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Case Report

Sternotomic resection of an anterior mediastinal cystic teratoma with pericardial fistula

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Abstract

Background: Mediastinal teratomas are rare tumors and the majority of them are mature teratomas, benign lesions characterized by slow growth and low malignant potential. Signs and symptoms are infrequent; hence the diagnosis is often incidental. Complicated cases are rare and are traditionally treated with elective surgery. Our case report presents a patient with a teratoma complicated by pericardial fistulization requiring emergency surgery: the only case described in the literature.

Case presentation: A 36-year-old male with no relevant medical history reported acute thoracic pain. Chest CT scan shows a mediastinal mass (compatible with cystic teratoma) compressing the superior vena cava, the heart, and pericardial involvement. The patient was first subjected to pericardiocentesis and then an en-bloc resection of the mediastinal lesion was performed via sternotomy.

Discussion and conclusion: Mediastinal lesions require an accurate differential diagnosis, despite the diagnosis often being incidental in the absence of clinical symptoms. Conversely, if symptomatic, complicated cases can lead to life-threatening situations, where the exeresis of such lesions might require urgent, complex, and multidisciplinary surgical intervention.

Abbreviations

MGCT: Mediastinal Germ Cell Tumor; SGCT: Seminomatous Germ Cell Tumor; NSGCTM: Nonseminomatous Germ Cell Tumor of the Mediastinum; TTE: Trans-Thoracic Echocardiography

Background

Germ cell tumors are rare and, in 5% of cases, they have extragonadal mediastinal localization (MGCT), as stated by Nichols, O'Donovan, et al. [1,2]. They are a heterogeneous group of neoplasms defined by variable histology and clinical course. According to Moran and Suster's classification, depending on histopathological characteristics, they can be distinguished into teratomas, Seminomatous Tumors (SGCTM) and Nonseminomatous Tumors (NSGCTM) [3].

Approximately 70% of all GCTMs are mature teratomas and their differentiation from the three embryonic layers justifies their varied morpho-histological composition [2]. Teratomas appear on CT scans as solid lesions with heterogeneous aspects, capsulated, with fluid and calcific content with different degrees of differentiation with dental residues, hair, or sebum [4,5].

These findings are usually collateral to investigations for other causes since most of these tumors keep growing without causing any pain or with rare and non-specific symptomatology (pain, cough, dyspnea) or possibly associated with syndromic pathologies (Klinefelter) [6,7]. However, at the mediastinal level, they can cause symptoms due to mechanical compression or invasion of neighboring structures or complications with erosion and/or fistulization in the airways, pleura, or pericardial spaces. Clinical presentation

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may include the Superior Vena Cava Syndrome (SVCS) [8], Horner's syndrome, chylothorax [9], or signs and symptoms related to the production of beta-hCG [10,11]. The treatment of choice is elective surgical resection.

The case we present is a peculiar massive mediastinal teratoma that become symptomatic only due to intra-pericardial fistulization and cardiac tamponade, treated with emergency surgery.

Case presentation

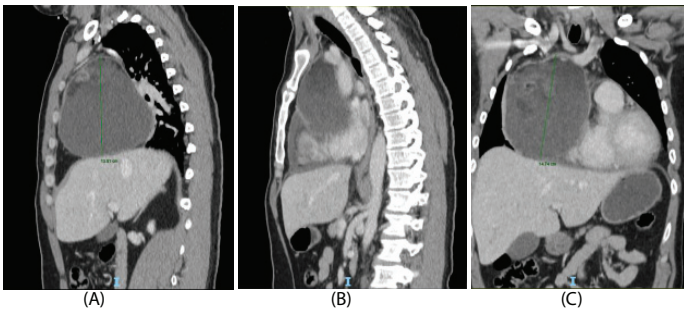
A 36-year-old male, without relevant medical history, was admitted to the emergency department with a sudden onset of anterior chest pain associated with nausea, vomiting, and arterial hypotension. None of the typical lymphoma B-symptoms such as fever, night sweats, itching, or weight loss were referred by the patient.

Chest CT scan with contrast medium shows a mediastinal mass with a maximum diameter of 13×12 cm (Figure 1A,C,E), heterogeneous (Figure 1D) with compression of the superior vena cava and pericardial effusion (Figure 1B). The lower part of the body was not included in the CT evaluation.

Blood testing detects neutrophilic leukocytosis (16.31 GB) and increased PCR (52.4). ECG reveals ST-T segment elevation with a superior concavity in DII, aVL, and V4-5-6. Trans-thoracic echocardiogram shows right para-atrial pericardial effusion, with a maximum thickness of 5 mm. A cardiologic evaluation was performed with the suggestion of close monitoring of the vital parameters and the daily control of troponine level.

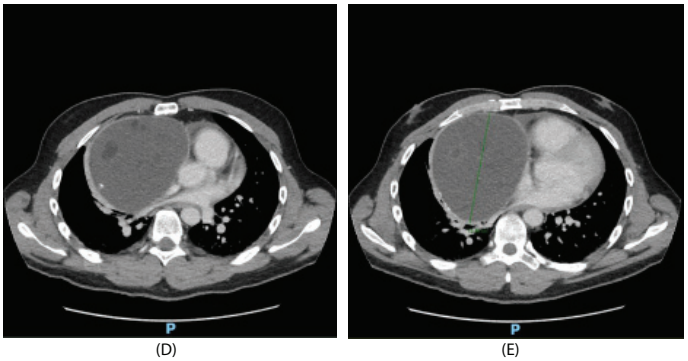
Thus, the patient was hospitalized in our intensive care unit for intensive monitoring.

Following the worsening of both retrosternal pain and hemodynamic instability with a concurrent increase in the inflammatory markers, a new TTE was performed showing an increase in the pericardial effusion. Therefore, we performed a pericardiocentesis for urgent evacuation by placing a pericardial chest tube which subsequently drained around 900 ml of purulent semi-cellular fluid. The culture tests performed on the sample were negative and an empirical antibiotic therapy with Vancomycin and Piperacillin-Tazobactam (VPT)



(A) (B) (C)

Figure 1A-C: The mass has a cranio-caudal dimension of 13 cm, deviating and compressing the main mediastinal strictures. B: CT scan image demonstrating the pericardial effusion. C: The mediastinal mass compressing and deviating the heart and the large vessels.



(D) (E)

Figure 1D,E: D: The heterogeneous aspects of the teratoma are well-highlighted. E: Image the shows the thick, contrast-enhanced wall delimitating the mass.

was established following an infectious disease specialist consultation.

On the first day after pericardiocentesis, both the clinical picture and lab results worsened with the onset of hyperpyrexia, dyspnea, and increased inflammatory markers. Remarkable at ECG was atrial fibrillation, reacutization of the clinical signs related to cardiac tamponade (pain, tachycardia, and agitation) with dyspnoea and hypotension, and an additional supply of 450 ml of cellular material from the pericardial drainage. Such a rapid deterioration of clinical conditions is an indication for urgent surgical exeresis of the lesion: the surgical procedure performed was a sternotomy with median longitudinal access.

The large mass was occupying the anterior mediastinum resulting inseparable from the pericardium, the right lung parenchyma, and the big venous vessels, due to the important perilesional fibro-inflammatory processes. We observed the symphysis with the pericardial serosa and the breach in the lowest portion of the lesion, which was responsible for the intrapericardial fistulization. The next step was opening the pericardium longitudinally from the supradiaphragmatic region to the ascending aorta, exposing a pericardial cavity filled with cellular material and epidermal derivatives (sebum, hair residues) (Figure 2A,B). At this point, we performed an en bloc resection of the mass with the pericardium, pointing out the tight adhesion of the specimen to the anonymous venous trunk and to the superior vena cava. Then we proceeded to the blunt dissection along the cleavage plane, reaching the thymic remnants cranially which were included in the en bloc, after ligation of Keynes' veins. We completed the dissection on the left pleuro-mediastinal side. On the right side, we performed an atypical resection of the anterior segment of the superior right lobe given the reported inseparability. Once we finalized the distal isolation from the mediastinal fat and the diaphragm, we concluded the specimen resection which included the known mediastinal lesion, the contiguous pericardial region, the thymic residue, the portion of lung parenchyma and of mediastinal fat (Figure 2C). After placing a right pleural drainage and a double mediastinal drainage (retrocardiac and retrosternal), we closed the sternotomic access.

After the intervention, the patient was transferred first to the ICU and then to the Department of Thoracic Surgery for further care on the second post-op day.

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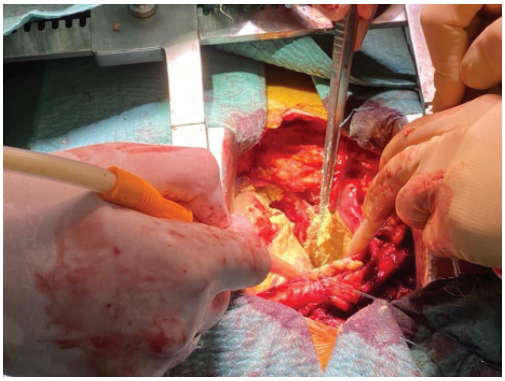
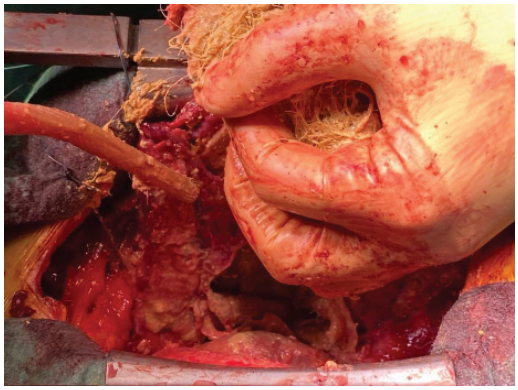


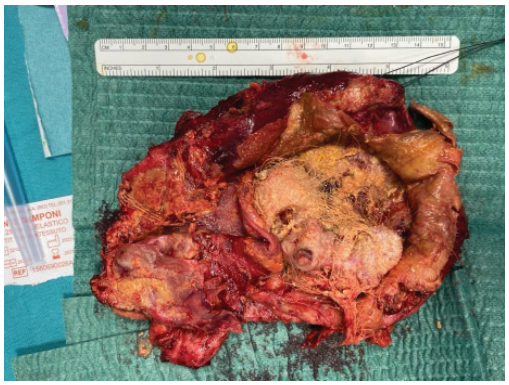
(A)

Figure 2A: Exposition of the pericardial cavity: sebum and hair can be seen.



(B)

Figure 2B: Exposition of the pericardial cavity: sebum and hair can be seen.



(C)

Figure 2C: Image of the mediastinal mass removed en-bloc.

The postoperative course was regular, void of complications, with gradual weaning of oxygen therapy and improving inflammatory markers. The removal of thoracic and posterior mediastinal drains occurred on the third post-op day, while the anterior mediastinal drainage was removed on the fifth day after surgery.

The culture tests on pericardial drain fluid, pleural fluid, blood cultures, swabs for sentinel germs, and intraoperative swabs were all negative.

None serologic tumor markers were evaluated.

Upon discharge, the patient was in good general clinical conditions: afebrile, eupnoeic on room air, with vital parameters within normal limits, and a clean surgical wound.

The definitive histological examination of the surgical specimen indicated a mature teratoma with a fibrosclerotic pseudocapsule, characterized by the presence of mature tissues, with both epithelial and mesenchymal differentiation.

Discussion

In most cases, GCTMs are clinically asymptomatic and found incidentally. The diagnosis is seldom reached secondary to the manifestation of clinical symptoms [12].

The complication into a pericardial fistula with stretching of the pericardium and concurrent inflammation led to a worsening clinical picture with the onset of symptoms associated with cardiac tamponade, despite the evacuative pericardiocentesis and the correct positioning of the intrapericardial device.

Chest CT scan with contrast medium is considered the gold standard among radiological examinations to identify the peculiar morphological characteristics of germ cell neoplasms (odontogenic and bone tissues) and helps discriminate the primary or metastatic nature of the mediastinal lesion unveiling, as in the present case, any anatomical-structural alterations possibly related to the clinic [13,14]. On CT imaging they present, predominantly in the prevascular compartment (85%), as cystic lesions characterized by a "rim-like wall enhancement" after administration of contrast medium [14]. Fat-fluid levels are typical radiological signs very specific to mediastinal teratomas, which therefore distinguishes them from other mediastinal lesions drawn in the differential diagnosis [15]. Necrosis and hemorrhages are most commonly found in lesions with malignant characteristics [16] or in immature teratomas [17]. In the present case, the oval appearance with regular margins and liquid-adipose content in the anterior mediastinum in the right paracardiac planes allowed us to reach an easy radiological diagnosis.

The intrapericardial fistulization of the sebaceous contents of the lesion made trans-thoracic echocardiography not only a diagnostic complement to the radiological images, highlighting the corpuscular nature of the lesion, but actually necessary for a proper functional evaluation of the patient and for an optimal treatment plan. Indeed, the information collected with this investigation allowed us to objectify the patient's clinical cardiological worsening, and document the presence of the fistula, helping us to understand why even after 24 hours from the position of the drainage there was still corpuscaluted-liquid to evacuate (so it was not just a pericardium abscess) and monitor the extent of the fistulization, and foster the indication for pericardial drainage placement first and the need for urgent surgery then.

In the context of GCTMs, overtly malignant lesions (seminomas, yolk sac tumors, embryonal carcinoma, etc.), are found incidentally and amenable to a diagnostic biopsy of nature, the standard therapy is neoadjuvant chemotherapy with subsequent surgical resection [18]. In pure seminomatous

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lesions, chemotherapy, and surgery may also be associated with radiotherapy [18]. In pure mature teratomatous lesions the only therapeutic option is surgery, which proves to be the gold standard regardless of the clinical picture.

In case of symptoms or complications, regardless of the benign or malignant nature, the initial treatment is merely surgical.

The rationale behind the elective surgical treatment is justified by the need for a definitive histological diagnosis, in order to promptly start a possible adjuvant treatment, but mostly to target the lesion before it could complicate or evolve into a cancerous lesion. The reported incidence of malignant transformation ranges from 0.5% to 3% and mostly into squamous cell carcinoma [19].

Our case presentation aims to draw attention to the possibility of encountering already complicated lesions("ab initio") during clinical practice, and to the potential for a further rapid deterioration of the patient's clinical conditions.

Literature suggests a total en bloc resection via open sternotomic approach (partial or total), bilateral thoracotomic with transverse sternotomy (total clam shell incision), or via lateral or postero-lateral thoracotomy. Minimally invasive surgical resection techniques, such as VATS and RATS, have given good results in terms of both oncological radicality and post-operative oncological clinical outcomes [20]. This result is achievable in selected cases and mainly for small lesions, with cleavage planes visible on radiological imaging and with an uncomplicated clinical course [21].

A recent study by Tian Z, and Liu H, et al. based on the experience in the treatment of MGCT with different surgical techniques (open and minimally invasive) on 108 patients, compared the outcomes between thoracoscopy and thoracotomy interventions for exeresis of mature teratomas of the anterior mediastinum. The minimally invasive approach was found to be superior to the classic approach in terms of intra-operative blood loss, days of hospitalization, post-operative pain, and in terms of days before chest drainage removal. Yet, the open approach remains the gold standard and is confirmed to be mandatory in the following cases: large lesions, with widespread adhesion of the tumor to the surrounding tissues; the need for en bloc removal of tissues such as the pericardium, lung parenchyma or vascular structures; preoperative symptoms (chest pain, fever, hemoptysis) [22].

In the present case, the conditions of the lesion and the acute symptomatic complication entailed the need for median sternotomy as our surgical access of choice.

Regarding histology, Ziad and Jae [23] highlighted how the most common cellular elements of a mature mediastinal teratoma are keratinizing or non-keratinizing squamous epithelium, adnexal tissues, and respiratory-type epithelium, cartilaginous and bone tissues [24]. Pancreatic tissue, whether endocrine or exocrine, has also been described in the literature, resulting predominantly in mediastinal teratomas than in teratomas located elsewhere [25,26].

Less frequently they can arise from the gastrointestinal system, neuroglia [27], smooth and skeletal muscles [28,29], parathyroid [30], liver tissue, retina [31], adrenal gland [32], thymic tissue [33-35] kidney and choroid [36].

In our case, the lesion appeared on microscopic examination as heterogeneous and made up of mature tissues, with both epithelial and mesenchymal differentiation: cartilaginous tissue, keratinizing and non-keratinizing squamous epithelium, respiratory and intestinal epithelium, skin appendages (hair follicles, sebaceous glands, apocrine and eccrine), sero-mucinous glands and finally endocrine and exocrine pancreatic parenchyma.

Conclusion

Mediastinal teratoma is a very rare tumor, and elective surgical excision remains the gold standard for asymptomatic patients. In our case, the complication with pericardial fistula and deterioration of the patient's clinical condition required a complex, which is the reason for the urgent and multidisciplinary surgical procedure. Therefore, the possibility of an abrupt symptomatic evolution of these lesions, even in young patients, may prompt the need for surgical treatment shortly after the histological diagnosis is achieved.

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