Mediastinal Cystic Teratoma Misdiagnosed as Pleural Tuberculosis: a Case Report and Review of 63 Cases Revealed by Pleural Effusion.

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Abstract

Background: Mediastinal cystic teratoma is a rare diagnosis in adolescence from low-income setting. Pleural effusion often mislead to infection and delay resection. We report a case from high burden tuberculosis country and analyze data from the literature. Methods: Clinical record of an adolescent patient from the Democratic Republic of Congo

Introduction

A teratoma is defined as a type of germ cell tumor that may contain several different types of tissue such as hair, muscle and bone.[1] It may be immature or mature, based on how normal cells looked under microscope; sometimes it is a mixture of both. They may occur in different parts depending on the sex (testicles, ovaries) or either in the chest, nervous system or abdomen. They may be benign or malignant.[1,2]

Mediastinal teratomas are the most common extra-gonadal germ cell tumors. They account approximately for 15% of anterior mediastinal masses in adults and 25% in children and 50-70% of mediastinal tumors.[3,4] They may be symptomatic through different ways either by mass effect, endocrine function impaired or by rupture creating pleural effusion.[3–8]The pleural effusion can often lead to a misdiagnosis of respiratory infections especially in low-income setting. We report on case of a cystic teratoma in a 15-year-old female from a low-income setting treated twice as tuberculosis. We also performed a review on cystic mediastinal teratoma with pleural effusion in the literature. In addition to our patient, we reviewed 63 records from other institutions identified via Medline between May 2017 and November 2017 that were well documented. Our review focused on analysis of clinical presentation, pathology findings, surgery procedure and immediate and follow-up outcomes.

Methods

Patients

All cases were identified through Medline from different institutions using pre-defined MeSh terms [mediastinal teratoma] OR [mature mediastinal teratoma] OR [mature teratoma]OR [immature teratoma] AND [pleural effusion] between May 2017 and November 2017. Mediastinal teratoma was diagnosed by anatomo-pathologic findings after surgery resection or during medico-legal expertise. The records of 64 patients were used for clinical analysis. We excluded two patients from whom clinical details and surgery outcomes were not obtained through our search. From full-text articles we extracted age of the patient at diagnosis, sex, country, first symptoms developed, type of surgery, anatomo-pathologic findings (mature/immature, benign/malignant, type of tissue found) as reported and patients outcomes after surgery (death/relapse). The time to diagnosis was calculated from the first onset of respiratory symptoms and the confirmation of diagnosis through histological analysis after surgery resection or autopsy. Chest X-ray and CT examination were

the major tools for establishing the preoperative diagnosis. Descriptive analysis was essentially conducted for the report of the literature review. Categorical variables were expressed as proportions and continuous variables were expressed by means and standard deviations (±SD). Data were captured on a Microsoft Excel spreadsheet and exported to STATA version 14.1 (StataCorp LP, College Station, Texas 77845 USA). A written consent was secured from our patient prior the study commenced.

Results

Case report

Patient presentation

A 15-year-old female was transferred to our hospital with pleural effusion symptomatology along with respiratory distress (RD) which had occurred on multiple occasions. Her respiratory distress could have been relieved only by repetitive pleural tapping as much as 36 times for the last three years at the sequence of every four weeks before the actual transfer. She come from a rural setting in the East of DR Congo and residing in an informal settlement. Her medical history acknowledged that she was born healthy to non-consanguineous parents, the sixth of eight healthy children. She apparently started developing the symptoms 3 years prior to the transfer with an increasing shortness of breath (SB) requiring medical assistance and incapacitated her to pursue studies and to accomplish ordinary activities. She had then been twice on six months of anti-tuberculosis drugs first line regimen and had completed the treatment six months before the transfer. Our clinical assessment confirmed a woody note and a silent at auscultation on the left thorax. The right thorax presented a normal vesicular breath sound. The chest x-ray and the CT-Scan revealed a giant mediastinal masse with a collapsed left lung and consolidation.

Surgical procedure

The surgery by left posterolateral thoracotomy noted a dense adhesion between the masse and the chest walls, pericardium and left lung. The left lower lung lobe was completely deflated and incarcerated by the masse with islands of epithelial tissue, hair and calcified tissue. Anatomopathological findings led to the diagnosis of a mediastinal mature cystic teratoma. The patient was then transferred to the intensive care unit for three days and required a prolonged stay in the wards due to the collapsed left lung. She benefited with a flexible bronchoscopy twice with an insertion in situ of a chest tube for drainage. The chest tube was finally removed 15 days later after few trials of chest tube clamping followed by chest x-ray.

Patient evolution

No other complication occurred during the intervention. With adequate antibiotics prescribed for 14 days along with planed physiotherapy exercises and diet restrictions, the patient did not present any late complications (infection, respiratory distress) or story of collapsed lung. Patient was then discharged with antalgic that she took for almost three weeks. She stays in the city within a benevolent family to avoid exposition to biomass fuel smoking and to reduce risk of pulmonary infections inherent to the rural area. After five years of follow-up, our patient did not complain for respiratory symptoms and she is now starting her first year at the university.

Review of 63 Cases of Mature Teratoma with Pleural Effusion

Sixty-three cases were reported and analysed from our search findings. 59% were females and mean (±SD) age was 26(±12) years. Most of them come from high-income countries (Japan 28%, France 25%, USA 18% following by India 7%). Cough, shortness of breath, fever and chest pain was mostly reported. Majority of cases were misdiagnosed with time to first clinical symptoms ranged from one week to two years. Further, our case was first treated twice as tuberculosis (different regimens). About 91% showed an immature aspect whilst 16% were malignant. Ovarian and pancreatic tissues were mostly retrieved as content. Duration of hospitalisation varied from four days to six months and septic as well neurologic complications were mostly reported. With a follow-up period (of half of cases) ranged from one week to two years, roughly five percent of patients died after surgery and 26 percent relapsed.

Discussion

To our knowledge, there has no previous review of literature reporting on mediastinal cystic teratoma with pleural effusion. We find that mature teratoma with pleural effusion presented a high proportion in female patients during their young adult life. However, the literature search demonstrates the occurrence of mature teratoma with pleural effusion in patients as young as at 25 weeks of gestation and as old as 51 years.[9–62] Respiratory symptoms are predominant and not specific which has led often to a delay in the diagnosis for as long as 2 years.[9–11,19,20,22,28,30,32–61] The majority of studies were reported in high-income countries. The requirement for the diagnosis to be made by CT scan and histopathology might explain the fact that such diagnosis is mostly reported in high-income countries. Tissues found in the tumour appeared mostly immature and of ovarian and pancreatic origin. Except from cases that presented complications, the duration of hospitalisation was short with variable lengths of follow-up periods. Complications related to infections and metastases were reported in less than 10 cases. Relapses occurred for some cases and death was a rare outcome and was reported in 10 cases due to respiratory complications essentially.[17,22,24,32]

The major strength of our study is the systematic search of the literature and the number of the cases reported. This will allow a more systematic assessment of the prevalence of the disease and could raise awareness among clinician from low and middle-income countries in particular. Our results have highlighted a substantial proportion of cases from low-income settings. Ascertainment misclassification following difficulties to diagnose mediastinal cystic teratoma might preside on the wrong impression of its rarity in poor settings. Regarding the high burden of respiratory infection diseases in such a region, clinicians might consider mediastinal tumours after an attempt to treat an infection disease not specified otherwise.

Although a systematic search of literature through Medline, among limitations of our study are the non-systematic search of [?]3 databases and the exclusion of studies without full-texts. Another limitation was related to the nature of the study that reported only on cases thus not representative of the populations from which the cases came from. However, the review has highlighted the need to consider such a differential diagnosis when assessing a patient with signs of pleural effusion especially in early childhood or young adult life.

Conclusion

Not all is about tuberculosis in sub-Saharan Africa. We describe an original report of a giant mature cyst teratoma that required excision with left lung decortication in a young female Congolese after she has been treated twice as having pulmonary tuberculosis. This is the first documented case in the region. Diagnosis is difficult via non-invasive procedure but clinicians should bear it in mind as a differential diagnosis to avoid delaying surgery.

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Availability of data and materials : The datasets used and/or analysed during the current study are available from the corresponding author on reasonable request.

Authors's contribution:

LB conceived and designed the study, supervised the patient follow-up, searched for reported cases, drafted the initial manuscript, and revised the manuscript after feedback. PDMCK contributed to design the study, supervised the patient care, collected the data, conceptualized the report, and reviewed and revised the manuscript. All authors read and approved the final manuscript.

Ethics approval and consent to participate

Written informed consent was given by the patient and by her guardians for their clinical records to be used in this review.

Consent for publication:

We received written informed consent from the patient and from her guardians to publish the information in this review.

Competing interests

The authors declare that they have no competing interests.

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List of figures and Table

Fig 1. Chest X-Ray (left: face, right: profile): a 15-year-old girl presenting with a large teratoma expressed by pleural effusion in South Kivu, Democratic Republic of Congo

Fig 2. Flow Chart Diagram of literature search of cases presenting a mature teratoma with pleural effusion through Medline between May 2017 and November 2017

Table 1. Demographic, Clinico-Pathology and Survival Findings of Reported Cases with Mediastinal Cystic Teratoma With Pleural Effusion

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