortly thereafte

III. Discussion:

- Pleural metastasis from extremity synovial sarcoma is exceptionally rare. Although the lungs represent the predominant metastatic site (70–80%), pleural involvement is usually related to pulmonary extension or primary pleuropulmonary synovial sarcoma [1–4]. To our knowledge, only one case of true pleural metastasis from an extremity synovial sarcoma has been reported, described by Iwata et al. [5].
- Jiang et al. reported a pleural synovial sarcoma nine years after resection of a plantar synovial sarcoma, suggesting a second primary pleuropulmonary tumor rather than metastasis [6]. In contrast, the present case strongly supports metastatic spread based on the patient's oncologic history and histopathological findings consistent with the initial tumor.
- Synovial sarcoma may recur late. Previous studies have reported a mean interval of 5.7 years (range 0.5–16.3) between diagnosis and metastasis, with 24% of deaths occurring after 10 years [7]. In our patient, pleural metastasis occurred twenty years after treatment, representing an exceptionally long latency period, which may reflect tumor dormancy and delayed reactivation of residual malignant cells.
- Diagnosis relies on imaging, histopathology, and molecular analysis. CT typically shows a pleural-based mass, often heterogeneous or necrotic and associated with pleural effusion. Histologically, the tumor presents monophasic or biphasic proliferation with strong TLE1 and epithelial marker expression. Molecular confirmation is provided by the characteristic t(X;18)(p11;q11) translocation, resulting in the SS18-SSX fusion gene [8].

- Management of metastatic synovial sarcoma relies on a multidisciplinary approach, including surgical resection of isolated metastases when feasible and systemic chemotherapy, most commonly based on doxorubicin and ifosfamide [3]. Prognosis depends on several factors such as tumor size, histological grade, disease-free interval, number of metastases, and completeness of surgical resection [3,7]. Despite treatment, metastatic disease remains associated with poor outcomes, with 5-year survival rates ranging from 20% to 50% [7]. In our patient, the clinical course was rapidly complicated by massive hemoptysis, leading to a fatal outcome before treatment could be initiated.
- Overall, this case highlights the extreme rarity of pleural metastasis from extremity synovial sarcoma and underscores the importance of long-term surveillance, as metastatic relapse may occur even decades after initial treatment.

IV. Conclusion:

- This report illustrates the exceptional occurrence of pleural metastasis from an extremity synovial sarcoma twenty years after initial treatment, highlighting the remarkable capacity of this tumor for very late relapse. Such an observation emphasizes the unpredictable biological behavior of synovial sarcoma and underlines the importance of long-term follow-up, even in patients who have remained disease-free for many years.

References:

[1]. Thway K, Fisher C. Synovial Sarcoma: Defining Features And Diagnostic Evolution. Ann Diagn Pathol. 2014;18(6):369-380.
[2]. Kransdorf MJ. Malignant Soft-Tissue Tumors In A Large Referral Population: Distribution Of Diagnoses By Age, Sex, And Location. AJR Am J Roentgenol. 1995;164(1):129-134.
[3]. Lewis JJ, Antonescu CR, Leung DH, Blumberg D, Healey JH, Woodruff JM, Et Al. Synovial Sarcoma: A Multivariate Analysis Of Prognostic Factors In 112 Patients With Primary Localized Tumors. Ann Surg. 2000;232(4):586-595.
[4]. Essary LR, Vargas SO, Fletcher CD. Primary Pleuropulmonary Synovial Sarcoma: Reappraisal Of A Series Of 25 Cases. Am J Surg Pathol. 2002;26(4):409-417.
[5]. Iwata T, Nishiyama N, Nagano K, Izumi N, Tsukioka T, Suehiro S. Metastatic Monophasic Synovial Sarcoma Of The Pleura. Ann Thorac Cardiovasc Surg. 2007;13(4):258-261.
[6]. Jiang M, Zhang J, Cheng D. A Second Primary Synovial Sarcoma Of Pleura Developed Nine Years After The First Synovial Sarcoma Of Plantar Pedis: A Case Report And Review Of Literature. Int J Clin Exp Pathol. 2019;12(7):2743-2748.
[7]. Bergh P, Meis-Kindblom JM, Gherlinzoni F, Berlin O, Bacchini P, Bertoni F, Et Al. Synovial Sarcoma: Identification Of Low And High-Risk Groups. Ann Oncol. 1999;10(9):1149-1156.
[8]. Kawai A, Woodruff J, Healey JH, Brennan MF, Antonescu CR, Ladanyi M. SYT-SSX Gene Fusion As A Determinant Of Morphology And Prognosis In Synovial Sarcoma. N Engl J Med. 1998;338(3):153-160.