

Epithelioid angiosarcoma arising from an oleothorax: An unreported case report

Karl Jackson, Parag Narkhede, Avinash Aujayeb

ABSTRACT

We describe the case of a male patient with a previous oleothorax who presented with constitutional symptoms and a mass growing out of the oleothorax. Biopsies showed an epithelioid angiosarcoma. This has not been previously reported. Oleothorax is a historical treatment for tuberculosis and patients can remain asymptomatic for years afterwards. Tumors can be associated with such foreign bodies and diagnosis requires a high degree of suspicion and perseverance in the investigative pathways.

Keywords: Angiosarcoma, Epithelioid, Oleothorax, Tuberculosis

How to cite this article

Jackson K, Narkhede P, Aujayeb A. Epithelioid angiosarcoma arising from an oleothorax: An unreported case report. Int J Case Rep Images 2020;11:101166Z01KJ2020.

Article ID: 101166Z01KJ2020

doi: 10.5348/101166Z01KJ2020CR

Karl Jackson¹, Parag Narkhede¹, Avinash Aujayeb¹

Affiliation: ¹Northumbria HealthCare NHS Foundation Trust, Care of Tracy Groom, Northumbria Way, Cramlington, Northumberland, UK.

Corresponding Author: Avinash Aujayeb, Northumbria HealthCare NHS Foundation Trust, Care of Tracy Groom, Northumbria Way, Cramlington, Northumberland, NE23 6NZ, UK; Email: avinash.aujayeb@nhct.nhs.uk

Received: 27 July 2020

Accepted: 22 August 2020

Published: 23 September 2020

INTRODUCTION

Oleothorax is a historical treatment method for upper lobe predominant tuberculosis. It was gradually discontinued after the development of effective anti-tuberculous drugs in 1950s. Epithelioid sarcomas are rare, slow-growing soft tissue cancers [1–4]. Chronic inflammation through persistent oxidative and nitrative tissue damage is a recognized risk factor for human cancers in various organs. Sarcomas can develop as a consequence of such inflammatory processes [5].

CASE REPORT

A 76-year-old male patient presented with left sided chest pain, dyspnea, and weight loss. His past medical history included treated hypertension and an oleothorax performed for pulmonary tuberculosis (TB) in the 1950s. This had been followed by streptomycin therapy. He was a never smoker and had no occupational dust exposure.

A chest radiograph (CXR) showed a large smooth-edged density in the left upper and mid zones, corresponding rib resections, and calcification in the right upper and mid zones (Figure 1). This was consistent with an oleothorax and previous TB.

A computer tomogram (CT) showed a large well-defined mass in the left hemithorax. It was of mixed attenuation with peripheral and internal calcification. There was soft tissue thickening in the chest wall (Figure 2).

The differential diagnoses included expansion of the oleothorax, malignancy associated with a foreign body and reactivation of pulmonary TB. A positron emission tomogram (PET) showed intense uptake in the anterior margin of the oleothorax (Figure 3). Initial CT guided and surgical biopsies showed a histiocytic inflammatory infiltrate in keeping with a chronic foreign body reaction. Extended cultures for acid fast bacilli were negative.

Ongoing clinical concern prompted repeat imaging. This showed an enlarging lesion in the anterior chest wall with irregular destruction of the left second rib and no change in the pleural mass (Figure 4).

Repeat biopsies showed tumor composed of epithelioid cells with abundant amphophilic cytoplasm,

pleomorphic vesicular nuclei, and prominent nucleoli. There was extensive hemorrhage, tumor necrosis, and mitoses. Immunohistochemistry showed diffuse strong expression of CD31 and factor 8 with moderate-strong staining with pancytokeratin (AE1/AE3) and cytokeratin 7. The tumor did not express CD34, calretinin, CK 5/6, D2-40, WT1(m), CD15, TTF-1, BerEP4, or CEA. The findings were consistent with a high grade epithelioid angiosarcoma. Unfortunately, the histological and immunohistochemistry slides from our patient have been archived and we are unable to retrieve them but Sakamoto et al describe the specific and similar pathological findings associated with our case [6]. Figure 5 is taken from Sakamoto et al. that shows tumor being composed of large, epithelioid cells with vesicular nuclei and prominent nucleoli, and immunohistochemically tumor cells express cytokeratins AE1/AE3 and CD31. In post-diagnosis, the condition of the patient deteriorated rapidly and he died.

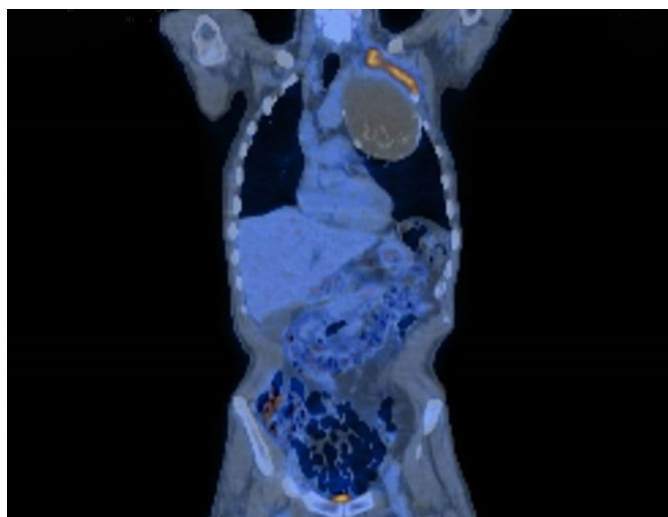


Figure 3: Positron emission tomogram (PET) showed intense uptake in the anterior margin of the oleothorax.



Figure 1: CXR showing a large smooth edged density in the left upper and mid zones, corresponding rib resections, and calcification in the right upper and mid zones.

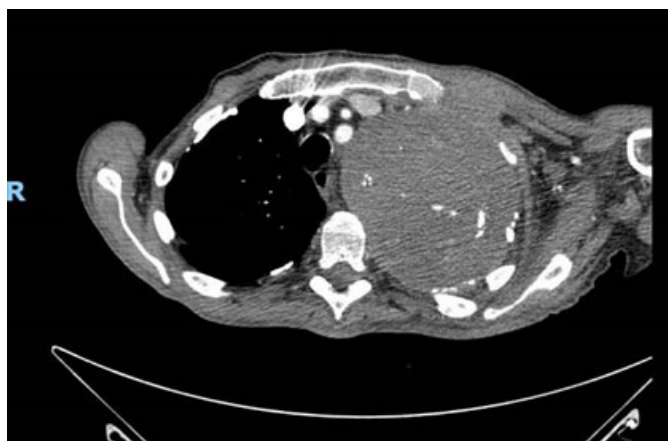


Figure 4: CT scan showing extension of the lesion into the anterior chest wall with irregular destruction of the left second rib and no change in the pleural mass.

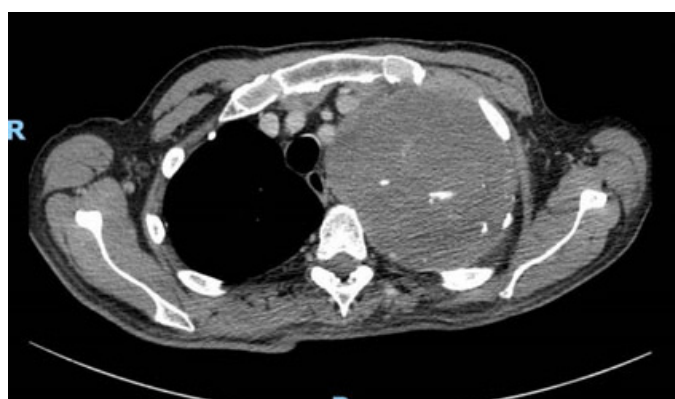


Figure 2: CT scan showing a large well defined mass in the left hemithorax of mixed attenuation with peripheral and internal calcification and with soft tissue thickening in the chest wall.

DISCUSSION

Mycobacterium tuberculosis was discovered and described by Robert Koch in 1882 [1]. Patients were initially treated in sanatoriums where rest was imposed and open-air shelters provide sufficient ventilation. Best rest was suggested as the pulmonary blood flow is gravity-dependent and thus the upright position creates a high oxygen tension in the apex of the lung which was felt to favor the growth of the mycobacteria. In those establishments, several therapeutic interventions were introduced and widely used as treatment for pulmonary tuberculosis. These included the induction of artificial pneumothoraces, artificial pneumoperitoneums, thoracoplasties, plombage, phrenic nerve crushes, and lung resections. All those methods were believed to reduce the amount of oxygen available for the pathogen to replicate. The discovery of anti-tuberculous drugs in the 1950s gradually led to a decline in the use of

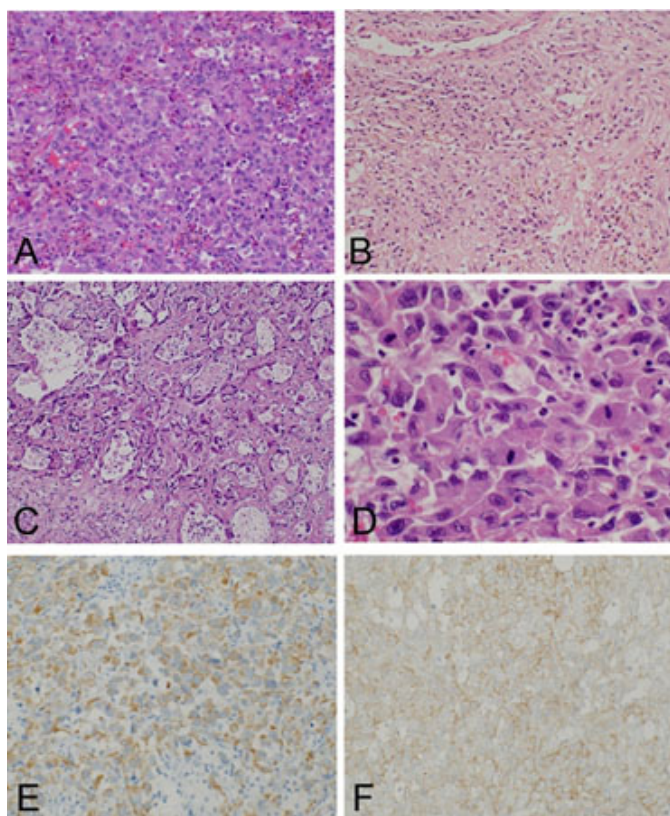


Figure 5: Slides showing classic epithelioid angiosarcoma histology: the tumor is composed of large, epithelioid cells with vesicular nuclei and prominent nucleoli (A), with fibrous stroma (B). (C) Vascular formation and malignant endothelial cells containing erythrocytes are observed. (D) Increased mitotic activity is observed. Immunohistochemically, tumor cells express cytokeratins AE1/AE3 (E), and the vascular marker CD31 (F) (taken from Sakamoto et al., Open Access article distributed under the terms of the Creative Commons Attribution License).

sanatoriums and an almost complete stop to the above surgical interventions [2].

Oleo thorax is oil in the pleural cavity and its insertion was one of the surgical interventions known as plombage. During plombage, a cavity was created under the ribs in the upper part of the thoracic cavity and inert material is inserted, either intra- or extra-pleurally. Those materials included air, olive, or mineral oil, gauze, paraffin wax, rubber sheeting, or bags and lucite balls. The amount of oil inserted varied between 100 mm and 2 L. The upper lobe of the lung would thus be forced to collapse and provide a relatively anaerobic environment for the mycobacterium pathogens.

In oleothoraces, the oil was aspirated at the completion of antibiotic treatment although often remained in situ and was found incidentally on thoracic imaging years later. Gomenol, an oil obtained from a plant, *Melaleuca viridiflora*, was sometimes added to paraffin oil for its germicidal actions. Oleothoraces tend to remain asymptomatic but complications include expansion and compressive symptoms, bronchopleural and pleuro-

cutaneous fistulae, reactivation of TB and empyema [3, 4].

Epithelioid angiosarcomas are rare, aggressive malignancies arising from epithelial cells and occur mostly in deep soft tissues. They account for less than 1% of all sarcomas. They have been associated with retained foreign material as well as schwannomas, trauma, surgery, and other malignancies [5–8]. There are no previous descriptions of angiosarcoma developing at the site of an oleothorax although an extra-skeletal chondrosarcoma at the site of lucite ball plombage has previously been reported [9].

Continued oxidative and nitrative tissue inflammation at the sites of foreign bodies is a recognized risk factor for cancer, such as sarcomas [10]. Histological analysis tends to demonstrate sheet-like, tubular, or nested growth patterns with focal vascular differentiation, and immunohistochemistry shows positivity for CD31, and partial positivity for Fli-1, CD34, and factor VIII-related antigen. The main differential diagnosis of epithelioid angiosarcomas are metastatic carcinomas and epithelioid hemangioendotheliomas. Identification of the presence of well-formed vascular channels and cytoplasmic vacuoles with red blood cell fragments helps with the differentiation. Furthermore, epithelioid hemangioendotheliomas are low-grade malignant vascular tumors with minimal cellular pleomorphism. Epithelioid hemangioendotheliomas are less aggressive than epithelioid angiosarcomas. Treatment usually involves chemotherapy and surgical excision of localized disease can improve outcomes but prognosis remains very guarded, with extensive metastasis possible, and thus conferring a survival range of 7–17 months [6–9].

CONCLUSION

Epithelioid angiosarcoma is a rare type of sarcoma that can be associated with foreign bodies and has a very poor prognosis. This case report is the first case described in the literature of an epithelioid angiosarcoma associated with an oleothorax.

REFERENCES

1. Brock TD. Robert Koch: A Life in Medicine and Bacteriology. Washington, D.C.: American Society of Microbiology; 1999.
2. Hochhegger B, Zanetti G, Marchiori E. An uncommon chest mass: Oleothorax. [Article in Portuguese]. J Bras Pneumol 2016;42(5):391.
3. Gillardin P, Mortelé K, Delens F, De Backeret AI. Oleothorax. JBR-BTR 2013;96(5):314.
4. Forouhi NG, Walker W, Leitch AG. Tuberculous empyema presenting 62 years after oleothorax: A case report. Respir Med 1994;88(5):391–2.
5. Jennings TA, Peterson L, Axiotis CA, Friedlaender GE, Cooke RA, Rosai J. Angiosarcoma associated with foreign body material: A report of three cases. Cancer

- 1988;62(11):2436–44.
6. Sakamoto A, Takahashi Y, Oda Y, Iwamoto Y. Aggressive clinical course of epithelioid angiosarcoma in the femur: A case report. *World J Surg Oncol* 2014;12:281.
 7. Rückert RI, Fleige B, Rogalla P, Woodruff JM. Schwannoma with angiosarcoma. Report of a case and comparison with other types of nerve tumors with angiosarcoma. *Cancer* 2000;89(7):1577–85.
 8. Meis-Kindblom JM, Kindblom LG. Angiosarcoma of soft tissue: A study of 80 cases. *Am J Surg Pathol* 1998;22(6):683–97.
 9. Thompson JR, Entin SD. Primary extraskeletal chondrosarcoma. Report of a case arising in conjunction with extrapleural lucite ball plombage. *Cancer* 1969;23(4):936–9.
 10. Tazawa H, Tatemichi M, Sawa T, et al. Oxidative and nitrative stress caused by subcutaneous implantation of a foreign body accelerates sarcoma development in Trp53+/- mice. *Carcinogenesis* 2007;28(1):191–8.

Author Contributions

Karl Jackson – Conception of the work, Design of the work, Acquisition of data, Analysis of data, Interpretation of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Parag Narkhede – Conception of the work, Design of the work, Acquisition of data, Analysis of data, Interpretation of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Avinash Aujayeb – Conception of the work, Design of the work, Acquisition of data, Analysis of data, Interpretation of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Guarantor of Submission

The corresponding author is the guarantor of submission.

Source of Support

None.

Consent Statement

Consent was obtained from the patient’s next of kin as the patient is now deceased.

Conflict of Interest

Authors declare no conflict of interest.

Data Availability

All relevant data are within the paper and its Supporting Information files.

Copyright

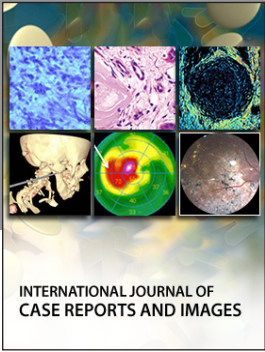
© 2020 Karl Jackson et al. This article is distributed under the terms of Creative Commons Attribution License which permits unrestricted use, distribution and reproduction in any medium provided the original author(s) and original publisher are properly credited. Please see the copyright policy on the journal website for more information.

Access full text article on other devices



Access PDF of article on other devices





Submit your manuscripts at
www.edoriumjournals.com

