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Multiple Primary Pulmonary Meningiomas: A Case Report

Authors : Wellemans Isabelle, Remmelink Myriam, Foucart Annick, Rusu Stefan, Compère Christophe

Abstract : Primary pulmonary meningioma (PPM) is a very rare tumor, and its occurrence has been reported only sporadically. Multiple PPMs are even more exceptional, and herein, we report, to the best of our knowledge, the fourth case, focusing on the clinicopathological features of the tumor. Moreover, the possible relationship between the use of progesterone–only contraceptives and the development of these neoplasms will be discussed. Case Report: We report a case of a 51-year-old female presenting three solid pulmonary nodules, with the following localizations: right upper lobe, middle lobe, and left lower lobe, described as incidental findings on computed tomography (CT) during a pre-bariatric surgery check-up. The patient revealed no drinking or smoking history. The physical exam was unremarkable except for the obesity. The lesions ranged in size between 6 and 24 mm and presented as solid nodules with lobulated contours. The largest lesion situated in the middle lobe had mild fluorodeoxyglucose (FDG) uptake on F-18 FDG positron emission tomography (PET)/CT, highly suggestive of primary lung neoplasm. For pathological assessment, video-assisted thoracoscopic middle lobectomy and wedge resection of the right upper nodule was performed. Histological examination revealed relatively well-circumscribed solid proliferation of bland meningothelial cells growing in whorls and lobular nests, presenting intranuclear pseudo-inclusions and psammoma bodies. No signs of anaplasia were observed. The meningothelial cells expressed diffusely Vimentin, focally Progesterone receptors and were negative for epithelial (cytokeratin (CK) AE1/AE3, CK7, CK20, Epithelial Membrane Antigen (EMA)), neuroendocrine markers (Synaptophysin, Chromogranin, CD56) and Estrogenic receptors. The proliferation labelling index Ki-67 was low (<5%). Metastatic meningioma was ruled out by brain and spine magnetic resonance imaging (MRI) scans. The third lesion localized in the left lower lobe was followed-up and resected three years later because of its slow but significant growth (14 mm to 16 mm), alongside two new infra centimetric lesions. Those three lesions showed a morphological and immunohistochemical profile similar to previously resected lesions. The patient was disease-free one year post-last surgery. Discussion: Although PPMs are mostly benign and slow-growing tumors with an excellent prognosis, they do not present specific radiological characteristics, and it is difficult to differentiate it from other lung tumors, histopathologic examination being essential. Aggressive behavior is associated with atypical or anaplastic features (WHO grades II–III) The etiology is still uncertain and different mechanisms have been proposed. A causal connection between sexual hormones and meningothelial proliferation has long been suspected and few studies examining progesterone only contraception and meningioma risk have all suggested an association. In line with this, our patient was treated with Levonorgestrel, a progesterone agonist, intra-uterine device (IUD). Conclusions: PPM, defined by the typical histological and immunohistochemical features of meningioma in the lungs and the absence of central nervous system lesions, is an extremely rare neoplasm, mainly solitary and associating, and indolent growth. Because of the unspecific radiologic findings, it should always be considered in the differential diagnosis of lung neoplasms. Regarding multiple PPM, only three cases are reported in the literature, and this is the first described in a woman treated by a progesterone-only IUD to the best of our knowledge.

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