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DOI: https://doi.org/10.33821/762 **Case report / Caso clínico**

Mediastinal mature cystic teratoma in an adolescent patient. Case report and literature review

Teratoma quístico maduro de mediastino en un paciente adolescente: reporte de caso y revisión de la literatura

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ABSTRACT

Introduction: Mediastinal mature cystic teratoma is a primary germ cell neoplasm with benign features. It is composed of fully differentiated tissues derived from more than one of the three embryonic germ cell layers and occurrs mainly in adolescents and young adults. They appear most frequently in the anterior compartment, secondarily at the level of the thymus and rarely in the posterior compartment. Clinical case: We present the case of a 13-year-old adolescent patient with an apparent mediastinal tumor, which caused moderate chest pain, dyspnea on moderate exertion accompanied by asthenia and progressive weight loss. Treatment: Total resection of the anterior mediastinal tumor is performed, which confirms the histopathological diagnosis of solid cystic mature tridermal cystic teratoma of the mediastinum, with negative lymph nodes for malignancy. Conclusion: Mature cystic teratoma is the most frequent pathology of mediastinal tumors, of benign characteristics, with non-specific symptomatology. Surgical resolution remains the gold standard in terms of treatment, with low recurrence rates after complete surgical resection and with good short, medium, and long term prognosis.

Keywords: MeSH: Mature Cystic Teratoma, Germinal Tumor, Mediastinal Lesion, Adolescent.

RESUMEN

Introducción: El teratoma quístico maduro de mediastino es una neoplasia de las células germinales primarias de características benignas, compuesta de tejidos completamente diferenciados, derivados de más de una de las tres capas de células germinales embrionarias, que aparece principalmente en adolescentes y adultos jóvenes. Se presenta con mayor frecuencia en el compartimento anterior, en segundo lugar, a nivel del timo y, rara vez, en el compartimento posterior. Caso clínico: Se expone el caso de un paciente adolescente de 13 años con aparente tumoración en mediastino que produce dolor torácico moderado, disnea de moderados esfuerzos acompañada de astenia y pérdida progresiva de peso. Tratamiento: Se realizó resección total de tumor de mediastino anterior, el cual confirmó el diagnóstico histopatológico de teratoma sólido quístico maduro tridérmico de mediastino con ganglios linfáticos negativos para malignidad. Conclusión: El teratoma quístico maduro es la patología más frecuente de los tumores mediastinales de características benignas con sintomatología inespecífica. La resolución quirúrgica sigue siendo el GOLD estándar, con poca tasa de recurrencia después de la resección quirúrgica completa y un buen pronóstico a corto, mediano y largo plazo.

Palabras Clave: DeCS: Teratoma quístico maduro, Tumor germinal, Lesión mediastinal, Adolescente.

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1. Introduction

Mediastinal thoracic neoplasms can occur in different compartments: anterior, middle, and posterior [1,5]. The anterior mediastinum is usually the seat of several types of neoplasms, germ cell tumors constitute 10% to 15% of them [1].

Teratoma is the most common germ cell tumor; it is characterized by the presence of tissues of origin from more than one of the three embryonic sheets. They account for 15% of such mediastinal germ cell tumors and usually occur in the anterior mediastinum near the thymus [2,10]. The incidence of teratomas is approximately 1 in 4,000 live births worldwide [3]. They mainly affect adolescents and young adults, with no sex predominance [4,5]. The clinical picture when it appears presents with a sudden onset of shortness of breath [6], with the most frequent symptoms being the product of chest compression such as cough, pain, lung infection, and dyspnea (or both) [7,8]. Most patients with mediastinal masses are asymptomatic and are only discovered by chance or incidentally [9]. Diagnosis is usually made with routine chest radiography (X-ray); however, other diagnostic methods are preferred, such as thoracic CT, magnetic resonance imaging (MRI), and even positron emission tomography (PET/CT). Radiologically, teratomas are characterized by rounded, lobulated, well-defined, anterior mediastinal masses that generally insinuate towards one side of the midline [10, 11].

The treatment of choice is undoubtedly surgical, with favorable prognosis [12]; therefore, surgical excision or total or complete resection of the tumor, including the tissues adherent to it, is preferred [13, 15].

The aim of this article is to report the case of a mediastinal mature cystic teratoma in an adolescent patient, along with the concerning exhaustive review of the literature regarding the subject.

2. Case Report

We present the case of a 13-year-old adolescent male with no pathological history of importance, who was transferred from another health center for presenting an apparent mediastinal tumor under study. It produced moderate chest pain, dyspnea on moderate exertion about 2 months ago and exacerbated in recent weeks accompanied by asthenia and progressive weight loss. No accompanying neurological or digestive symptoms or signs were evident. The patient initially underwent a chest X-ray and chest CT S/C, without a definitive diagnosis. These reported mediastinal mass with probable origin of Hodgkin's lymphoma.

On physical examination, there was palpable left submaxillary adenopathy and decreased bladder murmur in the right lung field with no intercostal pull.

2.1. Diagnostic workshop

The patient was evaluated by the Pediatrics and Thoracic Surgery service, under the presumptive diagnosis of mediastinal tumor of uncertain behavior, some complementary tests were requested, including paraclinical laboratory studies, a Chest X-ray (RX), spinal cord biopsy and Positron Emission Tomography (PET/CT). In the paraclinical laboratory studies, it was evidenced that the hemogram, blood biochemistry, coagulation times parameters performed in the Clinical Laboratory Department of SOLCA-Guayaguil were within normal ranges.

Special laboratory studies were also performed, which were within acceptable parameters and consisted of:

Human Chorionic Gonadotropin (HCG): 0.20 mIU/ml, Alpha Feto Protein: 1.14 IU/ml, Lactate dehydrogenase (LDH): 270 U/I.

The report of the standard and lateral Chest X-ray (RX) identified a radiopaque lesion observed in the upper and middle third of the right hemithorax with base in the mediastinum with partially defined lobulated contours. It does not cause displacement of the main bronchi and heart, measures approximately 12 x 10 cm in major axes, and is associated with increased density of hilar structures (Figure 1).

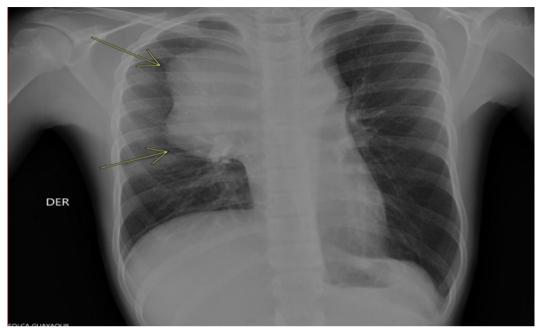
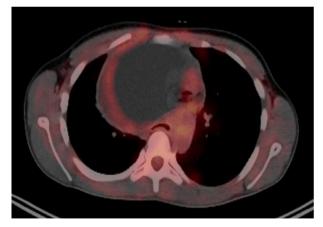


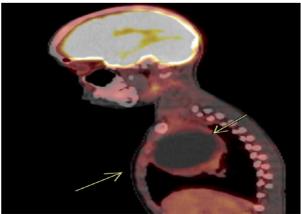
Figure 1. Standard and lateral chest X-ray showing radiopaque lesion in the upper and middle third of the right hemithorax.

Source: Department of Radiology and Imaging.

The bone marrow biopsy reported hypocellular bone marrow, free of neoplastic infiltration. Subsequently, it is evaluated with the results of complementary examinations. Since there was not a conclusive and definitive diagnosis, it is decided to plan for surgical procedure: thoracotomy vs sternotomy + tumor resection as a priority, in order to define the morphological and anatomical characteristics of the lesion and its relationship with adjacent structures. Additionally, it was requested to rule out a mediastinal mass vs Hodgkin's lymphoma, an oncological examination PET/CT, which reported single polylobulated, heterogeneous mass in the chest, with variable density (liquid, soft tissue and fat) with calcification in its wall. It measures approximately 7.3 x 11.3 x 16.6 cm, and it rejects but does not infiltrate neighboring structures (thyroid, trachea, esophagus, aortic arch) with hypermetabolism in its wall (Figure 2).

Α.





В.

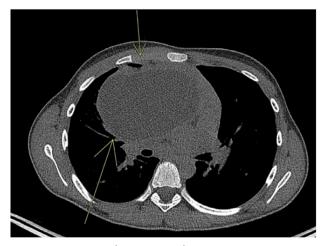




Figure 2. PET/CT. A. PET/CT. A. Axial section where the arrow shows a single polylobulated mass measuring 7.3 x 11.3 x 16.6 cm. B. Sagittal view showing with the arrow a single polylobed mass measuring $7.3 \times 11.3 \times 16.6 \text{ cm}$.

Source: Department of Radiology and Imaging.

2.2. Treatment and evolution

The patient underwent sternotomy + total resection of anterior mediastinal tumor + biopsy + placement of right chest tube. The surgical procedure lasted 4 hours, with an approximate bleeding of 600 ml. An incision was made in the midline of the sternum, finding intraoperatively: tumor in the anterior mediastinum region attached to the superior vena cava, with macroscopic characteristics: oval, brown, rough surface, irregular edges, multilobulated, measuring approximately 17.2 x 12.1 x 4.8 cm. A whitish pedunculated cystic formation measuring 4.5 x 3.3 x 2.3 cm with a rough surface with hairs was observed (Figure 3). In addition, during surgery, probable lymph nodes are isolated, measuring 0.7 x 1.5 cm.

The pathology report of the surgical specimen showed solid tridermal mediastinal mature cystic teratoma. The pathology study of the isolated lymph nodes revealed lymph nodes (3) negative for malignancy.

Cytology of the mediastinal tumor fluid reported hemorrhagic smear with isolated inflammatory cells (neutrophils), negative for malignancy.

Total exeresis of the solid tridermal mediastinal mature cystic teratoma was performed, with successful release of the superior vena cava adhesion, which was found to be undamaged, with no intraoperative complications, nor were there any complications in the postoperative period. Due to the complexity of the procedure, the postoperative period was managed in a multidisciplinary way by the Pediatrics Service and the Thoracic Surgery Service. The patient was initially admitted to the Pediatric ICU, where he remained 4 days after surgery, with the presence of a right chest tube with daily quantification placed in the operative room. It was removed on the sixth postoperative day when adequate bilateral pulmonary expansion was evidenced.

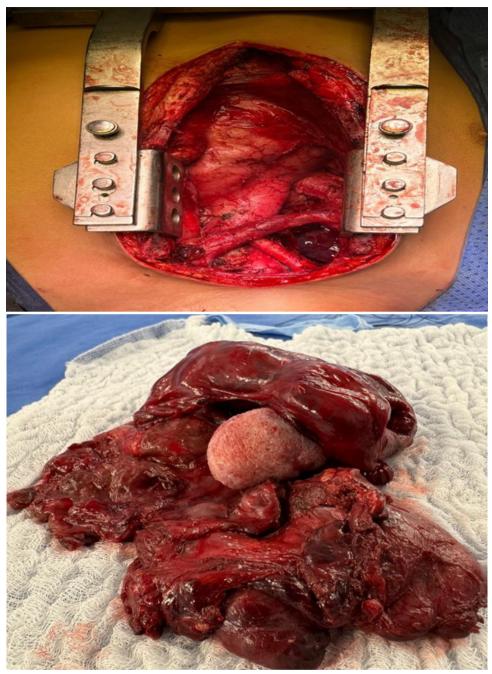


Figure 3. Intraoperative findings of anterior mediastinal tumor exeresis. Source: SOLCA-Guayaquil.

Finally, the patient was discharged on the ninth postoperative day without apparent postoperative complications. He consulted our service during the first three subsequent weeks, in which surgical wounds in good healing process, no signs of infection, with relative improvement of the initial symptomatology were evidenced. A control chest X-ray was performed, which reported no evident active lesions in the pulmonary parenchyma, free cost and cardiophrenic angles, no anomalous elevation of the diaphragmatic domes, osteosynthesis material in sternal topography and epigastrium (Figure 4).

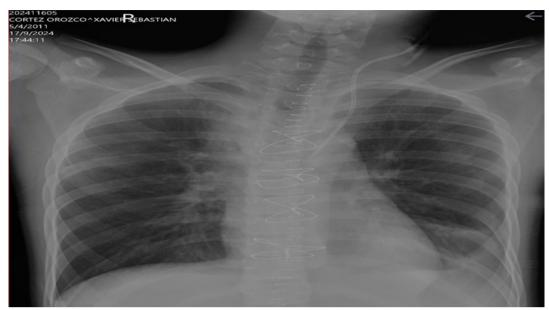


Figure 4. Radiographic control of postoperative evolution of anterior mediastinal tumor exeresis Source: SOLCA, Guayaquil.

Continuing with the clinical follow-up, the patient is scheduled for subsequent controls with high resolution thoracic CT scan, performed two months after surgery, showing no tumor recurrence. Having negative tumor markers, negative spinal studies, the patient does not receive specific oncologic adjuvant treatment. To date, he attends periodic controls at the Thoracic Surgery Service.

3. Discussion

Primary germinal tumors of the mediastinum have been classified into three main groups: teratomas, seminomas, and non-seminomatous tumors. Histologically they are classified into two groups: benign (mature teratoma) and malignant (immature teratoma) and teratoma with malignant component and teratoma with embryonal cell carcinoma [14].

Teratoma is a primary neoplasm with benign characteristics of germ cells of fully differentiated tissues derived from more than one of the three embryonic germ cell layers: ectoderm, mesoderm, and endoderm [1]. Of all primary germ cell tumors of the mediastinum, the most frequent is mature teratoma [2,8]. The frequency of mediastinal teratomas ranges from 1% to 5% [3,21]. They occur in young adults between the second and fourth decades of life, with equal frequency for both sexes [4,6]. Depending on the degree of differentiation their appearance can range from benign to malignant tumors and have cystic or solid areas, or both, of different sizes. This can be distinguished in their content [4]. Most mature or benign teratomas are cysts (dermoid) and contain well-differentiated somatic elements, such as hairs, fat, skin and cartilage [5]. According to the World Health Organization (WHO), mediastinal teratomas, according to their location, occur most frequently in the anterior compartment (prevascular); secondly, at the level of the thymus, and rarely in the posterior compartment [5].

Symptomatology is nonspecific, it may be asymptomatic or when symptoms are present, they may be due to compression, invasion of adjacent structures or secondary infection, including dyspnea, chest pain, cough, fever, weight loss, superior vena cava syndrome, dysphagia, orthopnea, hemoptysis and, rarely, cardiac tamponade [6,7,17]. There may also be nonspecific chest pain or pleural pain when intercostal nerves are involved [6,13,27]. However, tumor rupture in the bronchial tree may produce hemoptysis and cough with expectoration of hair or sebaceous material, indicating a fistula between the tumor and the tracheobronchial tree, which is pathognomonic of a teratoma [7,10,16]. In our patient, the most striking symptomatology was dyspnea, chest pain, and progressive weight loss, the latter being a more specific symptom, but not conclusive of a tumor of malignant origin or invading adjacent structures.

Laboratory tests are usually normal, with serum levels of human chorionic gonadotropin and alpha-fetoprotein being normal in patients with benign teratomas, but the immaturity of the histopathological elements present in the tumor, as well as elevated blood levels of alpha-fetoprotein hormone, are indicators of worse prognosis [1,8,12]. According to the literature consulted, tumor marker assays are crucial, as they can indicate the presence of a malignant component, such as seminomas or nonseminomatous germ cell tumors. Lactate dehydrogenase levels are also evaluated in cases where lymphomas are suspected [9,20]. In our patient, the results of these tumor markers yielded normal results.

The diagnosis is usually made by chance on routine radiography [10]. Chest radiography, in posteroanterior and lateral projections, is the initial radiological examination for mediastinal tumors [11,14]. Radiology examinations usually reveal a well-circumscribed mediastinal mass that often protrudes into one of the lung fields [12].

To complement the radiological diagnosis, a computed tomography (CT) scan of the chest should be performed, which provides information on tissue density and tumor delineation. Mature teratomas usually appear on CT as an anterior mediastinal mass containing soft tissue, fluid, fat or calcium deposits [13,17]. Pleural effusion can also be found on this imaging method and is more common in ruptured mediastinal teratomas [12].

Homogeneity or heterogeneity on chest CT enables distinguishing between ruptured and unruptured teratomas [13,15]. Magnetic resonance imaging (MRI) can also be used because it assesses the anatomic relationships with adjacent structures and allows safer and more appropriate planning of the surgical approach and tumor resectability [16].

Positron emission tomography (PET/CT) is an objective and useful modality in the differential diagnosis and treatment of anterior mediastinal tumors, including teratomas [14,17]. In our specific case, PET/CT was used as an adjunctive examination because high-resolution chest CT was not immediately available and considering that a mediastinal mass with probable Hodgkin's lymphoma origin was initially thought to be present.

A wide range of conditions can be considered in the differential diagnosis of a lesion containing heterogeneous fat in the anterior mediastinum. In distinguishing between mature cystic teratomas and other entities such as mediastinal lipoma, mediastinal lipomatosis, thymoma, liposarcoma, may also include hydatid cyst, lung abscess and parenchymal lung tumor [16] the presence of calcification and fluid elements is a key distinguishing factor [17,18].

The diagnosis of mature cystic teratomas is confirmed by anatomopathologic examination, which also provides valuable information on the extent of resection [19,22].

Surgical treatment is the gold standard, since mature teratomas are usually curable by complete excision of the tumor. However, it should be kept in mind that a thorough pathologic review is mandatory and definitive to exclude other differential diagnoses, such as small immature tissue tumors, other germ cell tumors or carcinomas [3,7,23].

The most frequently used access route is the median sternotomy [22,25], which helps to avoid complications such as compression of adjacent structures, rupture (rare) into the lung or bronchial tree, with consequent hemoptysis into the pleural or pericardial spaces and less common malignant transformation [1,26]. In cases where the tumor is localized to a specific hemithorax, an anterolateral thoracotomy is often favored. While complete removal of the tumor is generally achievable, it can be challenging because it is often found adherent to neighboring structures such as the thymus, pleura, and pericardium. In some cases, endoscopic removal may be a viable option for small tumors [23,28]. In our particular case, a median stereotomy with complete surgical excision was performed, without apparent complications, since the surgical specimen, despite its large size and adherence to the vena cava, was completely excised.

There are several complications of the surgical technique, such as infection at the surgical site, empyema, sepsis, and death due to the large size of the tumor and its adhesion to the surrounding organs [24,25]. Other complications included persistent atelectasis and rupture in some area of cystic mass during difficult dissection with tumor content material [26].

The prognosis of mature mediastinal teratomas is good after complete resection. Recurrences are rare, and are associated with incomplete tumor resection [12,27,30]. In children, the postoperative prognosis for mature teratomas is typically favorable, with a survival rate exceeding 96 % [28].

Long-term clinical, laboratory, and imaging surveillance is necessary for individuals who have undergone surgical removal (resection) of a mediastinal teratoma. This surveillance should be performed at shorter intervals for the first 5 years after surgery and then annually thereafter [9, 29].

4. Conclusions

Mature cystic teratoma is the most frequent pathology of mediastinal tumors, which presents with very nonspecific symptomatology, especially in adolescent patients, such as the patient in our clinical case. Total or complete surgical resection is the ideal treatment, with low recurrence rates and good short, medium and long term prognosis.

This particular case highlights the importance of considering mediastinal mature cystic teratoma among the most frequent pathologies of the mediastinum that should be studied to have adequate management and clinical follow-up. In this context, it is of utmost importance to have a correct diagnostic and therapeutic approach, which will allow proposing a series of similar clinical cases as multicenter studies for future medical research.

Abbreviations

PET CT: Positron Emission Tomography

CT: Computed Axial Tomography

MRI: Magnetic Resonance Imaging

RX: X-ray

5. Administrative Information

5.1. Additional Files

None declared by the authors.

5.2. Acknowledgments

We thank the patient and his family, who agreed to the dissemination of this scientific work.

5.3. Authors' contributions

All authors read and approved the final version of the manuscript.

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5.5 Availability of data and materials

Data are available on request from the corresponding author. No other materials are reported.

5.6 Statements

5.6.1 Informed consent

The patient's legal guardians gave written informed consent for publication of this case report and accompanying images. The Editor-in-Chief of this journal has a copy of the written consent for review.

5.6.2 Conflicts of Interest

The authors declare that they have no conflicts of competence or interest.

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