Clinical note

Diagnostic pitfalls in the preoperative $^{18}$F-FDG PET/CT evaluation of a case of giant malignant solitary fibrous tumor of the pleura

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Solitary fibrous tumor of the pleura (SFTP) is an uncommon entity, generally with an indolent behavior. Nevertheless, some malignant forms have been rarely reported. These, often have an aggressive biological behavior with pathological findings of invasiveness. The preoperative diagnosis and evaluation of the grade of malignancy are extremely challenging. Herein we report a case of a 64-year-old man who presented with a left giant intra-thoracic mass imaged with fluorine-$^{18}$ fluorodeoxyglucose positron emission tomography/computed tomography ($^{18}$F-FDG/PET-CT) and sampled via fine-needle aspiration biopsy (FNAB). Imaging and FNAB findings showed suspicion of a benign form of SFTP. Surgical radical resection of the giant mass was performed. The definitive histological diagnosis showed a malignant SFTP. Based on this report, we take the opportunity to briefly discuss the insidious pitfalls concerning the radiological and $^{18}$F-FDG/PET-CT features as well as cyto/histological findings in the pre-operative diagnostic work-up examination of this rare entity.

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Dificultades diagnósticas en la evaluación preoperatoria con $^{18}$F-FDG PET/TC en un caso de tumor fibroso solitario maligno gigante de la pleura

R E S U M E N

El tumor fibroso solitario de la pleura (TFSP) es una entidad poco frecuente, en general con un comportamiento indolente. Sin embargo, algunas formas malignas rara vez han sido publicadas, presentando a menudo un comportamiento biológico agresivo con hallazgos patológicos de invasión. El diagnóstico preoperatorio y la evaluación del grado de malignidad es extremadamente difícil. Presentamos el caso de un paciente de 64 años de edad con una masa intratorácica gigante. Se realizó TC, $^{18}$F-FDG/PET-TC y biopsia por aspiración con aguja fina. Los hallazgos de imagen y de la biopsia hacían sospechar de una forma benigna de TFSP. Se realizó la resección quirúrgica radical de la masa gigante. El diagnóstico histológico definitivo mostró el TFSP maligno. Aprovechamos la oportunidad de este caso para revisar los aspectos relativos a los estudios radiológicos, características de la $^{18}$F-FDG/PET-CT y los hallazgos cito-histológicos en la evaluación preoperatoria de esta rara entidad.

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Introduction

Solitary fibrous tumor of the pleura (SFTP) is a rare neoplasm that accounts for less than 5% of all pleural tumors. $^1$ Although the majority of these neoplasms are benign, about 10–20% of the cases reported in literature are malignant. $^2$–$^5$ The distinction between benign and malignant SFTPs based on specific radiological signs is usually very difficult and, similarly, percutaneous transthoracic fine-needle aspiration biopsy (FNAB) of the mass rarely provides enough tissue to give a definitive diagnosis $^6$ and to establish the exact degree of malignancy. Although fluorine-$^{18}$ fluorodeoxyglucose positron emission tomography ($^{18}$F-FDG/PET-CT) has been shown in very small cohorts of patients to provide some differential diagnosis elements to distinguish between benign and malignant SFTP, $^6$ the efficacy of this diagnostic tool is very controversial. Herein we report a very challenging case of a 64-year-old man who presented with a left giant malignant SFTP and discuss on the controversial pre-operative diagnostic imaging.

Clinical case

A 64-year-old man, with a previous unremarkable history, presented with an increasing dyspnea, persisting cough and a moderate weight loss. A chest X-ray and a CT-scan showed a giant intra-thoracic mass partially occupying the left hemithorax. In detail, the density of the neoplasm (measuring about $18 \text{ cm} \times 17 \text{ cm} \times 10 \text{ cm}$) was quite low (40 U.H.) and very

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inhomogeneous because of different areas with various patterns of enhancement (Fig. 1A and B); in addition no clear signs of local invasiveness (invasion of the chest wall or ipsilateral diaphragm) were detected, apart from a small bilateral pleural effusion (proved to be negative for malignant cells after thoracentesis). After a multidisciplinary discussion of the case, we planned a 18F-FDG/PET-CT scan. Image acquisition for the whole-body scans started 1 h after the intravenous administration of FDG (3.7 MBq/kg body weight). At FDG administration, the plasma glucose level of the patients was 96 mg/dL. The CT-scans from the brain to the pelvis were performed immediately prior to the PET-scan with a multi-detector spiral CT-scanner (3.75 mm slice thickness). The attenuation-corrected PET, CT and fused PET/CT images were available for review in axial, coronal and sagittal planes, as was a cine display of maximum intensity projection of the PET data. FDG-uptake was considered to be abnormal on visual inspection when it was distinguishable from the background uptake in the mediastinum. The standard uptake value (SUV) was calculated using the commercially available software provided by the manufacturer and embedded into the imaging machinery.

The 18F-FDG/PET-CT showed a very inhomogeneous pattern of the mass (Fig. 1C) characterized by the simultaneous presence of areas with no radiopharmaceutical uptake and others with only a mild tracer uptake (SUVmax = 2.2). No areas of increased 18F-FDG uptake were found in correspondence of the pleural effusion nor in other sites. These imaging findings were strongly indicative for a benign (or “low-grade”) intra-thoracic tumor.

Moreover, we carried out a transthoracic FNAB that revealed bland appearing spindle cells, suggesting the diagnosis of a benign pleural fibrous tumor (Fig. 1D).

Due to the relevant symptoms, a surgical excision of the mass was performed via right lateral thoracotomy. At surgery, the tumor originated from the visceral pleura of the left lower pulmonary lobe, being the vascularization guaranteed by a large pedicle located in pulmonary ligament. No signs of local invasiveness were found but a pulmonary wedge resection was necessary together with the mass excision in order to achieve a radical “en bloc” resection (Fig. 2A).

Microscopically the tumor showed solid spindle and oval cells arranged in random arrays (a “patternless pattern”), with relevant variation in cellularity from region to region within the neoplasm (Fig. 2B). A moderate mitotic activity was present (5 mitoses out of 10 HPF) and the tumor demonstrated reactivity for CD34 (Fig. 2C) and bcl2 (Fig. 2D). According with the England’s criteria, the final morphological and immunohistochemical diagnosis was indicative for a huge aggressive SFTP.

The postoperative course was uneventful and the patient was discharged in 7th postoperative day.

Although no adjuvant radiotherapy was administered due to R0 resection, a very strict follow-up program including 6-monthly CT-scan and physical examination was planned. At the moment of writing this report (9 months after surgery), the patient is alive with no signs of tumor recurrence.

**Discussion**

Although SFTPs may be still considered as uncommon neoplasms, their incidence is increasing in the last decade, substantially due to a better knowledge of the pathological features and a more accurate terminology of such rare entity. In fact, while only 800 cases were described from 1931 to 2002, a total of 960 further cases have been subsequently reported from 2002 to 2012. Although the majority of these neoplasms are benign, about 10–20% show a more aggressive biological behavior, with pathological findings of invasiveness, appearance of distant metastases and local relapse after resection.

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Although the chest CT scanning remains the test of choice, a firm differential diagnosis of malignancy based on specific radiological signs is usually impossible albeit some radiological features (large size and central necrosis) are more commonly associated with malignant forms. In this scenario, 18F-FDG/PET-CT has been advocated as a potentially useful functional imaging method.
for distinguishing malignant from benign SFTPs, although this assumption is still based on few single cases or small – seldom contradictory – retrospective series.8

Reporting this challenging case, we would highlight the insidious pitfalls of pre-operative diagnostic work-up in the differential diagnosis between benign and malignant SFTP.

In the present case, the pre-operative imaging (CT-scan and 18F-FDG/PET-CT-scan) showed a very inhomogeneous pattern with no clear signs of malignancy. Similarly, the cytological findings were bland. Although the results of both examinations failed to be in accordance with the final diagnosis, this fact may be easily explained with the specific characteristic of the tumor itself. This giant mass, similarly with others reported in literature,9 consisted of different areas with different pathological characteristics (necrotic/fibrosis tissue, hemorrhagic/ischemic zones and areas with high/low cellularity). Considering that the 18F-FDG/PET-CT findings are influenced by the metabolic activity of the mass (this being directly related with the proliferation-index and the malignancy grade of the tumor), the inhomogeneous metabolic pattern reflects substantially the pathological pattern of the tumor.

Similarly, the transthoracic FNAB per se may be useful in the differential diagnosis between SFTP and other neoplasms but has some strong limitations in the definition of the grade of malignancy because the biopsy is limited to a portion of the mass.

In fact, as reported by Cardillo et al.,1 the pre-operative cytological assessment of malignant SFTPs is invariably unsatisfactory since the accuracy of the CT-guided FNAB ranges in levels below 50%.

The case described would suggest that 18F-FDG/PET-CT could have only a limited role in evaluating SFTP. For example, whenever technically feasible, this functional imaging method could serve as a “guide” for a FNAB in large SFTPs, because 18F-FDG/PET-CT is able to detect the areas with the highest metabolic activity into a large tumor with inhomogeneous density. Nevertheless, it should be taken into account that the achievement of a pre-operative accurate pathological diagnosis often does not change the decision making process because the compression of the mass to the neighboring structures (and the symptoms related to it) represents per se a clear indication for a surgical excision. Therefore, in our opinion, a FNAB, although useful in differential diagnosis, should be reserved preferentially to those cases where the clinical condition contra-indicates surgery. Based on our case and literature data,3,7,10 the role of 18F-FDG/PET-CT in the definition of the malignancy grade of SFTPs should be further investigated in large clinical series before recommend its use in the differential diagnosis between malignant and benign SFTPs.

Conflicts of interest

The authors have no conflicts of interest to declare.

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