Case report - Thoracic oncologic

Contralateral recurrence of a malignant solitary fibrous tumor of the pleura

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Abstract

We present an unusual case of a contralateral recurrence of malignant solitary fibrous tumor of the pleura (SFTP) nine years after a complete resection. Recurrence of malignant SFTP has already been described, but is usually localized. In our case the patient underwent surgical resection for a malignant SFTP of the left upper lobe in 2000. Nine years later computed tomography (CT)-scans showed lesions that were suspicious of tumor recurrence in the right lung. Thoracoscopy, wedge-resections and pathological findings revealed four nodules of a malignant SFTP of the right middle and lower lobe, histopathologically identical to the tumor, which had been resected nine years ago. A coincidental mucinous bronchioloalveolar carcinoma of the left lower lobe was resected by thoracotomy. To our knowledge this is the first report of contralateral recurrence of a malignant SFTP years after complete resection in the literature. The possibility of a new primary tumor on the right with local metastasis could not be excluded in the clinical and histopathological examinations. Therefore, contralateral recurrence of malignant SFTP should be considered in the postoperative follow-up even years after complete resection.

Keywords: Solitary fibrous tumor of the pleura; Contralateral recurrence; CD34

1. Case report

Nine years previously a 10 cm pleural tumor was resected completely by thoracotomy in the left thorax in a 55-year-old patient. Immunohistologically the tumor met the criteria for a solitary fibrous tumor of the pleura (SFTP) with increased malignant potential.

Nine years later the patient presented again with cough and shortness of breath. Computed tomography (CT)-scans revealed four sharply delimited tumors in the right thorax (40 mm, 18 mm, 13 mm and 3 mm, respectively) in the pleura of the middle and lower lobe and an opacity (11 mm) in the lingula of the left lung (Fig. 1). We performed a thoracoscopy of the right lung with wedge-resections of the middle and lower lobe. Histopathologically the cellularity of the spindle cell component and the mitotic rate (7–8 per mm²) were higher than in the previously diagnosed SFTP. The epithelial component was positive for CK22 and TTF-1, whereas the stromal component was strongly reactive for CD34 and bcl-2 and CD99 (Fig. 2). The markers CD31, CK22, BER-EP4, EMA, CD117, Actin, CD57, S100, HMB-45 and SMA were negative. The tumors were classified as malignant SFTP.

The tumor in the lingula and a small palpable tumor of the left lower lobe were resected later by thoracotomy due to adhesions. The lesion in the lingula turned out to be a postoperative scar from the previous SFTP-resection. The palpable lesion in the left lower lobe showed an incidental bronchioloalveolar carcinoma (4 mm, mucinous subtype). The rest of the hospital stay was uneventful.

SFTP are rarities with an incidence of 2.8/100,000 and are unrelated to pleural mesothelioma or smoking [1, 2]. The majority of patients present with cough, chest pain or...
dyspnea [1]. Histopathologically SFTP originate from mesenchymal cells in the submesothelial tissue. They are immunohistochemically negative for cytokeratines and positive for CD34, bcl-2 and CD99 [3]. Cell density, necrosis, number of mitotic figures and cell atypia indicate malignancy [4]. From 800 reported cases in the literature 80% of SFTP were benign [1]. Adequate therapy consists of a complete resection, because malignant transformation and metastases of SFTP are unpredictable [5]. SFTP-recurrence occurs within 24–120 months after resection in 2–14% in benign and in 14–63% in malignant variants localized in the area of the former resection due to contamination, incomplete resection and satellite metastasis [1,4]. In the case of local recurrence, re-resection has been strongly recommended as a potentially curative treatment [1].

In this case report we present an unusual case of contralateral recurrence of malignant SFTP nine years after complete resection. To our knowledge this is the first report in the literature.

The malignant potential of the first SFTP was indicated by high mitotic rate and focal necrosis. Due to the known higher recurrence rate in malignant variants of SFTP we initially performed a complete resection. Although a local recurrence of SFTP 17 years after complete resection has been described [6], the contralateral recurrence of SFTP nine years after resection is unusual. The genesis of the contralateral recurrence of the malignant SFTP in this case stays unclear, although the time between primary resection and recurrence make the hypothesis of a metastatic event improbable. The immunohistological findings could not clearly differentiate between a second primary and a contralateral metastasis of the previous malignant SFTP, but both possibilities would be extraordinary.

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References