Mediastinal, Endobronchial, and Pleural Metastases of Immature Testicular Teratoma

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Abstract

This is the report of a 32 year old man presenting with dyspnea and chest pain that was hospitalized with findings of right massive pleural effusion. He had undergone orchiectomy one year previously and diagnosed as testicular teratoma. Thoracal computed tomography showed a necrotic mass occupying right upper and middle lobes, having variable internal echos, and regular contours, right-sided pleural effusion and another mass in anterior mediastinum. Bronchoscopy revealed a vegetal mass partly obstructing the intermediate bronchus. Biopsies of the endobronchial lesion and of the plevra were reported to indicate a metastasis of an immature testicular teratoma. Serum alpha-fetoprotein and pleural effusion alpha-fetoprotein and Ca19-9 levels were high. Dyspnea was relieved partially by repeated thoracentesis. The patient considered to be at stage IV, expired 13 months after the initial diagnosis. We reported this case as rare, having endobronchial, pleural, mediastinal metastases of immature testicular teratoma.

Turkish Respiratory Journal, 2003;4:(3):156-159

Key words: *immature testicular teratoma, endobronchial metastasis, pleural metastasis, mediastinal metastasis*

Introduction

Tumors of the testis are uncommon with an annual incidence of 2 per 100 000 males. They occur more commonly in whites. The peak age of incidence is 20 to 35 years. It is the most common malignancy in this age group.

About 95% of testicular tumors are malignant and drive from germ cells. Teratomas are classified as nonseminomatous germ cell tumors and represent a group of complex tumors having various cellular or organoid components remisniscent of normal derivates from more than one germ layer (1). Histologically three variants are recognized, based on the degree of differentiation. Mature teratomas are composed of a heterogeneous helter-skelter collection of differentiated cells or organoid structures, such as neural tissue, muscle bundles, islands of cartilage, clusters of squamous epithelium, structures reminiscent of thyroid gland, bronchial or bronchiolar epithelium, bits of intestinal wall or brain substance. Immature teratomas can be considered to have a structure intermediate between mature teratoma and embryonal carsinoma. In contrast to mature teratoma, in these tumours the elements of the germ cell layers are incompletely differentiated and not arranged in organoid fashion. However, even if the differentiation is

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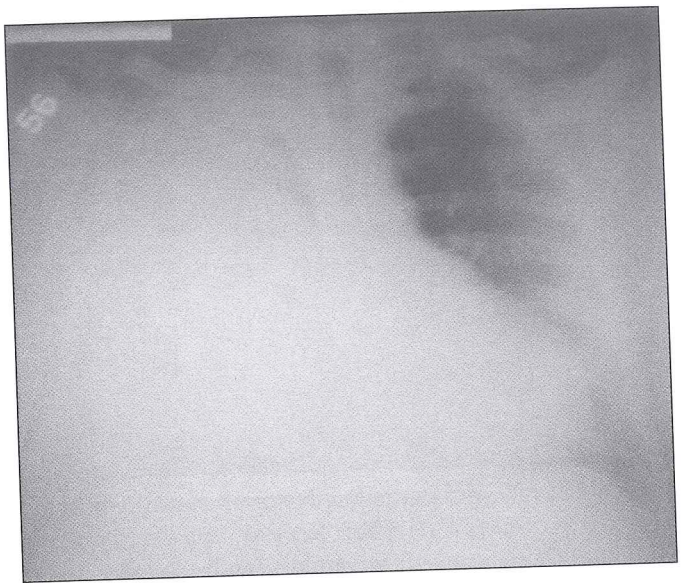

Figure 1. *Radiographic appearance of the chest*

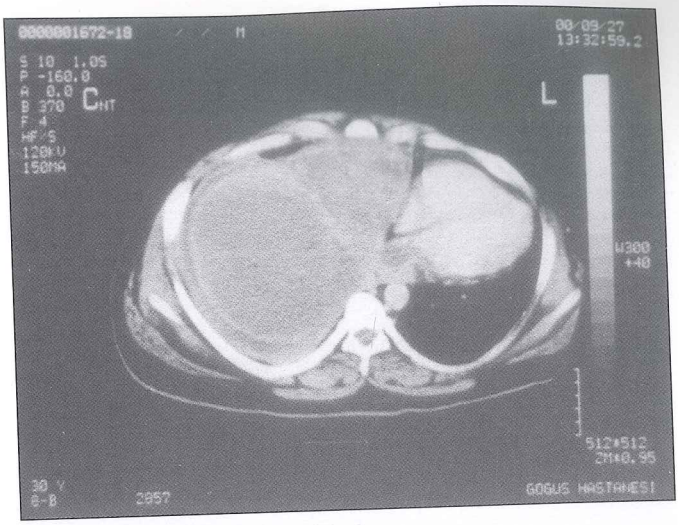

Figure 2. *Computed tomography of the thorax*

incomplete, the nature of the embryonic tissue can be clearly identified. Although these tumors are clearly malignant, they may not display clear-cut cytological features of malignancy.

Nonseminomas metastasize by both lymphatic and hematogenous routes. The liver and lungs are the main sites for the metastases and they are radioresistant.

Most patients with germ cell tumors present with a painless mass in the testis. A painful testicular mass is usually caused by bleeding into the neoplasm. Other presenting symptoms may be back or abdominal pain caused by retroperitoneal lymphadenopathy and shortness of breath caused by diffuse pulmonary metastases.

Germ cell tumors, especially nonseminomatous cancers, often secrete biological markers. Alpha-fetoprotein (α-FP) elevation in serum implies the presence of nonseminomatous elements in the tumor or metastases. Human choriogonadotropin (HCG) is secreted by almost all choriocarsinomas, by approximately one third of embryonal cell carsinomas and teratocarsinomas and, rarely, by pure seminomas. These tumor markers may be used to monitor the response to therapy.

Today advances in treatment have transformed testicular cancer from the most common cause of death in the second decade of life into one of the most curable of all cancers (1). In nonseminomatous testicular cancer, the addition of cisplatin to agressive multiple-drug chemotherapeutic regimens has resulted in response rates of over 90% and long-term remission in 50 to 90% of patients.

Case Report

The patient, a 32 year old man, presented with dyspnea and right sided chest pain. A massive pleural effusion was detected on the right side and was admitted for further investigation and treatment. Patient's history revealed that he had undergone an orchiectomy one year ago with a suspicion of inguinal hernia, and a diagnosis of testicular teratoma was made.

The patient was an obese mentally retarded male, who had dyspnea as a result of mediastinal shift. Breath sounds and vocal fremitus were absent on the right chest and dullness was present by percussion. There were no other noteworthy clinical findings.

Laboratory tests showed an erythrocyte sedimentation rate of 105 mm/h and white cell account of 10 000/mm^3 and an albumin level of 3.3 g/dl. Arterial blood gas findings were as follows: pH: 7.41, pCO_2: 32 mmHg, pO_2: 81 mmHg, HCO_3: 20 mmol/L, O_2 saturation: 95%. Other blood parameters were normal.

Chest radiogram showed a homogeneous mass, which occupied the right hemithorax from apex to diaphragma and causing contralateral mediastinal and tracheal shift (Figure 1). Ultrasonography of the abdomen was normal.

Computed tomography of the chest showed a mass which had regular contours but was necrotic in appearance. The mass occupied a large portion of the right upper and middle lobes. the tomography revealed a right sided pleural effusion and thickening and another mass in the mediastinum, 7 cm in diameter (Figure 2).

Bronchoscopy revealed that intermediate bronchus was narrowed 70% with a vegetal mass that originated from the medial wall (Figure 3). The histology of the biopsy specimen of this lesion was reported as a metastasis of immature testis teratoma (Figure 4).

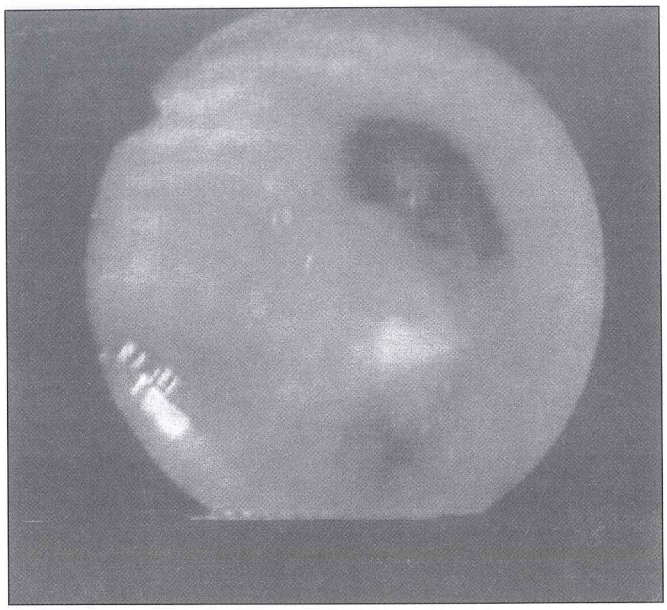

Figure 3. *Bronchoscopic appearance of the lesion in the intermedieate bronchus*

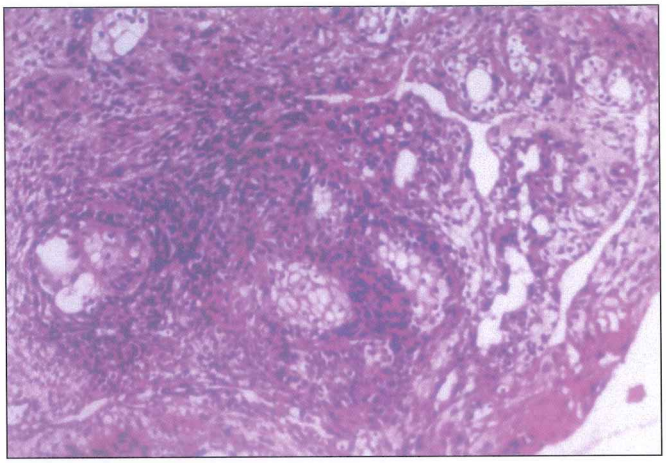

Figure 4. Histo*pathological appearance of the pleural metastasis.*

Table 1. Tumor marker levels in serum and pleural effusion

	α-FP	Ca 19-9	CEA	HCG
Serum	1430 (increased)	5.2	<20	<1
Pleural effusion	1516 (increased)	47.4 (increased)	7.5	15.6

CEA: Carsinoembryonic antigen
α-FP: Alpha-fetoprotein
HCG: Human choriogonadotropin

Tumor marker levels in the serum and pleural effusion are given in Table 1.

The case was evaluated as stage IV disease according to the Royal Marsden staging system. Dyspnea was relieved partially by repeated thoracentesis. Chemotherapy could not be administered because of patient incomplience. The patient expired 13 months after the initial diagnosis.

Discussion

The most frequent localizations of teratomas are testes and ovaries. Intrapulmonary teratoma are rare only 8 cases have been reported to date (2). Rare cases of primary endobronchial teratoma have also been reported (3,4).

Testicular immature teratoma is a rarely seen tumor. Usually testicular tumors spread first to the medial iliac and paraaortic lymph nodes, then to the mediastinal and supraclavicular lymph nodes. Hematogenous invasion to lungs, brain, and bones usually occurs in advanced stages. Rare cases of pulmonary metastasis have been reported (5,6). In our patient, metastases were found to develop simultaneously in pulmonary parenchyma, bronchial mucosa, pleura and mediastinum.

Teratomas tend to rupture. This phenomenon has been attributed to ischemia and/or necrosis caused by the increase in size of the tumor or to infection. It has also been suggested that sebaceous materials or digestive enzymes drived from tumor tissue tend to cause inflammation and necrosis, leading to rupture (7). Southgate et al, suggested that the tendency of a mediastinal teratoma to adhere and erode the surrounding structures was caused by proteolytic enzymes produced by the tumor (8). Choi et al, in 17 cases of operated mediastinal teratoma studied by computed tomography, concluded that the CT findings of homogeneity of the internal components and changes in the pleura, or pericardium adjacent to the lung parenchyma can be used as diagnostic signs of teratoma rupture (9). In our case, it was thought that the homogenous structure of the mediastinal and parenchymal tumors seen in thoracal CT and the presence of hemorrhagic pleural fluid could be taken as signs of rupture, but it has not been possible to evaluate the serum and pleural fluid amylase levels.

Teratomas localized in thorax usually present clinical symptoms related to their localization or complications. Cases exhibiting hemothorax and pyothorax because of rupture of mediastinal teratoma have been reported (7,10). Teratomas are seen typically in young men, and so was our patient (11).

Mediastinal teratomas are usually localized in anterior mediastinum. They present with symptoms of local thoracic compression and invasion. Coughing, hemoptysis and chest pain are frequent symptoms (11). Dyspnea and chest pain which were present in our patient were thought to be related to pleural invasion and compression.

Pathological diagnosis is essential for the definitive diagnosis of teratoma. In our case, the diagnosis was confirmed by testicular, pleural and bronchial biopsies. In nonseminomatous malign germ cell tumors, increased levels of HCG or α-FP are diagnostic (11). Our case had high levels of α-FP in plasma and in the pleural fluid, and a high level of Ca 19-9 in the pleural fluid.

Orchiectomy continues to be the primary treatment method of disseminated nonseminomatous germ cell tumors of testis. Chemotherapy regimens based on cisplatin serve to improve the prognosis and high cure rates have been reported (12). However, after a complete biochemical response to polychemotherapy, metastatic tissue may remain in the retroperitoneum and/or the lungs. In this particular group of cases, surgical resection is necessary to distinguish patologically the nature of the mass which could be a mature teratoma or a viable cancer, or simply represent necrotic or fibrotic tissue. In our case because of the patient's incompliance there was no possibility to administer chemotherapy and orchiectomy was the only treatment the patient recieved.

In conclusion, we have reported this case, because of the rare combination of endobronchial, pleural, and mediastinalmetastases, occuring together in the same patient.

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Yedigöller 2003, Turgay Çelikel MD