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Chest wall resection for a giant angiomatosis lesion: A case report

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ABSTRACT

INTRODUCTION: Angiomatosis is a rare and benign vascular lesion which can be located in various sites throughout the body, most commonly diagnosed in females in the first two decades of life.
PRESENTATION OF CASE: A 54-year-old female with no previous medical history presented with significant pain and discomfort associated with a 12.3 × 11.3 × 4.4 cm posterior chest wall mass invading the 4th and 5th ribs and extending into the thoracic cavity.
DISCUSSION: Angiomatosis is a benign vascular lesion that can affect any soft tissue in the body. Typically, it has a female predilection and tends to involve the lower extremities. It is histologically characterized by a proliferation of blood vessels of varying sizes and has a high recurrence rate after excision. Significant blood loss can occur during resection.
CONCLUSION: Management of these lesions is challenging based on the infiltrative and vascular nature of the disease. Neoadjuvant therapy can be considered in select cases. Close surveillance is recommended due to high rates of recurrence.

1. Introduction

Angiomatosis is a rare and benign vascular lesion which can be located in various sites throughout the body. Angiomatosis lesions have been described in the breast, bone, cerebro-meningeal, retinal and other locations [1–4]. Overall, however, this lesion is extremely rare. We present a case of complete resection of a giant chest wall angiomatosis lesion. This work has been reported in line with the SCARE criteria and cites the corresponding paper in the reference section [5].

2. Case report

A 54-year-old female with no previous medical history presented to our institution with significant pain and discomfort associated with a large protuberant chest wall mass. Computed tomography (CT) scan showed the lesion to be 12.3 × 11.3 × 4.4 cm. The lesion invaded the 4th and 5th ribs and extended into the thoracic cavity (Fig. 1). Percutaneous biopsy was non-diagnostic but showed no evidence of malignancy.

The patient subsequently underwent a resection of the mass with en bloc chest wall resection. The surgery was remarkable for the very large feeding vessels arising from the chest wall and the vascular nature of the mass. Portions of the 4th, 5th and 6th ribs were removed with the mass. The patient tolerated the procedure well and was ultimately discharged in good condition. Her pain was significantly reduced after surgery.

Pathologic examination revealed angiomatosis with no malignant elements (Fig. 2). The mass measured 14 cm in maximum diameter and margins were negative for disease.

3. Discussion

The first case report of angiomatosis was published in 1927 by Snell and affected the retina of a patient [6]. Angiomatosis can affect any soft tissue in the body and is defined as a benign vascular lesion that usually presents in the first two decades of life. Angiomatosis typically has a female predilection and tends to involve the lower extremities [7]. It is histologically characterized by a proliferation of blood vessels of varying sizes. These blood vessels appear in clusters and are intimately associated with a large amount of adipose tissue [8].

Angiomatosis is a lesion which is composed of a widespread proliferation of blood vessels surrounded by fatty tissue. Histologically there is a haphazard collection and cluster of vessels of varying sizes associated with an abundance of fat. The diagnosis of angiomatosis

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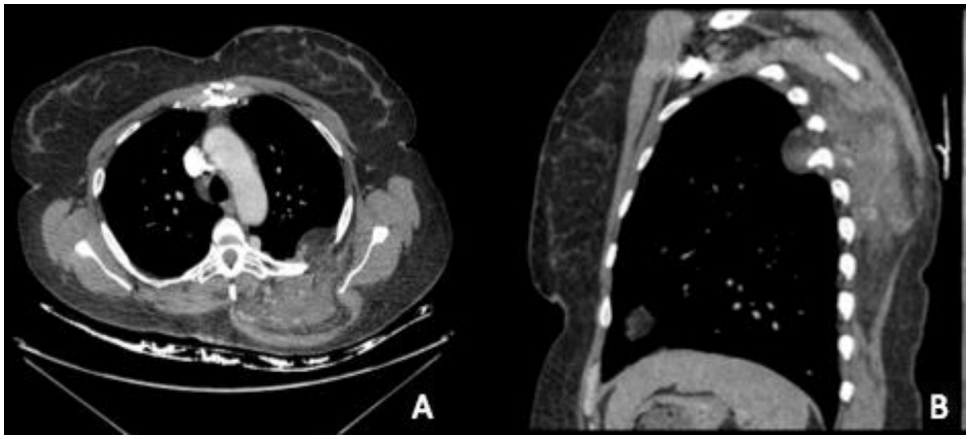

Fig. 1. CT scan showing chest wall angiomatosis lesion with intrathoracic extension in A) axial and B) sagittal views.

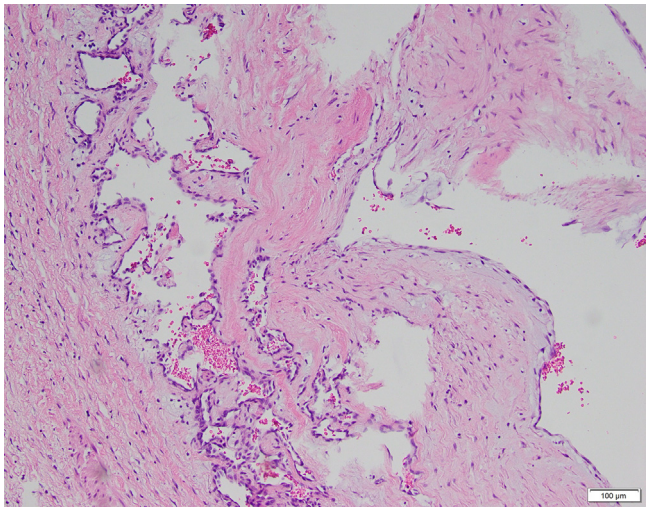

Fig. 2. High-powered hematoxylin and eosin view x 400 showing angiomatosis lesion.

should be suspected when a lesion has a tremendous amount of blood vessels coursing through it, especially when there are very large blood vessels within the central portion of the lesion [8]. A tissue diagnosis can be achieved with a percutaneous biopsy, but these biopsies should be performed using image guidance if possible to help guide the needle away from some potentially large blood vessels within the lesion. It is our opinion that percutaneous biopsy should be attempted when the lesion is very large and the technical difficulty of the resection may be quite difficult, as some reports have detailed very challenging surgeries to resect these lesions [9].

Angiomatosis can mimic hematomas, vascular malformations and metastatic tumors [10]. Although a benign entity, management of these lesions can be challenging because the recurrence rate after resection is estimated to be 90%. Complete excision is the preferred approach, and en bloc resection of adjacent structures should be performed if necessary to achieve negative margins [9]. In our patient, resection of 3 ribs were required to remove the lesion completely. Also, these lesions are usually associated with significantly enlarged collateral vessels and there can be significant blood loss associated with resection. Our patient had very enlarged collateral blood vessels, measuring almost one centimeter in diameter, and care had to be taken to control these vessels during division of the ribs. Coordination with the anesthesia team is important to prepare for the possibility of significant blood loss.

Though surgical resection is the treatment of choice, non-surgical options are available if patient is a poor surgical candidate or if the lesion is unresectable given its location [11]. Embolization can be done to reduce blood supply to the mass and potentially reduce its size [12]. Given the rarity of this tumor, the absolute efficacy of embolization is unknown. Other options for management of angiomatosis include angiogenesis inhibitors and bisphosphonates when bone is involved [13]. If there is widespread disease then thalidomide, zoledronate and interferon alpha can be considered [14]. Radiation can also be considered preoperatively for very large lesions [11]. Because angiomatosis was very low on our differential diagnosis, we opted for upfront surgical resection. Given the size and location of the lesion, however, if the diagnosis had been known preoperatively we would have favored neoadjuvant treatment with one of these agents.

The overall prognosis for angiomatosis varies depending on the size of the lesion and the ability to resect the lesion completely. Though benign, these lesions can create morbidity from local complications. Small lesions have an excellent prognosis because they can be resected completely with minimal morbidity. Larger lesions have a very high recurrence rate, especially when only partially removed [15].

4. Conclusion

Angiomatosis is an extremely rare disease. Management of these lesions is challenging based on the infiltrative nature of the disease. Surgical resection is the ideal treatment, but preoperative treatment with a variety of agents should be considered if reducing the size of the lesion will appreciably lessen the extent and morbidity of surgical resection. Close surveillance is recommended since the overall recurrence rate is quite high.

Conflicts of interest

None.

Funding

None.

Ethical approval

Our report is exempt from ethical approval from our institution.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request. Identifying details were withheld if they were not essential.

Author contribution

Ulises Garza contributed to the writing of the manuscript. Navyatha Mohan contributed to the writing of the manuscript. Catherine Miller prepared the slides and interpreted the specimen. Suimin Qiu prepared the slides and supervised the histologic analysis. Ikenna Okereke, contributed to the writing of the manuscript, organization of the report and was the supervisor of this project.

Registration of research studies

N/A.

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