Localized benign pleural mesothelioma: a case report

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Primary neoplasms of the pleura are rare tumors and the majority are generally mesotheliomas. Mesotheliomas are either localized and mostly benign, or diffuse and uniformly malignant neoplasms. Localised benign pulmonary mesothelioma (solitary fibrous tumor of the pleura) are originally thought to be a variant of diffuse pleural mesothelioma because they consists of a spindle cell stroma associated with branching tubular structures lined by cuboidal cells. Our case which is reported below shows the clinical spectrum of the more common benign variant. Clinical differential diagnosis of benign and malign mesotheliomas is not clear. Complete surgical resection is the preferred treatment for both types and usually curative with the benign mesothelioma.

The localised pleural variant is benign in most cases, and it is even less common, constituting only 10% of all mesotheliomas [1]. The importance of localised benign mesothelioma is that it is almost impossible to differentiate from a malignant neoplasm preoperatively and it may occasionally recur, sometimes with a malignant change. Monaldi Arch Chest Dis 2003; 59: 2, 166-168

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Case report

A 44-year old builder and former smoker was seen in the chest policlinic. He was generally in good health, but complaining of chest pains, dyspnea and a cough. In his past history, he was an alcoholic. He had been working as a builder for approximately 25 years. In physical examination, there were no significant findings except for breathing sounds heard less clearly in the basal part of the left lung and digital clubbing was not seen. In the laboratory, haemogram and biochemical results were normal. Sedimentation rate was 2 mm / hr. The chest radiograph showed a 10 cm diameter mass in the left base adjacent to the diaphragm (figure 1). A Computed Tomographic (CT) scan of the chest showed 12.5 x 9 x 10 cm, smooth-contoured mass in the left lower lobe abutting the diaphragm (figure 2). The bronchoscopy was normal. No distant metastases was found.

A left posterolateral thoracotomy was performed, and a solitary, 12 x 15 x 20 cm, firm, pale yellow mass arising on a long, thin pedicle from the diaphragmatic visceral pleura of the left lower lobe was found. The tumor was rich with vessels and excised with a margin of normal lung. The pathological diagnosis was a benign fibrous mesothelioma with no nuclear aberrations or mitotic figures (figure 3).

The patient had an uncomplicated recovery. There was no recurrence reported during his follow up, but he died two years after the operation due to an alcoholic coma.
Localised benign pleural mesotheliomas (solitary fibrous tumours of the pleura) are tumours originating from the mesenchymal tissue underlying the mesothelial layer of the pleura. The differential diagnosis of localised benign mesothelioma include localised malign mesothelioma, soft tissue neoplasm of the chest wall involving the pleural cavity, or solitary metastasis from an occult primary.

Localised benign pleural mesotheliomas are rare tumors. In the largest single hospital series reported, OKIKE et al at the Mayo Clinic found 60 patients with this neoplasm over 25 years, an incidence of 2.8 cases per 100,000 registrations [2]. In general, about one benign pleural mesothelioma will present for every 10 of the malignant neoplasms. The mere presentation of a localised pleural neoplasm, however, does not guarantee it is benign. Meyer reported that there were 15 detected solitary fibrous tumours of the pleura cases between 1981 and June 1998, but two of them showed malign recurrence [3].

The localised benign tumor tends to occur with a 2:1 ratio of women to men [2, 4]. This contrasts with the male preponderance in the malignant neoplasms localised as well as the diffuse mesotheliomas [4]. The mean age at presentation for the localised benign variant is 51 to 53 years (range 15-73) [2, 5, 6] and a mean 55 years for the localised malign tumors [2]. The benign solitary tumor has no association with asbestos exposure nor with a history of tobacco use [2, 4]. The diffuse malignant mesothelioma, however, has a well-recognised relationship to asbestos exposure [4].

Localised benign mesothelioma is found mostly as an asymptomatic lesion on routine chest radiography [2, 5]. When symptomatic, cough, chest pain, dyspnea and fever occurred most commonly and were found predominantly in the larger tumors [2, 6]. Extrapulmonary symptoms of pulmonary hypertrophic osteoarthropathy and digital clubbing are seen in as many as 17 to 20 percent of patients [2, 4]. Other less common symptoms include pleurisy, weight loss, hemoptysis, pneumonitis, and rarely hypoglycemia [2, 6]. Hypoglycemia is related to a pathological incretion of insulin-like growth factor by the tumour [6].

Chest radiography is the first step in detection of the lesion. It is especially important in the asymptomatic cases. Then a Chest CT is an important test in staging and developing a surgical strategy. A lesion of well-circumscribed lobulated borders but with no evidence of invasion is important in benign variety cases [7]. Ultrasound or magnetic resonance imaging (MRI) may also be occasionally useful in planning surgery with very large tumors, but like th CT, the findings are non-specific. However, in a study undertaken by PADOVANI et al. it was said that MR seemed to be the most accurate imaging method in the assessment of the diagnosis [8]. Bronchoscopy is important in the exclusion of other lesions. A transthoracic needle biopsy is not generally recommended, since a definitive diagnosis is rarely made and does not influence the surgical therapy with this tumor. Thoracotomy with open biopsy is the generally accepted as method of establishing a definitive diagnosis. Video-assisted thoracic surgical techniques may be used in resection of the smaller benign tumors [9, 10]. The larger tumors require a standard thoracotomy approach for both diagnosis and treatment with an emphasis on complete resection, which is generally curative. The benign tumors are usually pedunculated, although care should be taken with the pedicle, which may be highly vascular. The malignant variety more commonly originates from the parietal pleura and virtually never is pedunculated [2]. In most patients, local excision of the tumor is possible with sparing of lung tissue. Pleural adhesions frequently seen in the large (> 8 cm diameter) tumors may complicate the procedure [5].

Classification of mesotheliomas is based on several clinicopathologic criteria. Grossly the tumor is divided into diffuse and localised types. Microscopically the tumor is judged by the degree of pleomorphism and angiogenesis, and the number of mitoses. Histologically subdivisions are made based on tissue organisation: epithelial (carcinomatous), mesenchymal (fibrous), or mixed [11]. Grossly, the tumor is a firm, encapsulated and lobulated mass with a characteristic whorled ap-
pereance in the benign tumor and a more homoge-
nous texture in the malignant neoplasm. Necrosis
and haemorrhage can be seen in both types of cal-
cification and is generally confined to the benign
type. The size may vary from 1 cm to 36 cm in di-
ameter with a mean of 6 to 8 cm. The tumors may
reach gigantic sizes especially in the benign type,
but malignant tumors tend to be smaller [2, 5].
Size itself appears to have no bearing on whether
the tumor is benign or malignant, resectable, or
curable long term. Microscopically, the smaller be-
nign tumors (< 8 cm-diameter) tend to be poorly
vascularized, with no mitoses and with uniform
elongated spindle cells with varying amounts of
collagen and reticulin fibers. The larger tumors
more often have variable pleomorphism and a few
mitoses usually < 4 mitoses / 10 high-powered
fields [2, 5]. The localised malignant neoplasm has
a fibrous tubulopapillary, cubimorphic pattern in-
terspaced with bizarre stroma. Nuclear anaplasia
and a high mitotic rate are common [2, 5].
The separation of benign from malignant
mesothelial proliferations has emerged as a major
problem in the pathology of the serosal mem-
branes. There is no definitive pathological criteria
in differential diagnosis of benign and malign loca-
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