International Journal of Innovative Research in Medical Science (IJIRMS)
Volume 08, Issue 01, January 2023,
https://doi.org/10.23958/ijirms/vol08-i01/1602

Case report

Spontaneous Regression of a Thymoma with Mild Pleural Effusion: A case report

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Received 24 December 2022; Accepted 10 January 2023; Published 13 January 2023

Abstract

Introduction: Spontaneous regression of a thymoma is an unusual and rare phenomenon witnessed across the field of thoracic medicine. ***Presentation of Case:*** A 33-year-old man presented with persistent right shoulder pain and shortness of breath which started 1 month prior to hospital visit. Computed tomography of the chest was done which showed an 11.0 cm anterior mediastinal mass with bilateral pleural effusion. The man refused surgery and was discharged without any treatment. 7 months later upon evaluation, the mass had regressed more than 70% of its size with subsequent disappearance of the pleural effusion. Right thoracoscopic thymectomy was performed safely without complications. Post thymectomy, the surgical pathology report and analysis of the specimen was consistent with the diagnosis of thymoma. ***Clinical Discussion:*** Our reported case is an unusual example of a large thymoma undergoing spontaneous regression over time without any therapy provided. Previous studies have reported similar phenomena with limited insight towards the understanding of such an event. Thymomas are eventually managed by surgical resection which remains the preferred modality of treatment, most commonly via thoracoscopy consistent with our case along with past reports. ***Conclusion:*** Spontaneous regression of thymomas is widely unwitnessed in practice, thus, recognition of this rare occurrence could provide further insight in disease outcomes.

Keywords: ***Thymoma; Regression; Pleural effusion; Myasthenia gravis; Thoracoscopy***

Introduction

Thymoma is considered the most common neoplasm arising from the thymus gland, accounting for the majority of neoplasms related to the anterior mediastinum in adults after the age of 40 [1,2]. Thymomas can occur in association with various autoimmune diseases such as Myasthenia Gravis (MG), which potentially account for approximately 20-30% of all thymomas [2]. The phenomenon of spontaneous thymoma regression without therapy is rare, with very few cases reported in previous literature [2-4]. This is a report of an uncommon and rare case of spontaneous regression (SR) of a large thymoma in an adult patient. This case report has been reported in line with the SCARE Criteria [9].

Presentation of Case

A 33-year-old man not known to have any past medical or surgical history was admitted with a complaint of persistent right shoulder pain with mild shortness of breath on exertion which started 1 month prior to hospital visit. Physical examination was unremarkable. The patient was vitally stable, weighed 122 kg with a respiratory rate of 20 beats per minute, blood pressure of 146/86 mmHg and SpO2 of 96%. Lab values were within normal limits except for a high WBC level of 14.600 per microliter and Glucose level of 8.81 mmol/L. Radiography of the chest revealed a large opacity silhouetting the upper right cardiac shadow and ascending aorta suggestive of an anterior mediastinal mass. Moreover, Chest computed tomography scan (CT) was performed which revealed an anterior mediastinal mass of 11 cm x 8 cm in size with mild bilateral pleural effusion [**Figure 1**]. The mass was biopsied and sent for histopathology twice without complications under CT guidance by an interventional radiologist (IR) using 18g tru-cut biopsy needle, with the first result being non-conclusive, and the second being suggestive of a thymoma. Precise histological interpretation was difficult due to extensive necrosis, fibrosis and cystic degeneration. The patient was provided the opinion of undergoing a thymectomy, though, he refused surgery and wanted to seek secondary opinion, and was discharged from the hospital without any medications or treatment. Seven months later upon follow up in the clinic, the patient underwent a second CT scan of the chest which revealed spontaneous regression of approximately 70% of the mass size without any applied treatment or intervention [**Figure 2**]. Moreover, the patient agreed to the opinion of surgery and was scheduled for minimally invasive right thoracoscopic thymectomy. Under general anesthesia, a total of 3 ports were inserted in the right side, two 5mm and one 10mm port with no adhesions observed. The thymus gland was then identified and dissected all around with preservation of the right phrenic nerve, superior vena cava (SVC) and innominate veins. A 5mm port was also inserted in the left side with identification and preservation of the left phrenic nerve. Complete dissection and resection of the thymoma was achieved smoothly with no major complications, followed by right chest tube insertion and fixation. Post op, the patient recovered well without morbidities, pain management was sought with encouragement of mobilization and use of incentive spirometry for optimal recovery. Surgical pathology report of the specimen revealed a large area of fibrosis, macrophages reaction and hemosiderin deposition. Immunohistochemical findings showed immature T-cells positive for CD3, CD1a and CD99.

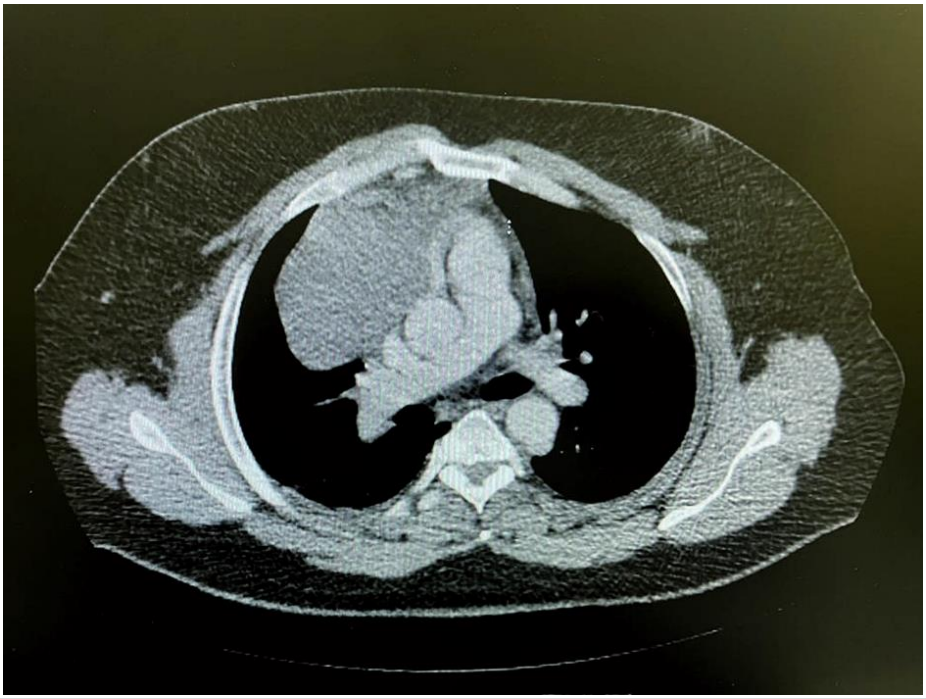

Figure 1: Chest CT performed at the initial visit revealing an 11.0 cm anterior mediastinal mass with mild bilateral pleural effusion.

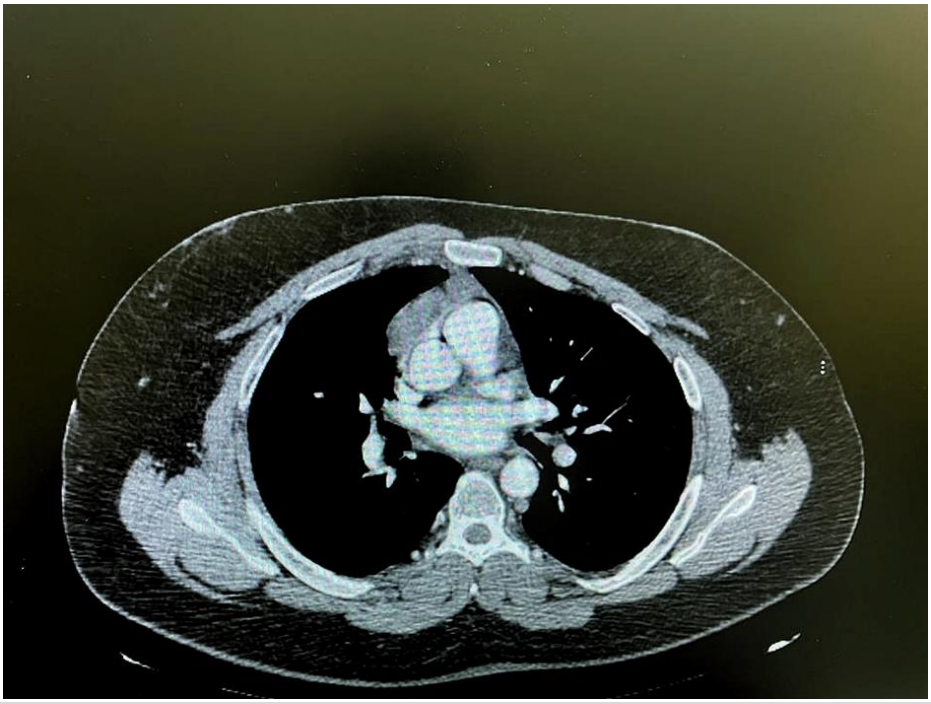

Figure 2: Chest CT repeated 7 months later showing almost 70% spontaneous regression of the anterior mediastinal mass.

Discussion

Thymomas in the adult population are considered the most common neoplasm affecting the anterior mediastinum, with surgical resection remaining the treatment of choice. Thymomas are often seen in adults after the age of 30 years with high association to myasthenia gravis (MG) and in some cases can be highly invasive if left untreated with the possibility of second malignancy [1]. In this present case, the patient denied symptoms of MG. Although considered unusual, spontaneous regression of mediastinal neoplasms have been witnessed in the past involving cases of renal cell carcinoma, malignant melanoma, and neuroblastoma [4]. The mechanisms in which a tumor can regress can be either via necrosis, apoptosis or rupture [3,4]. Nonetheless, SR of thymoma without therapy is a rare phenomenon, with some cases witnessing SR upon treatment with corticosteroids [4,5]. The mechanism in which thymomas spontaneously regress remain unclear, with several reports suggesting that SR could be due to extensive necrosis and vascular compromise or absorption of intratumoral components [2-4]. In our case, histological interpretation of the biopsied specimen was a challenge due to extensive necrosis, fibrosis, and cystic degeneration. The cause of necrosis itself has not been clearly determined, though, one study stated that rapid tumor growth could result in massive necrosis due to increased internal pressure [4]. Generally, thymomas are asymptomatic except those associated with myasthenia gravis in approximately 20-30% of cases [2]. Moreover, almost all cases of thymomas witnessing SR in past reports have been associated with symptoms ranging from fever, chest pain and shortness of breath with findings of pleural effusion, consistent with our report [2-4]. These clinical findings associated with elevation of serum inflammatory markers are mainly consistent with extensive necrosis [4]. The mechanism underlying these symptoms could be due to the rapid progression in size and enlargement of the mass, compromising vascular supply with creation of an inflammatory

reaction ultimately resulting in a pleural effusion [2-4]. In our case, the mild bilateral effusion resolved subsequent with SR of the thymoma. Thymomas are generally managed by complete surgical resection using open techniques or minimally invasive approach with video-assisted thoracoscopy [7]. The minimally invasive intercostal approach was used in the present case without complications, with single port techniques gaining popularity provided the advantages of good visibility assisting with intraoperative hemostasis, reduced postoperative pain and recovery length [8]. During the postoperative period, histopathology studies and subtyping of the resected mass could provide great value in further treatment decisions which may involve introducing radiotherapy for thymomas with more aggressive features. Markers considered to be useful for immunohistochemical characterization of thymomas include cytokeratins 19 and 20, P63, P40, and others such as CD5 and CD20 [6].

Conclusion

We herein present a rare case of a large thymoma exhibiting spontaneous regression, although the occurrence of this phenomenon is unusual, possible malignancy should be considered, with surgical intervention and resection of the remaining lesion as the mainstay treatment method.

Abbreviations

MG: Myasthenia Gravis
SR: Spontaneous regression
CT: Computed tomography
IR: Interventional radiologist
SVC: Superior vena cava

Declarations

Ethics approval and consent to participate

Not applicable

Consent for publication

Written informed consent was obtained from the patient for publication of this case report and any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Availability of data and materials

The case history and reports used and/or analyzed during the current study are available from the corresponding author on reasonable request.

Competing interests

The authors declare that they have no competing interests

Funding

None

Authors contributions

Abdulaziz Alnassar contributed to the conception and design of the study. He performed the collection and interpretation of data and wrote the manuscript.

Acknowledgements

Not applicable

Research registration

N/A

Guarantor

I, as the author of the study, accept full responsibility for the work and/or the conduct of the study, had access to the data, and controlled the decision to publish.

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