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Letter to the Editor

Sarcoid-Like Mediastinal Reaction in the Context of Histiocytic Sarcoma

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Dear Editor,

The link between sarcoidosis and sarcoma is both infrequent and scarcely documented in the literature.[1] Histiocytic sarcoma (HS) is still a rare entity whose differential diagnosis includes histiocytic/dendritic tumors, myeloid, or lymphoproliferative neoplasm, melanoma, or carcinoma[2]

A 47-year-old male, without any remarkable personal records, consulted a fourth proximal right-hand phalanx and gummy lump. Ultrasound (US) revealed a 4-5 cm solid lump, hypervascular, and not fixed to deep layers without periosteum invasion. Magnetic resonance imaging (MRI), T2, T1, and short tau inversion recovery (STIR) showed peripheral enhancement, suggesting a mesenchymal origin. After extirpation, the pathology study was compatible with HS (atypical mesenchymal hystiocitary cell proliferation).

Therefore, an extension study was performed using computerized tomography (CT), gammagraphy, and positron emission tomography–CT. Computerized tomography showed multilevel mediastinal adenopathies in 5 (SUVmax 4.8), 4R (SUVmax 14.9), 7 (SUVmax 13.6), 11R (SUVmax 13.2), and 11L (SUVmax 13.1) territories. Endobronchial US-guided transbronchial needle aspiration (EBUS-TBNA) revealed multilevel mediastinal adenopathies suggestive of malignancy (hypoechoic round, marked margins, no hilar core) in 7 and 11R territories. Samples were taken with an ECHO-HD-22-EBU S-P-C® using 3-needle punctures. The results demonstrated both lymphoid tissue and histiocytic non-necrotizing granulomatous areas, which tested negative for mycobacteria in both the Ziehl–Neelsen stain and Löwenstein–Jensen culture.

In addition to EBUS-TBNA, further videomediastinoscopy (VM) by the Carlens technique and biopsy of 2R and 4R territories were obtained, showing non-necrotizing granulomas containing epithelioid and giant multinucleated cells. Based on these findings, a sarcoid-like reaction (SLR) was diagnosed. No relapse was observed after 1 year of follow-up with periodic CT and MRI.

Sarcoid neoplasms may be associated with other malignancies in up to 2.5%. However, the link between sarcomas and sarcoidosis remains unclear. Sarcoidosis is a well-known risk factor for skin, hematologic, digestive tract, and renal or liver malignancies.[3] Some authors have reported a lower-risk link between mediastinal non-caseating granulomas (NG) and locoregional relapse in patients with lung cancer.[4]

Unfortunately, in patients already diagnosed with both sarcoidosis and cancer, the discovery of NG may lead to a mistaken belief in sarcoidosis relapse when an SLR (NG in the absence of systemic symptoms)[5] may develop, which may lead to overestimation of tumor stratification and therefore alter correct management.[1]

The correlation between SLR and sarcomas has rarely been reported in the literature.[1] Although the pathogenesis remains obscure, there is evidence that the presence of circulating tumoral antigens and/or chemotherapy drugs may lead to a host immune response.[5] Second, there is a well established occurrence of sarcoidosis as paraneoplastic syndrome. Finally, chronic inflammation status or cancer therapy drugs may modulate the cytokine environment increasing lymphocyte rates which may lead to SLR.

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A mandatory extension study must be performed, including histological sampling for every patient affected by sarcoma. Although EBUS-TBNA allows for a minimally invasive mediastinal approach, identification of malignancy-matched characteristics, and sample collection, VM remains the gold standard.

In the presented case, only after VM, malignancy was ruled out and it prevented from diagnosing a sarcoma metastasis when an SLR was occurring. Despite rare association with sarcomas, SLR must be included in the differential diagnosis of every patient suffering from sarcoma and presenting with lung NG.

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REFERENCES

1. Espejo AP, Ramdial JL, Wilky BA, Kerr DA, Trent JC. A non-random association of sarcoidosis in patients with gastrointestinal stromal tumor and other sarcomas. *Rare Tumors*. 2018; 10:2036361318787626. [CrossRef]
2. Hung YP, Lovitch SB, Qian X. Histiocytic sarcoma: new insights into FNA cytomorphology and molecular characteristics. *Cancer Cytopathol*. 2017;125(8):604-614. [CrossRef]
3. Bonifazi M, Bravi F, Gasparini S, et al. Sarcoidosis and cancer risk: systematic review and meta-analysis of observational studies. *Chest*. 2015;147(3):778-791. [CrossRef]
4. Steinfort DP, Tsui A, Grieve J, Hibbs ML, Anderson GP, Irving LB. Sarcoidal reactions in regional lymph nodes of patients with early stage non-small cell lung cancer predict improved disease-free survival: a pilot case-control study. *Hum Pathol*. 2012;43(3):333-338. [CrossRef]
5. Ravaglia C, Gurioli C, Casoni GL, et al. Sarcoid-like lesion is a frequent benign cause of lymphadenopathy in neoplastic patients. *Eur Respir J*. 2013;41(3):754-755. [CrossRef]