ISSN: 2584-0355
DOI: doi.org/10.51219/MCCRJ/Ifeanyichukwu-Dupe-Nwanji/13

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https://urfpublishers.com/journal/case-reports

Vol: 1 & Iss: 2

Case Report

Rare Presentation of Pleural Mesothelioma with Associated Dysphagia in an Elderly Female Cement Trader and the Resultant Diagnostic Challenges: A Case Report

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Citation: Nwanji ID, Adegoke OO, Awosusi BL. Rare Presentation of Pleural Mesothelioma with Associated Dysphagia in an Elderly Female Cement Trader and the Resultant Diagnostic Challenges: A Case Report. *Medi Clin Case Rep J* 2023;1(2):43-46. DOI: doi.org/10.51219/MCCRJ/Ifeanyichukwu-Dupe-Nwanji/13

Received: 31 August, 2023; **Accepted:** 04 September, 2023; **Published:** 07 September, 2023

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ABSTRACT

Mesotheliomas are rare mesenchymal tumours that arise from the neoplastic proliferation of mesothelial cells lining various body cavities. Dysphagia is a rare complication of advanced mesothelioma usually occurring as a result of tumour compression or direct invasion of the oesophagus. Here we describe the case of an elderly female cement trader who presented on account of complaints of difficulty in swallowing of three months duration initially to solid foods and later to liquids. Chest X-Ray done showed a homogenous opacity in the left lower lung zone with obliteration of the left costophrenic angle. Computed tomography scan revealed thickened left pleura with reduced left lung volume. Upper gastrointestinal endoscopy revealed a vague lower oesophageal stricture suggestive of metastatic cancer from a lung primary. Patient had a gastrostomy on account of severe dysphagia. Her clinical condition deteriorated during the third week of hospital admission. Despite interventions and intensive care admission, she eventually succumbed to her illness and was certified dead after 25 days of hospital admission. At autopsy, there was a predominantly left-sided pleural-based mass with irregular thickening of the pleura maximal at the costo-diaphragmatic region with nodular involvement of the pericardium. Even though the oesophageal lumen was free of tumour along its length, the closely related aorta was fixed to the vertebral bones by tumour causing a mass effect. A conclusive diagnosis of epithelioid mesothelioma of the left pleura was made based on the gross and microscopic features of this tumour. This case suggests that in addition to recognized risks among construction workers exposed to asbestos, cement sellers/traders may also constitute an epidemiologic risk group for developing pleural mesotheliomas. Large-scale population based prospective studies may be needed to further explore this risk.

Keywords: Pleural mesothelioma; Dysphagia; Cement trader

Introduction

Mesotheliomas are rare mesenchymal tumours that arise from the neoplastic proliferation of mesothelial cells lining various body cavities including the pleura, pericardium, peritoneum, and the tunica vaginalis[1]. The majority of mesotheliomas arise in the pleural cavity with a significant male predominance[2]. The major risk factor for the development of mesothelioma is prolonged occupational or environmental exposure to asbestos[3]. The latency period from exposure to asbestos and the development of mesothelioma ranges from 20 years to more than 40 years[4]. Dysphagia is a rare, but recognised complication of advanced mesothelioma usually occurring as a result of tumour compression or direct invasion of the oesophagus[5].

This case report highlights the unusual and rare finding of dysphagia complicating pleural mesothelioma in an elderly female cement trader. It also brings to fore the diagnostic dilemma and fatality that can arise due to a low index of suspicion.

Case Presentation

History and examination findings

The decedent was a 68-year-old Nigerian female who presented at the University College Hospital, Ibadan, Nigeria on account of complaints of difficulty in swallowing of three months duration. The difficulty in swallowing was initially to solid foods and then gradually progressed to difficulty in taking liquids. There was associated retrosternal chest pain and weight loss. There was no history of ingestion of any corrosive substance. She is a known hypertensive. She did not take alcohol or use tobacco in any form. She was a cement trader (who had been in the cement trade for 20 years). Examination findings were those of a chronically ill-looking elderly woman with significant chest findings of dull percussion notes, and reduced breath sounds on the left lower lung zone. The remaining physical examination was essentially normal.

Laboratory and radiologic investigations

Laboratory work-up showed normal full blood count, urea, creatinine, and liver function tests. However, there was hypokalaemia with serum K+ value of 2.0mmol/l (normal reference range is 3.5-5.0mmol/l) and an elevated erythrocyte sedimentation rate of 102 mm/hr (normal reference range is 0-15mm/hr.).

Chest X-Ray done showed a homogenous opacity in the left lower lung zone with obliteration of the left costophrenic angle. The thoracic computed tomography (CT) scan revealed thickened left pleura with reduced left lung volume. The oesophagus was dilated. Upper gastrointestinal endoscopy revealed a vague lower third oesophageal stricture suggestive of metastatic oesophageal malignancy from a lung primary and a provisional diagnosis of an oesophageal malignancy was entertained. However, the oesophageal biopsy done was negative for malignancy.

Clinical diagnosis, treatment and patient outcome

The patient then had a gastrostomy on account of severe dysphagia. Her clinical condition deteriorated around the third week of hospital admission. She developed marked dyspnoea and subsequently had a cardiovascular arrest. Despite interventions and intensive care admission, she eventually succumbed to her illness and was certified dead after 25 days of hospital admission.

Autopsy Findings

The significant findings at autopsy were in the thoracic cavity. The left lung and visceral pleura were morbidly adherent to the parietal pleural at the superior and inferior aspects of the left hemithorax (**Figure 1A**). There were areas of pleural thickening and firm greyish-white tumour noted at the aforementioned sites. The maximum thickness of the pleura is 1.1cm (at the costodiaphgragmatic area, **Figure 1B**). At the left lung apex, the tumour encompassed the subclavian vessels. There were multiple tumour nodules seen at the pericardial pleura ranging from 0.5cm to 1.5cm in diameter (**Figure 1C**). The tumour extended to the midline and involved segments of the thoracic aorta, firmly attaching it to the vertebral bones. The oesophagus; however, was free of tumour along its entire length (**Figure 1D**).

There was atelectasis of the left lung. The right lung was moderately heavy and weighed 650g (reference range: 280-500g). The cut sections of the right lung showed moderate oedema and congestion.

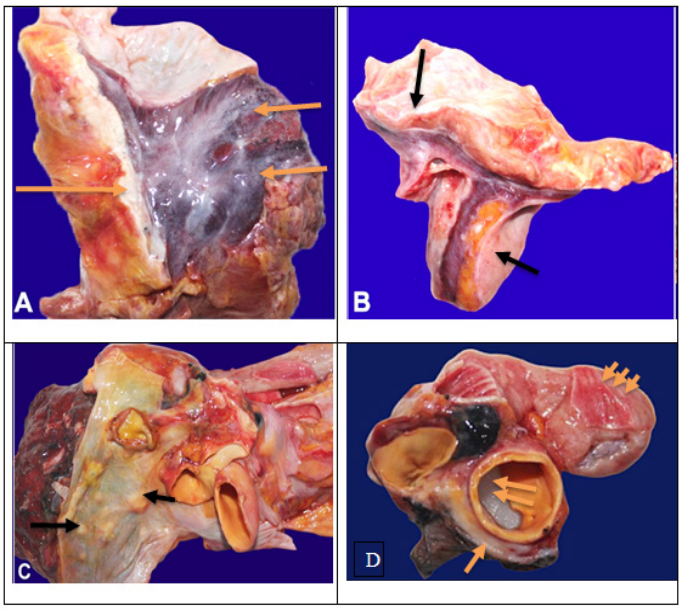

Figure 1. A - Photomicrograph showing the inferior surface of the left lung base (double green arrows). There is marked pleural thickening by greyish white tumour (single green arrow).

B - Photomicrograph showing cut sections of the lung at the costophrenic angle with markedly thickened pleural tissue (black arrows).

C - Pericardial surface with multiple greyish-white tumour nodules (black arrows).

D- Transverse section at the apical mediastinum showing greyish-white tumour (single green arrow) infiltrating the posterior aspect of the aorta (double green arrows). The oesophagus is located anteriorly (three green arrows) and it is free from tumour involvement.

Definitive histopathologic diagnosis

The histologic examination of the pleural mass showed a neoplasm composed of infiltrative cells disposed in pseudo-acinar patterns. The tumour cells were relatively monomorphic with bland nuclei and moderate amphophilic cytoplasm (**Figure 2A**). No cytoplasmic mucin vacuoles were seen. These tumour cells were seen to be infiltrating the wall of the aorta (**Figure 2B**). The underlying lung tissue showed atelectatic changes. A conclusion of epithelioid mesothelioma of the left pleural was made based on the gross and histologic features of this tumour.

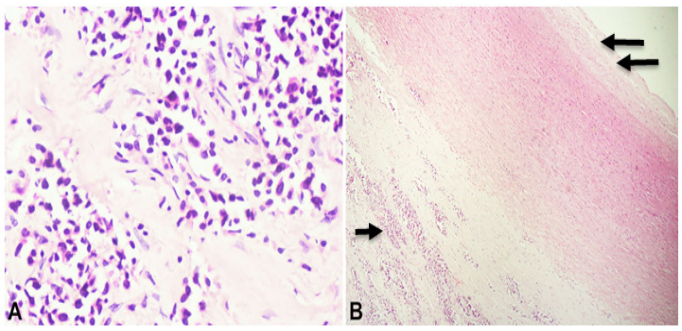

Figure 2. A: Photomicrograph of the pleural tissue showing infiltrative, mildly pleomorphic cuboidal cells disposed in pseudoglandular patterns (Hematoxylin and eosin stain, x 400 magnification).

B - Photomicrograph shows sections of the wall of the aorta with adventitial infiltration by tumour cells (single black arrow). Double black arrows highlight the tunica intima. (Hematoxylin and eosin, x40).

Discussion

The decedent presented primarily with symptoms of dysphagia which is a common presentation of oesophageal disorders. Even though the chest imaging showed a lung mass, the presenting symptom of dysphagia and findings of mild oesophageal stenosis in the lower third initially was considered as indicative of oesophageal involvement by a primary or metastatic lesion. The biopsies of the constricted portion of the oesophagus were negative for malignancy, resulting in a clinical diagnostic dilemma which could not be resolved until her demise.

At autopsy, pleural-based masses involving the apical and costophrenic angle and the pericardium were seen. The tumour had also encased and infiltrated the fibrous wall of the large vessels (including the aorta and left subclavian vessels). Regarding the aorta, the tumour caused an adherence of the aorta to the vertebral wall. Interestingly, no evidence of direct oesophageal involvement by the tumour was found. Nevertheless, the close anatomic relationship of the aorta and the compressive mass effect on the oesophagus must have been responsible for the difficulty in swallowing. Dysphagia is a rare but recognized complication of advanced mesothelioma and this may be due to mass effect of the tumour or direct infiltration into oesophageal lumen[5].

Histologic sections of the tumour showed features in keeping with mesothelioma. Mesotheliomas can be histologically categorized into three main subgroups: epithelioid type, sarcomatoid type and biphasic type (combining epithelioid and sarcomatoid features).6 The epithelioid type is the most common subtype and it shows epithelioid cells with ovoid-to-cuboidal nuclei with scanty to moderate cytoplasm[6].

The differential diagnoses in this case were oesophageal malignancy and primary lung adenocarcinoma. As earlier mentioned, the oesophageal mucosa was clean along its entire length with no evidence of neoplastic involvement. The differential diagnosis of primary lung cancer was also ruled out as the underlying lung tissue only showed features of atelectasis. Also, the nuclear features typical of adenocarcinomas such as eccentric or overlapping nuclei, vesicular chromatin, nuclear pleomorphism, and cytoplasmic mucin vacuoles were lacking in this case [4].

Ancillary investigations like immunohistochemistry can be helpful to further confirm this diagnosis. Mesotheliomas are usually positive for Calretinin, D2-40 (podoplanin) and WT1 while negative for epithelial markers such as Ber-EP4 and MOC-31[7]. These antibodies could not be tested for because they are not readily available locally in a resource-limited setting.

In this index case, the significant risk factor identified appears to be prolonged exposure to cement dust. In many developing countries such as Nigeria, regulation of the cement and asbestos content used in construction materials is limited. This has resulted in continued use of adulterated cement, asbestos and other construction materials with attendant health risks as seen in this case.

In spite of the World Health Organization and International Labour Organization's call for the adoption of a program to eliminate asbestos-related disease among nations through a ban on asbestos-containing materials, much is left to be seen in this area in the West African sub-region[8].

This case suggests that in addition to recognized risks among construction workers exposed to asbestos, cement sellers/traders may also constitute an epidemiologic risk group for developing pleural mesotheliomas. Large-scale population based prospective studies may be needed to further explore this risk.

Conclusion

This case describes pleural mesothelioma presenting primarily with dysphagia in a Nigerian female cement trader and highlights the diagnostic challenges and fatality involved when there is a low index of suspicion. It also brings to bare the need for sustained public health policies, oversight and proper regulation of the production, sale and use of cements, asbestos and other constriction materials.

Declaration

Ethical approval and consent to participate

Not applicable

Consent for publication

Verbal and written consent was obtained from the deceased's relatives to present the case for publication.

Availability of data and material

Not applicable.

Conflicts of interest

The authors have no conflict of interest to declare.

Funding

The authors did not receive any external funding in the course of writing and publishing this case report.

Author's contribution

Authors IDN and OOA performed the post-mortem examination. All authors reviewed the patient's clinical records and autopsy findings and made the definitive histopathologic diagnosis. Authors IDN and BLA conceptualized the idea of presenting it as a case report. All authors participated in the writing and editing of the final manuscript.

Acknowledgement

Not applicable

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