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CASE REPORT

Multilocular thymic cysts can be easily misdiagnosed as malignant tumor on computer tomography: A case report

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Abstract

BACKGROUND

Multilocular thymic cyst (MTC) is a rare mediastinal lesion which is considered to occur in the process of acquired inflammation. It is usually characterized by well-defined cystic density and is filled with transparent liquid.

CASE SUMMARY

We report on a 39-year-old male with a cystic-solid mass in the anterior mediastinum. Computer tomography (CT) imaging showed that the mass was irregular with unclear boundaries. After injection of contrast agent, there was a slight enhancement of stripes and nodules. According to CT findings, it was diagnosed as thymic cancer.

CONCLUSION

After surgery, MTC accompanied by bleeding and infection was confirmed by pathological examination. The main lesson of this case was that malignant thymic tumor and MTC of the anterior mediastinum sometimes exhibit similar CT findings. Caution is necessary in clinical work to avoid misdiagnosis.

Key Words: Multilocular thymic cyst; Computer tomography; Misdiagnosis; Hemorrhage; Infection; Case report

Core Tip: Multilocular thymic cyst (MTC) is a rare mediastinal lesion, which is considered to occur in the process of acquired inflammation. It is usually characterized by well-defined cystic density and is filled with transparent liquid. Here we reported on a 39-year-old male with a cystic-solid mass in the anterior mediastinum. Computer tomography (CT) imaging showed that the mass was irregular with unclear boundaries. After injection of contrast agent, there was a slight enhancement of stripes and nodules. According to the CT findings, it was diagnosed as thymic cancer. After surgery, MTC accompanied by bleeding and infection was confirmed by pathological examination.

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INTRODUCTION

Thymic cysts account for about 1%-5% of all mediastinal masses[1-3], mainly located in the anterior mediastinum, with a few cases located in the neck[4]. Most of them are circular, elliptical, and irregular in shape, with or without separation. The clinical symptoms of thymic cysts depend upon the location. If the surrounding tissues and organs of the cyst are compressed, breathing difficulties, coughing, thymic pain, and Horner's syndrome may occur. Most thymic cysts have cystic density, a clear boundary and are easy to be diagnosed by medical imaging[5-8]. Some thymic cysts are difficult to diagnose before surgery and can be misdiagnosed as thymoma based on their location and computer tomography (CT) findings. Here, we reported on a case of multilocular thymic cyst (MTC) with hemorrhage and infection in a 39-year-old patient, which was misdiagnosed as thymic carcinoma.

CASE PRESENTATION

Chief complaints

A 39-year-old man came to our hospital with the symptoms of fever, headache and occasional palpitations.

History of present illness

The patient had the symptoms of fever, headache and occasional palpitations.

History of past illness

There was a history of hyperglycemia and no history of muscle weakness, joint pain, chest trauma or any surgery. Human immunodeficiency virus (HIV) was negative.

Laboratory examinations

The random blood glucose was 21.16 mmol/L. Laboratory tests showed that lactate dehydrogenase was 572 U/L, α-hydroxybutyrate dehydrogenase was 441 U/L and the leukocyte count was 15.7 × 10^9/L. Electrocardiogram showed atrial fibrillation, ventricular tachycardia, and elevated T-waves. All tumor markers were negative (Table 1).

Imaging examinations

After chest X-ray examination, a mediastinal mass was found (Figure 1). CT examination showed a mass in the anterior mediastinum, with a size of 5.6 cm × 11.3 cm × 10.2 cm. It had uneven density, showing a mixture of cystic density and solid density, with CT values ranging from 30 to 50 HU. Its shape was irregular and the boundary was unclear. No calcifications were observed. After contrast agent injection, a slight enhancement of stripe and nodular shape were found. The mass was adjacent to the ascending aorta, superior vena cava and left brachial vein. The boundary with the above structure was unclear, but the vascular lumen was not invaded. Bilateral pleural effusion was found (Figure 1). According to CT findings, it was diagnosed as a thymic cancer.

FINAL DIAGNOSIS

This patient was finally diagnosed with acute myocarditis, acute upper respiratory tract infection, MTC with infection in the anterior mediastinum, and diabetes.

Table 1 Tumor markers of the patient

Tumor marker	ng/mL
Alpha-fetoprotein	1.5
Carcinoembryonic antigen	3
Carbohydrate antigen 19-9	48.4
Neuro-specific enolase	24.8
Ferritin	1000
Tumor specific factors	66.2

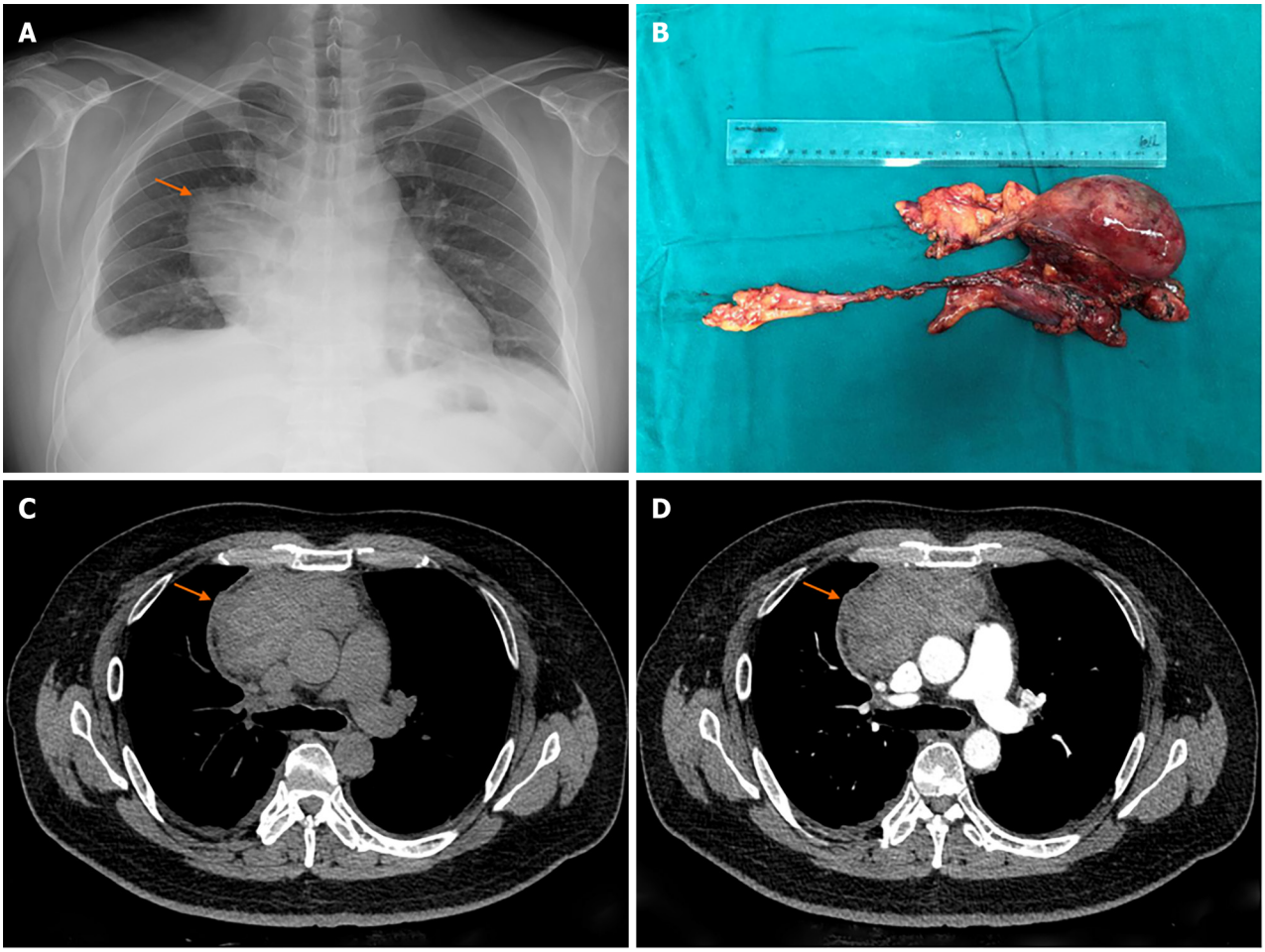

Figure 1 Radiology and macroscopic pathological image. A: Chest X ray image; B: Macroscopic pathological image; C: Plain computer tomography (CT) image; D: CT image after contrast agent injection.

TREATMENT

Myocarditis was diagnosed based on the abnormal electrocardiogram, high levels of lactate dehydrogenase and α-hydroxybutyrate dehydrogenase, and the clinical symptoms of fever and palpitations. After treatment with cefuroxime, fructose, sodium diphosphate, betaloc, insulin and acarbose, the lactate dehydrogenase and α-Hydroxybutyrate dehydrogenase levels decreased and the leukocytes returned to normal. The mediastinal mass resection was performed 13 d after admission. The mass consisted of a cystic part and a solid part, attached to the adjacent tissues and surrounded the right brachiocephalic vein.

After surgery, histopathological examination was performed. Squamous epithelium, cholesterol crystals, bleeding and reactive lymphoid hyperplasia with CD1 α (-), CD20 (partially positive), CD3 (partially positive), CD5 (partially positive), CD99 (+), CK (+), CK7 (partially positive), CK19 (+), CK5/6 (+), EMA (+), Ki-67 (5% +), p63 (+), CD117 (-) were found and MTC was diagnosed (Figure 2).

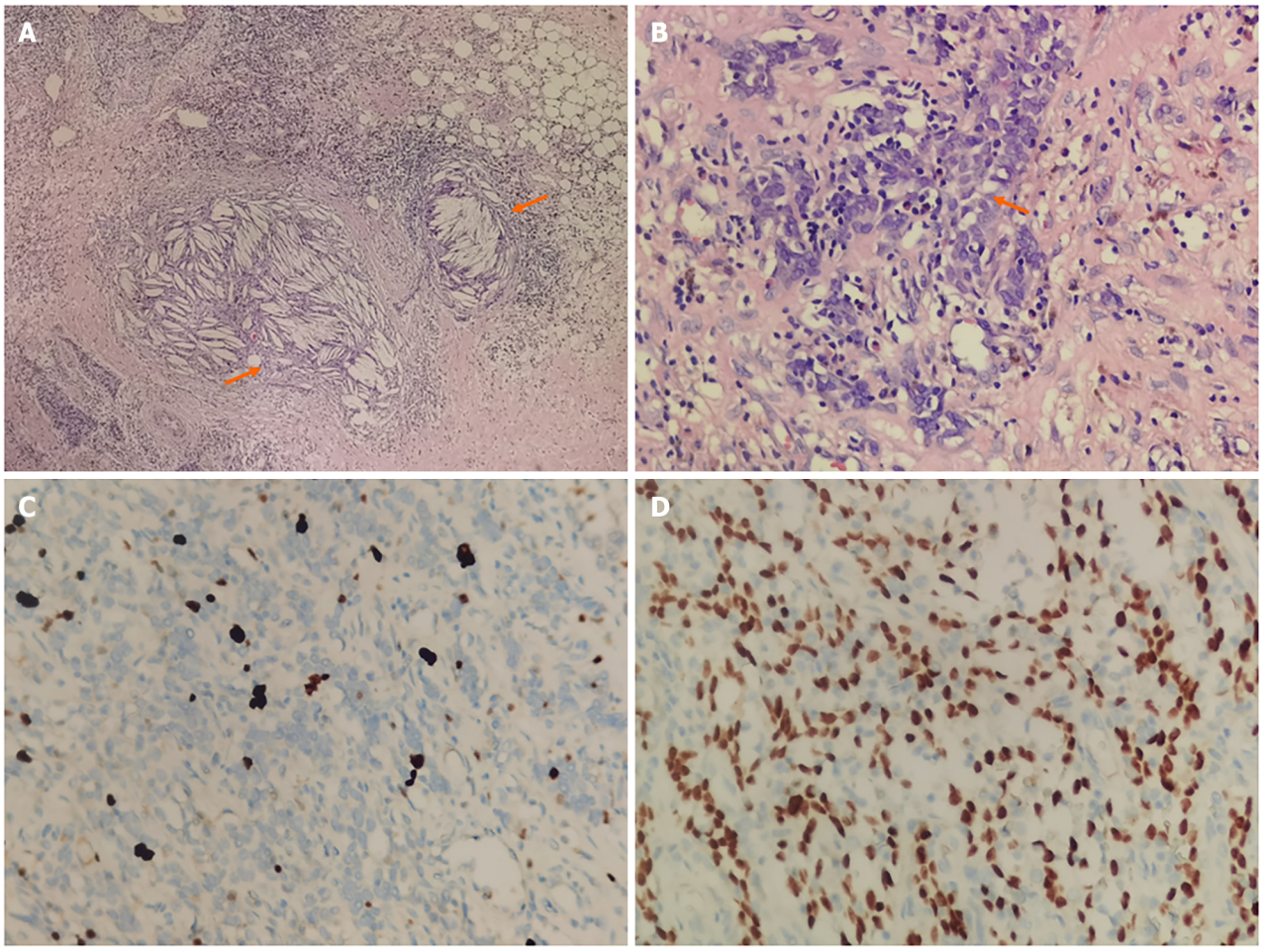

Figure 2 Histopathological picture of this case of multilocular thymic cyst. A: The cholesterol crystals in multilocular thymic cyst (MTC); B: Squamous epithelium and lymphoid hyperplasia; C and D: Immunohistochemistry of Ki-67 and p63 in MTC. The Ki-67 was 5% positive and p63 was positive.

OUTCOME AND FOLLOW-UP

After treatment, all symptoms were improved and the patient was discharged successfully. The patient was tracked for 9 months and no signs of recurrence was found (Figure 3).

DISCUSSION

According to the morphology, thymic cysts could be divided into two subtypes: monocular thymic cyst and MTC. Monocular thymic cysts are common, while MTC is rare. MTC is generally considered to be formed during inflammation and usually accompanied by immune deficiency diseases (such as HIV), autoimmune diseases (such as arthritis or Sjögren syndrome[9,10]) or thymic trauma[11]. Therefore, MTC is also called an acquired thymic cyst. It is usually filled with transparent liquids, and sometimes may contain cloudy liquids or gelatinous substances due to bleeding. The main histologic features of MTC included the following: multiple cystic cavities, the wall of which was composed of fibrous wall, squamous epithelium, columnar epithelium or cubic epithelium; acute and chronic inflammation with fibrovascular proliferation; necrosis, hemorrhage, cholesterol granuloma and reactive lymphoproliferative[9]. The pathological diagnostic criteria of the World Health Organization are the gold standard for diagnosing MTC. If MTC is small and asymptomatic, surgery is not necessary, which can protect patients from surgical trauma and reduce psychological stress. Imaging evaluation plays an important role in the diagnosis, treatment and decision-making with MTC.

Most MTCs showed cystic density with separation, no enhancement, and no complicated pleural effusion. The CT manifestation of this case was a mixed density mass with cystic and solid components. After injection of contrast agent, there was a slight enhancement of striped and nodular shape. It is very difficult to differentiate it from other mediastinal tumors, especially thymoma. According to the surgical and pathological results, the solid density was formed by bleeding, turbid liquid or colloidal material. The striped and nodular enhancement were caused by cyst wall, septum and reactive lymphoid hyperplasia. Previously, Jin *et al*[12] has reported 13 cases of thymic cyst with solid density which were misdiagnosed as thymoma. Marvasti *et al*[13] have reported five mediastinal cysts containing viscous liquid, which showed solid density on CT. The solid density in thymic cysts may be caused by bleeding, lymphoid tissue, viscous liquid containing high levels of protein and hyperplastic thymic tissue at different stages. The solid density has also been

Table 2 Reported cases of multilocular thymic cyst containing solid density in adults

Ref.	Number of cases	CT value	Histopathologic of solid density
Jin *et al* [12]	28	38.61 ± 15.01 HU	N
Matsumoto *et al*[14]	1	NA	Lymphoid hyperplasia
Chalaoui *et al*[15]	1	35 HU	Hemorrhage
Kim *et al*[16]	1	NA	Lymphoid hyperplasia and fibrous septa
Mohakud *et al*[17]	1	60-80 HU (high density areas)	Hemorrhage
Nagata *et al*[19]	1	NA	Cholesterol granuloma
Choi *et al*[18]	8	Ranged -20 to 17 HU	Thick inflammatory thymic tissue, thymic hyperplasia and calcification
Izumi *et al*[7]	4	NA	Foreign body granulomas
Ridder *et al*[20]	2	NA	Hemorrhage and cholesterol granuloma
Damaskos *et al*[1]	1	NA	Calcification and fragments of thymic tissue

CT: Computer tomography; NA: Not applicable.

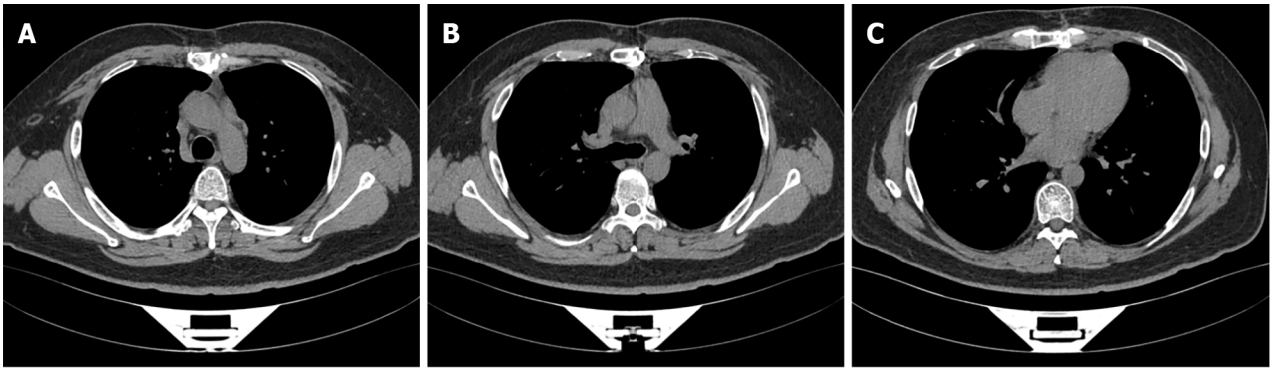

Figure 3 Rechecked after 9 months and no signs of recurrence.

reported as cholesterol granuloma and calcification[1,7,14-20] (Table 2). In this case, the boundary of the lesion was not clear, which was easy to be misdiagnosed as a malignant tumor. Usually, unclear boundary was considered as an invasive marker of malignant tumors. According to the histopathological results, there was bleeding, inflammation and reactive lymphoproliferation in this lesion. The unclear boundary might be caused by these factors, which also explains the significant increase of leukocytes. Pleural effusion is also a sign of malignant tumor, which misleads the diagnosis of this disease to a certain extent. Previously, there was no report of MTC complicated with pleural effusion. In this case, the bilateral pleural effusion may be caused by acute myocarditis. However, due to the lack of pleural fluid cytology results, the cause of pleural effusion was not clear.

CONCLUSION

Through this case, we found that MTC can have both cystic density and solid density simultaneously. Sometimes, due to combined inflammation, bleeding and other reasons, the edge may be unclear and pleural effusion may occur. In these conditions, it is easy to be misdiagnosed with malignant tumors. Full understanding of the special manifestations of the disease, combined with clinical and laboratory examination, is very important for accurate diagnosis.

FOOTNOTES

Co-first authors: Jun Sun and Qing-Ning Yang.

Co-corresponding authors: Yi Guo and Chuan-Ming Li.

Author contributions: Sun J and Yang QN drafted the manuscript; Guo Y and Li CM performed the final approval of the manuscript; Zeng P, Ma LY, Kong LW and Zhao BY collected the clinical, pathology and computer tomography data; All authors read and approved

the final manuscript. Sun J and Yang QN contributed equally to this work as co-first authors; Guo Y and Li CM contributed equally to this work as co-corresponding authors. The reasons for designating Guo Y and Li CM as co-corresponding authors are: First, the research was performed as a collaborative effort, and the designation of co-corresponding authorship accurately reflects the distribution of responsibilities and burdens associated with the time and effort required to complete the study and the resultant paper; Second, Guo Y and Li CM contributed efforts of equal substance throughout the research process. In summary, we believe that designating Guo Y and Li CM as co-corresponding authors is fitting for our manuscript.

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