ELSEVIER

Contents lists available at ScienceDirect

Respiratory Medicine Case Reports

journal homepage: http://www.elsevier.com/locate/rmcr





Rare case of cavernous hemangioma of the thymus

Samah Joobeur ^a, Ahmed Ben Saad ^{a,*}, Asma Migaou ^a, Manel Njima ^b, Asma Achour ^c, Nesrine Fahem ^a, Saousen Cheikh Mhamed ^a, Naceur Rouatbi ^a

- ^a Pulmonology Department, Fattouma Bourguiba Hospital, Monastir, Tunisia
- ^b Department of Pathology, Fattouma Bourguiba Hospital, Monastir, Tunisia
- c Radiology Department, Fattouma Bourguiba Hospital, Monastir, Tunisia

ARTICLE INFO

Keywords: Cavernous hemangioma Surgery Thymus Mediastinal neoplasm Vascular malformations

ABSTRACT

Cavernous hemangioma (CH) of the thymus is an extremely rare congenital venous malformation. Related symptoms are non-specific and patients are often asymptomatic. The diagnosis is difficult to make either by non-invasive or mini-invasive procedures. Surgical resection is usually required for diagnosis and treatment. We report a case of a 46-year-old men with an incidental finding of an anterior mediastinal tissue mass on chest computed tomography scan. A complete surgical resection of the mass was performed. Histopathological examination concluded to a thymic CH.

1. Introduction

Cavernous hemangioma (CH) is a congenital venous malformation which was initially described by Lungenschmid in 1990 [1]. This malformation has the potential to develop in all parts of the body especially in the cutaneous and mucosal tissues of the face, mouth, and limbs particularly in children. Whereas it is very rarely seen in the thymus. the incidence of hemangioma, including cavernous hemangiomas, is 0.5% or less in most studies of mediastinal tumors [2]. The diagnosis is difficult to make promptly because both invasive and noninvasive examinations mostly fail to distinguish it from other tumors of the mediastinum. Surgical resection is necessary to provide material for histopathologic diagnosis. We report a case of an incidental finding of a thymus cavernous hemangioma in a 46-year-old male diagnosed after surgical resection and histopathological examination.

2. Case presentation

A 46-year-old men presented to our hospital for a physical examination after a chest trauma (public road accident). He was complaining of chest pain that appeared after the trauma. The patient was asymptomatic before the accident and had only a history of cigarette smoking. Physical examination revealed normal breath sounds in both lung fields. Complete blood count and blood biochemistry were within normal

limits. Chest computed tomography (CT) scan revealed an hypodense left anterior mediastinal tissue mass measuring 2.3 * 1.9 cm in size (Fig. 1). A surgical approach was recommended to provide a definitive diagnosis and treatment of the mass because a mediastinal tumor such as thymoma was suspected. Under general anesthesia, left thoracotomy followed by a complete surgical resection of the mass was performed. A histopathological examination using hematoxylin and eosin (HE) staining revealed thymic tissue presenting a vascular proliferation formed by thick-walled dilated vessels, lined by a regular endothelium (Fig. 2). The diagnosis of a CH in the thymus was made. The patient was discharged 5 days after surgery without complications.

3. Discussion

The International Society for the Study of Vascular Anomalies (ISSVA) classified CH as a congenital venous malformation in the low-flow lesion category [3]. It is an uncommon abnormality and the thymic location is extremely rare. To the best of our knowledge, only ten cases of cavernous hemangioma of the thymus have been reported in English literature [4].

Histologically, hemangioma originates from residual embryonic vascular cells. It is caused by abnormal vascular development occurring before birth [4]. CH occurs mainly in the liver (30% of known cases), as well as in the brain and skin [5]. Vascular proliferations of the

^{*} Corresponding author. Pulmonology Department, Fattouma Bourguiba Hospital, 5000, Monastir, Tunisia.

E-mail addresses: samah.joobeur@rns.tn (S. Joobeur), ahmedbensaad28@yahoo.fr (A. Ben Saad), migaou.asma@gmail.com (A. Migaou), manelnjima@yahoo.fr (M. Njima), doc.asma.achour@hotmail.com (A. Achour), nesrinefahem@yahoo.fr (N. Fahem), saoussen.cheikh@gmail.com (S.C. Mhamed), naceur.rouatbi@rns.tn (N. Rouatbi).

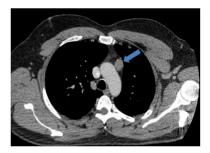


Fig. 1. Chest CT scan (contrast enhanced axial image, mediastinal window) revealing an hypodense left anterior mediastinal tissue mass with a thin peripheral calcification (blue arrow). (For interpretation of the references to colour in this figure legend, the reader is referred to the Web version of this article.)

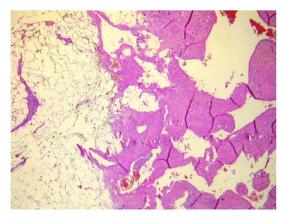


Fig. 2. Thymic tissue presenting a vascular proliferation formed by thick-walled dilated vessels, lined by a regular endothelium (HEx40).

mediastinum include arteriovenous hemangiomas, venous hemangiomas, glomus tumors, hemangiomas, angiofibromas, hemangiopericytomas, angiolipomas, and CH [6].

Reviewing the reported literature, CH can occur at all ages with no gender predominance [4]. The majority of patients were asymptomatic, and the tumor was discovered incidentally as is the case with our patient. Patients may present with different symptoms, like chest pain, cough or dyspnea [4]. These symptoms are due to the growth of the mass and the pressure on the surrounding organs.

Imaging examinations including chest CT scan and magnetic resonance imaging (MRI) are beneficial for assessing the tumor size, border and vascular condition but they don't allow to determine whether a mass is benign or malignant. Enhanced CT may be very effective, as it can reveal a phlebolith in some cases of venous malformation [4,7]. In our case, we noticed a thin peripheral calcification on CT scan.

When occurring in the thymus, CH is toughly diagnosed either by invasive or non-invasive explorations. Indeed, there is a risk of bleeding by biopsy of the mass. Also, it is difficult to distinguish CH from other tumors of the mediastinum such as a thymoma [7,8]. Thus, a CH requires often a resection for diagnosis and treatment. Histopathological findings generally show cavernous or cystic venous channels with a flat single layer of endothelial cells without atypia or mitosis [7].

The choice of treatment depends on the related organ. It can include embolization, sclerotherapy, and surgical resection [4]. However,

according to the published literature concerning mediastinal location, complete resection is considered the most successful and effective treatment [4,7,8]. The surgical approach may be sternotomy, thoracotomy, or a video-assisted thoracoscopic surgery (VATS) depending on the mass size and the local resources [4]. A complete extirpation of the lesion is the best therapeutic choice. In fact, in case of incomplete resection, the residual tissue has the potential for recurrence with abnormal vessel dilation and regeneration [7]. Surgical resection is usually associated with excellent long-term prognosis [4]. Our patient had a complete surgical resection with favorable outcomes.

4. Conclusion

We describe this very rare case of thymic cavernous hemangioma that was definitively diagnosed after surgery. This congenital malformation is often discovered incidentally. Complete surgical resection represent a safe and effective treatment. Other reported cases are needed for a better understanding of this abnormality with a standardized therapeutic approach.

Funding

This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

Declaration of competing interest

None.

Acknowledgements

none.

Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.rmcr.2019.100986.

References

- D. Lungenschmid, R. Schöpf, O. Dietze, A. Furtschegger, Computerized tomography diagnosis of cavernous thymus hemangioma and differentiation from other mediastinal hemangiomas, Rontgenblatter 43 (7) (1990 Jul) 301–304.
- [2] N. Yoshino, D. Okada, H. Ujiie, H. Akiyama, Y. Nishimura, K. Koizumi, et al., Venous hemangioma of the posterior mediastinum, Ann. Thorac. Cardiovasc. Surg. 18 (2012) 247–250, https://doi.org/10.5761/atcs.cr.11.01706.
- [3] O. Enjolras, Classification and management of the various superficial vascular anomalies: hemangiomas and vascular malformations, J. Dermatol. 24 (1997) 701–710, https://doi.org/10.1111/j.1346-8138.1997.tb02522.x.
- [4] C. Zheng, F. Zhang, S. Tu, X. Zhang, C. Zhao, Cavernous hemangioma of the thymus: a case report and review of the literature, Medicine (Baltim.) 97 (30) (2018), e11698, https://doi.org/10.1097/MD.000000000011698.
- [5] A. Yamazaki, H. Miyamoto, Y. Saito, H. Matsuzawa, Y. Sakao, Y. Anami, Cavernous hemangioma of the anterior mediastinum: case report and 50-year review of Japanese cases, Jpn. J. Thorac. Cardiovasc. Surg. 54 (5) (2006) 221–224.
- [6] G.P. Romeo, P.N. Fiedler, C.S. Hecht, D. Nuñez, Thrombosed cavernous hemangioma arising in cervical ectopic thymus tissue: a case report, Conn. Med. 76 (7) (2012) 401–404.
- [7] N. Ose, Y. Kobori, Y. Takeuchi, Y. Susaki, S. Taniguchi, H. Maeda, Cavernous hemangioma in the thymus: a case report, Surg Case Rep 2 (1) (2016) 10, https://doi.org/10.1186/s40792-016-0137-6.
- [8] C. Shen, Y. Liang, H. Xu, L. Liu, G. Che, Cavernous hemangioma of thymus misdiagnosed as thymoma: a case report, World J. Surg. Oncol. 12 (2014) 323, https://doi.org/10.1186/1477-7819-12-323.