

A huge neoplasm occupying the right hemithorax in a pregnancy

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Abstract: Germ cell tumors, like teratoma, typically occur in young adults in their second to fourth decade with equal sex distribution. We firstly report a very rare case of a huge tumor compressed the vital structures of the mediastinum that was diagnosed in a 21-year-old woman at 39 weeks of gestation during a routine prenatal examination. The patient underwent complete en-bloc resection and the size of the tumor was extremely large although no invasion to the vessels or to the airway had occurred. Adherence to the adjacent right pulmonary artery and right main bronchus was present, but without erosion or fistulization. The patient has remained well for over 2 months after the treatment without any signs of disease recurrence.

Keywords: Teratoma; surgery

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Introduction

Germ cell tumors typically occur in young adults. Female ascendency has been reported by some authors with a ratio of 1.27-2.05 for female to male (1-3). In 1953, Willis defined the teratomas as true tumors composed of tissues that are foreign to the part or organ of the body (4). Anterior mediastinum is the most frequent site of occurrence of the tumors in thorax, and benign teratomas constitute about 3-12% (5). According to another review, 3-8% is located in the posterior portion of the visceral compartment (6). The incidence given for germ cell tumors complicating pregnancy varies in the literatures (7,8). We firstly report a very rare case of a huge tumor compressed the vital structures of the mediastinum that was diagnosed in a 21-year-old woman at 39 weeks of gestation during a routine prenatal examination.

Case report

An ultrasound scan was performed at 39th gestational week in a 21-year-old primigravida which revealed a 10 cm × 10 cm × 8 cm smooth-walled, mixed echogenic mass with irregular inner contents in the right thorax and presented

to our hospital with cough for 2 weeks. A repeat scan four weeks later after she underwent a cesarean section, showed the same in the size of the mass. Physical examination showed dullness to percussion and diminished breath sounds on the right side of the chest. Laboratory data, including serum tumor markers were all within normal limits. Chest X-ray showed total atelectasis of the right lung and contained some calcified components in the middle of the thorax (*Figure 1*). The contrast-enhanced computed tomography (CT) scan revealed a large heterogeneous well-defined mass of 16 cm × 10 cm in size on the right side abutting the chest wall and extended to the whole right pleural space (*Figure 2*). The mass showed heterogeneous density containing soft tissue elements, fat, cystic areas and foci of calcification. The echocardiography showed squeezing of the right atrium (*Figure 3*).

The patient, however, refused to have the Computed tomography-guided percutaneous aspiration examination. As diagnosis was not established through imaging, surgical management was first in the priority list of therapeutic options. The patient was subjected to the right posterolateral thoracotomy through the fifth intercostal space. Many adhesions existed with the right pulmonary artery, the right main bronchus, the pericardium, the

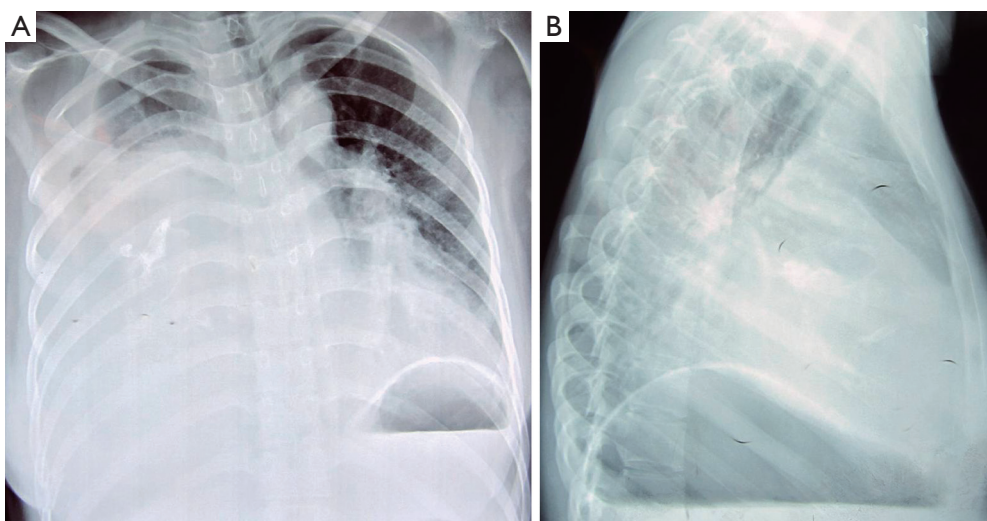


Figure 1 Chest X-ray of the case. Chest X-ray showed total atelectasis of the right lung and contained some calcified components in the middle of the thorax (A,B).

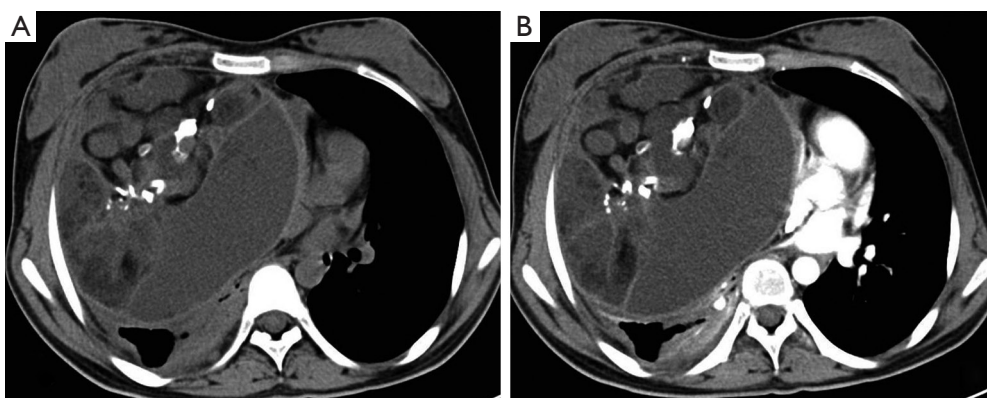


Figure 2 Contrast-enhanced CT features of the case. The contrast-enhanced CT scan revealed a large heterogeneous well-defined mass of 16 cm × 10 cm in size on the right thorax (A,B).

superior vena cava, and the diaphragm, and a combination of blunt and sharp dissection for the division was applied uneventfully. Because of difficulty in the mobilization of such a huge mass, a purse string suture permitted aspiration of sebaceous content via a small incision in the wall. As the size diminished, manipulation was facilitated. The tumor was completely resected and the collapsed right lung was easily re-expanded. Macroscopic examination of the resected tumor was white-gray colored, well circumscribed, and measured 16 cm × 13 cm × 10 cm in size. Histologically, it contained skin components with sebaceous glands, vessels, and fat, and muscles with bone, bone marrow, and gastrointestinal epithelium (*Figure 4*). No malignant or immature component was found. She is doing well without

evidence of tumor recurrence 2 months following her initial diagnosis.

Discussion

Teratomas are uncommon neoplasms comprised of mixed dermal elements derived from the three germ cell layers and are characterized by the presence of virtually any tissue type. According to the mediastinal germ cell tumor classification system proposed in 1986 by Mullen and Richardson, there are three categories: benign germ cell tumors, seminomas, and nonseminomatous germ cell tumors, also called malignant teratomas (9). Germ cell tumors comprise 15-20% of all anterior mediastinal tumors and benign

mediastinal teratomas accounts for 60% of all germ cell tumors (10). Benign mature teratomas contain components derived from more than one of the three primitive germ cell layers: ectoderm: hair, skin; mesoderm: bone, cartilage, muscle, fat; endoderm: bronchial, intestinal, pancreatic tissue (11). A PubMed searching of the period from 1990 to 2014 revealed little information on the “huge teratoma” of gravida as seen in our patient. We firstly report a very rare

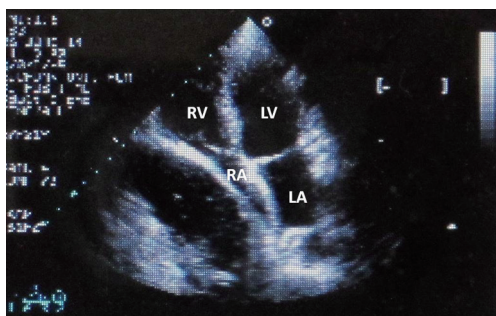


Figure 3 Echocardiography of the case. The echocardiography showed squeezing of the right atrium.

case of a huge teratoma that was diagnosed in a 21-year-old woman at 39 weeks of gestation.

Benign teratomas are usually asymptomatic and may be diagnosed as an incidental finding. Common presenting symptoms include chest or shoulder pain, dyspnea, cough, fever, pleural effusion, and bulging of the chest wall (12). In the case under discussion, the patient presented cough for two weeks. With regard to radiologic evaluation, the most characteristic radiologic finding of these neoplasms is identification of a complex mass containing a well-circumscribed fluid volume, fat-fluid level and calcifications. These findings are best identified by CT. CT offers superior identification of fat as either sebum or adipose tissue than by ultrasound. A fat-fluid level, however, has also been described in a case of well-differentiated liposarcoma of the retroperitoneum (13). There is also a word of caution in our case. Patient examined with chest X-ray showed total atelectasis of the right lung and contained some calcified components and the contrast-enhanced CT scan revealed a huge heterogeneous well-defined mass on the right side abutting the chest wall, which probably extended to the whole right pleural space. While CT was the standard for

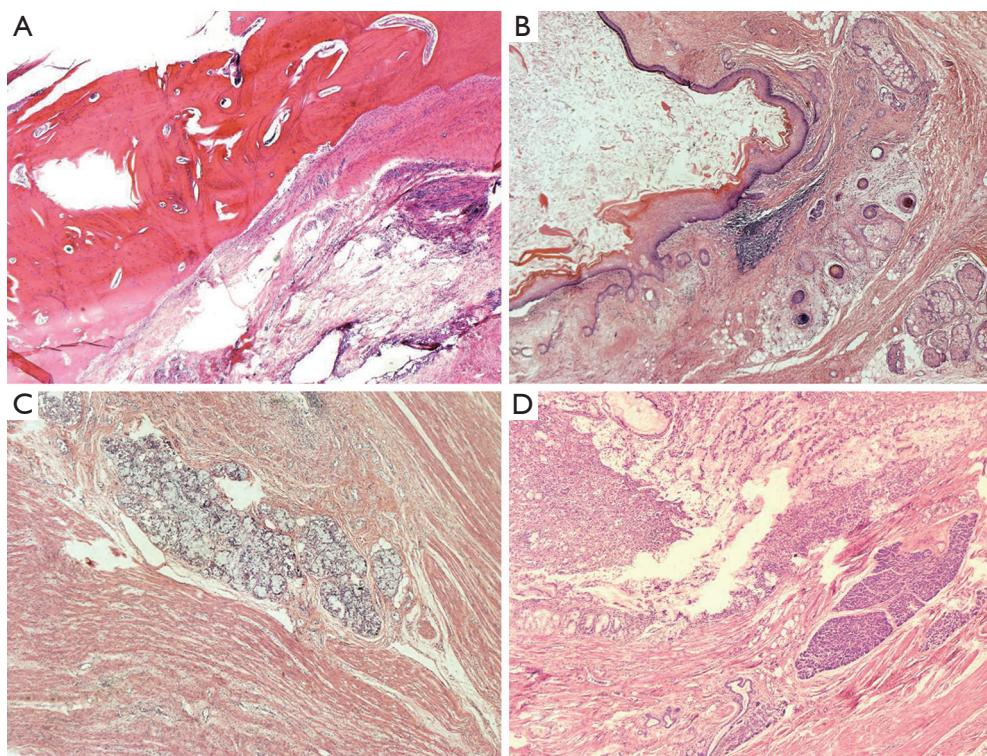


Figure 4 Histological and immunoistochemical features. Histologically, it contained skin components with bone marrow (A), sebaceous glands (B), muscles and muciparous glands (C) and gastrointestinal epithelium (D). (40×).

retroperitoneal imaging, MRI allows for improved soft tissue resolution. It is useful in assessing for encasement or invasion of blood vessels, which aids in the determination of malignant potential and resectability. Fat is suggested by high-intensity signal on T1-weighted images (14). In our case, the patient refused to have the examination of MRI for the high cost.

Macroscopically, there are two variants. Cystic teratomas are composed of fully mature elements and are usually benign. Solid teratomas are more likely to be malignant and formed of immature embryonic tissue, fibrous tissue, fat, cartilage, and bone (15). Microscopically, cysts lined by a variety of epithelia, calcification and ossification are often seen. Benign mature cystic teratomas include epidermis, brain and glial tissue, teeth, cartilage, peripheral nerve, smooth muscle, respiratory epithelium, connective tissue, and intestinal epithelium (16). The teratoma classification system introduced by Gonzalez-Crussi (17), allows for stratification based on the nature of the tissues present within the tumor, their proportion, and their characteristics.

The differential diagnosis of benign teratoma is malignant teratoma. Malignant transformation has been reported to occur in 1% to 2% of ovarian cystic teratomas and in a few cases of mediastinal teratoma, especially giant mediastinal teratoma (18). The possibility of malignant transformation should be considered in any cystic tumor with an invasive solid portion in the wall. Malignant teratomas tend to progress rapidly and are generally diagnosed at advanced stages when symptoms develop. Renato *et al.* (19) reviewed ten cases of malignant retroperitoneal teratomas and found that the most common presenting symptoms were abdominal pain, an abdominal mass, vomiting, weight loss, and fever. Germinomas, while malignant, are radiosensitive and multi-modality treatment (chemotherapy, radiation, and surgery) often leads to effective tumor control.

Surgical resection is the treatment of choice and radical extirpation secures a long survival out of recurrence. Chemotherapy and radiotherapy have relatively small roles in the management of these tumors. Median sternotomy is usually preferred for tumor removal, but access via either posterolateral or anteroposterior thoracotomy depends on the size, location, and expansion of the tumor. Difficulty in surgical maneuvers may be a result of the vital structures involved (20). In the case reported, right posterolateral access was chosen, as the tumor was almost entirely located into the right hemithorax, reaching and adherent to the left hemidiaphragm. Such a location precludes median

sternotomy, otherwise the preferred approach, as surgical manipulations are impossible on the lower lobe of the right lung and on the right hemidiaphragm. CT-scan revealed the relations with the mediastinum vital anatomical structures because it was critical to clarify whether there was an invasion or simply compression. At last, the tumor was completely resected and the collapsed right lung was easily re-expanded. Microscopic examination showed mature cystic teratoma.

Conclusions

Huge teratoma is rare neoplasm that is diagnosed in a pregnant woman. Teratoma is suggested by CT demonstrating a complex mass with a well-circumscribed fluid volume, fat-fluid level, and calcifications. While the majority of these neoplasms are benign, a variety of malignant components may be present or develop from clonal transformation. In the case reported, the primigravida presented only the cough for 2 weeks and the growth of the huge tumor has little influenced the pregnancy. Surgical resection and careful histologic evaluation is required for diagnosis. To facilitate the preoperative diagnosis and avoid the misdiagnosis of such rare disease, more cases will need to be reported.

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