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Short Communication

Primary pleural hybrid cellular schwannoma/perineurioma: A case report

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ABSTRACT

Hybrid schwannoma/perineurioma is a recently characterized benign nerve sheath tumor, most commonly affecting the lower limb and limb girdle. Hybrid tumors located in the subcutis of the trunk have not previously been reported to affect the pleura. We describe a 52-year-old man with dyspnea and thoracic pain due to a large mass in the right pleura, histologically composed of densely packed, S-100-positive spindle cells, intermixed with cells containing slender nuclei positive for epithelial membrane antigen, Glut-1, and claudin-1. To our knowledge, this is the first report of hybrid schwannoma/perineurioma in the pleura.

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1. Introduction

Benign peripheral nerve sheath tumor types include schwannoma, perineurioma, and neurofibroma, derived from Schwann cells, perineurial cells, and a mixture of endoneurial components, respectively [1]. Schwannoma is composed of compact spindle cells with twisted nuclei that are strongly and diffusely positive for S-100 protein and negative for epithelial membrane antigen (EMA), CD34, Glut-1, and claudin-1 [1]. Cellular schwannoma is a highly cellular variant of schwannoma, showing predominantly compact spindle cell proliferation, with few or no Verocay bodies, and is often associated with xanthomatous histiocytes and lymphoid clusters [2,3]. Perineuriomas are far less common than schwannomas and neurofibromas and consist of a proliferation of spindle cells with wavy nuclei that have extremely thin, elongated, and bipolar cytoplasmic processes and a storiform, focally whorled growth pattern. These tumor cells

Abbreviations: EMA, epithelial membrane antigen

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are positive for EMA, Glut-1, CD34, and claudin-1 and are generally negative for S-100 protein [1,4].

In recent years, a number of nerve sheath tumors showing areas of more than one histologic type have been described, and these tumors exhibit an abrupt transition and/or intimate admixture of peripheral nerve cell types. These composite tumors have been termed "hybrid" peripheral nerve sheath tumors. Well-documented examples have been reported that include hybrid schwannoma/neurofibroma, schwannoma/perineurioma, neurofibroma/perineurioma [5–8], and cellular schwannoma/perineurioma [9]. These hybrid tumors usually arise in the dermis and subcutis and have been reported to occur over a wide range of ages and anatomical sites [5]. The pathogenesis of these hybrid tumors remains unknown, but these tumors might be associated with localized microenvironmental changes or clonal genetic alterations in primitive tumor cells [8].

Intrathoracic peripheral nerve sheath tumors are rare and usually occur in the paraspinal region of the mediastinum, where they involve spinal nerve roots and sympathetic ganglia, raising the possibility of a pleural tumor [10]. Herein, we describe the case of a 52-year-old man with a pleural tumor exhibiting morphological and immunohistochemical features of a hybrid cellular schwannoma/perineurioma.

2. Case report

A 52-year-old man presented to the ABC Medical Center in Mexico City with a history of dyspnea, thoracic pain, and sporadic cough for 3 months. There was no clinical evidence of neurofibromatosis or a history of other peripheral nerve sheath tumors. Computed tomography and 2-deoxy-2-[(18)F] fluoro-D-glucose positron emission tomography/computed tomography revealed an $18 \times 11 \times 10\ cm^3$ bilobed mass in the right hemithorax that involved the visceral pleura, with a standard uptake value of 7.8 and extensive pleural effusion with ipsilateral lung collapse (Fig. 1 A–C). The tumor was

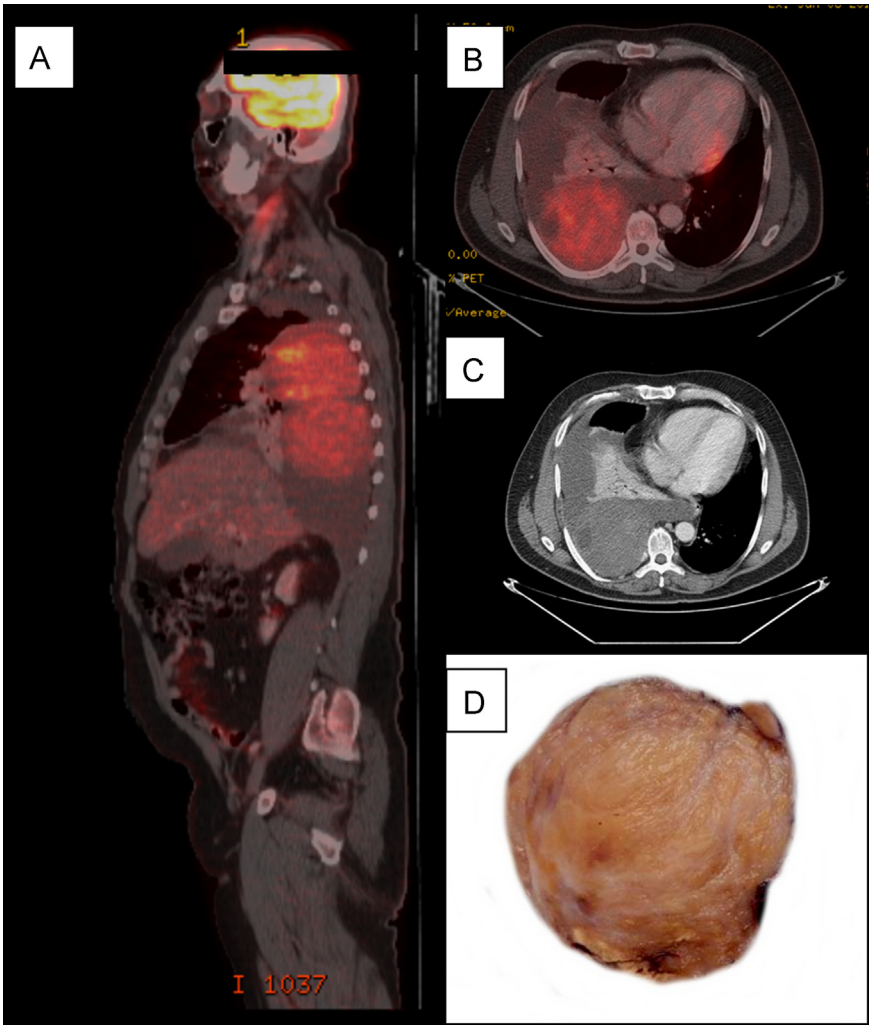

Fig. 1 – (A–C) Computed tomography and 2-deoxy-2-[(18)F]fluoro-D-glucose positron emission tomography/computed tomography revealing a mass located in the right hemithorax that involved the visceral pleura, with a standard uptake value of 7.8, ipsilateral pleural effusion, and lung collapse. (D) On sectioning, the cut surface showed a solid, firm, yellow/white fascicular mass. (For interpretation of the references to color in this figure legend, the reader is referred to the web version of this article.)

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