BJCR
Brazilian Journal of Case Reports

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Publicações Científicas de Acesso Aberto

Case Report

Aortoesophageal fistula with formation of secondary aortic pseudoaneurysm due to esophageal neoplasia: case report

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Abstract: Aortoesophageal fistula as a cause of upper gastrointestinal bleeding is a very rare event with a high immediate mortality rate. Some of the causes include foreign bodies, post-surgical complications, and malignancy. The following case describes how esophageal cancer invaded the walls of the descending aorta, leading to the formation of a pseudoaneurysm that subsequently fistulized into the esophagus, secondary to radiotherapy treatment. This case highlights the importance of evaluating the thoracic aorta before and during radiotherapy treatments, to investigate the existence of aortic vascular compromise adjacent to the esophagus. If the existence of an aortic pseudoaneurysm is detected, radiotherapy treatment should be suspended until the pseudoaneurysm is corrected, reducing the lethality of patients undergoing advanced treatments.

Keywords: Aortoesophageal fistula; Aortic pseudoaneurysm; Esophageal neoplasm; Upper gastrointestinal bleeding; Radiotherapy.

Citation: Frizon A, Schaffel EH, Donato F, Bello R, Macagnan MR, Silva FG, Sobrinho HR. Aortoesophageal fistula with formation of secondary aortic pseudoaneurysm due to esophageal neoplasia: case report. Brazilian Journal of Case Reports. 2024 Oct-Dec;04(4):18-22.

Received: 26 September 2023
Accepted: 13 February 2024
Published: 15 February 2024

1. Introduction

Aortoesophageal fistula (AEF) is a rare cause of massive upper gastrointestinal bleeding. It is a catastrophic condition that requires immediate treatment due to its high mortality rate. Thoracic aortic aneurysm, foreign body ingestion, esophageal malignancy and post-surgical complications are common causes of AEF [1, 2]. However, radiation-induced AEF is a rare phenomenon, and little described in the literature, but with high lethality even in specialized centers and with immediate therapy, making prevention the best option [3–5].

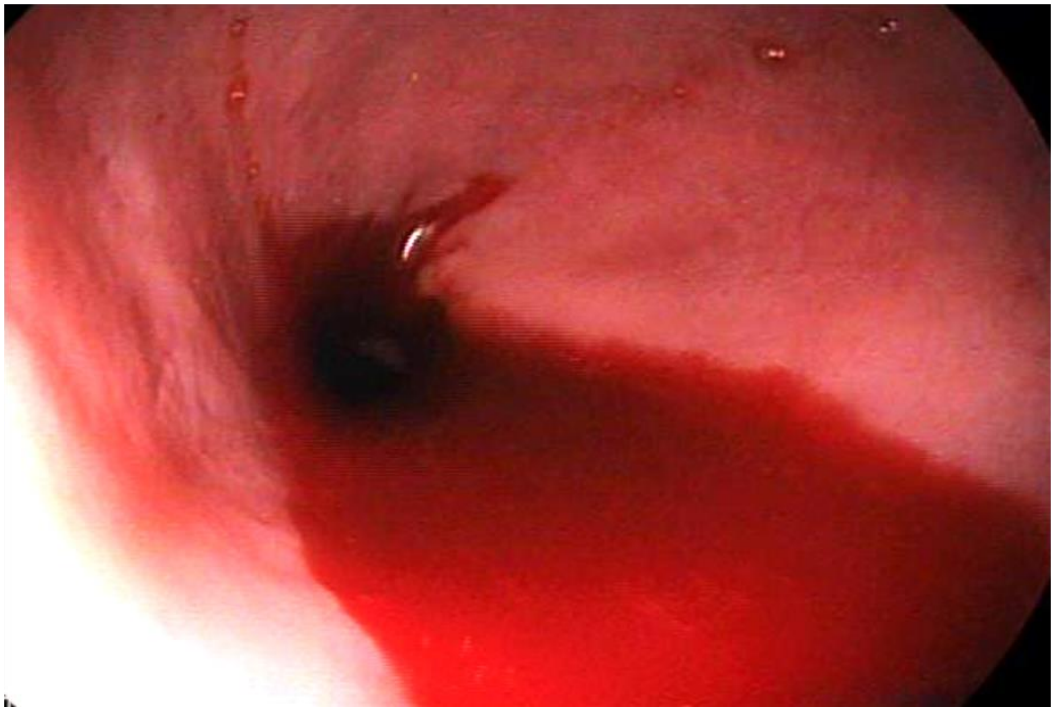
This case demonstrates that esophageal carcinoma can give rise to a pseudoaneurysm and progress to AEF in patients undergoing radiotherapy treatment. Tumor invasion determines local tissue fragility in the aortic wall and subsequent rupture and extension into adjacent soft tissues or channeling into the esophageal lumen. This work addresses the need to use diagnostic methods for vascular assessment such as CT angiography before and during radiotherapy treatments to detect pseudoaneurysms before rupture.

The objective of this work is to provide more details about this rare pathology and warn about the need to perform an aortic vascular study before starting radiotherapy treatment, especially in cases where esophageal cancer may be invading the thoracic aorta.

2. Case Report

A 52-year-old male patient long-time smoker and alcoholic with a history of dysphagia and weight loss, presented with an upper digestive endoscopy showing a vegetative lesion in the upper third of the esophagus, with pathology showing moderately differentiated squamous cell carcinoma. He underwent chemotherapy and radiotherapy and after 5 months of treatment (26 sessions of localized radiotherapy and 7 sessions of chemotherapy), hematemesis and melena began. After 7 days of these symptoms, he was taken to the hospital emergency department in hypovolemic shock with reduced consciousness due to massive upper gastrointestinal bleeding. He underwent urgent upper gastrointestinal endoscopy which revealed profuse bleeding around the tumor, with no evidence of esophageal varices (Figure 1). Local application of adrenaline was performed to control the bleeding without success. A thoracic tomography angiography was performed, which demonstrated AEF and vascularized nodulation compatible with a pseudoaneurysm (Figure 2 and 3).

Figure 1. Upper gastrointestinal endoscopy demonstrating profuse active bleeding.



3. Discussion and conclusions

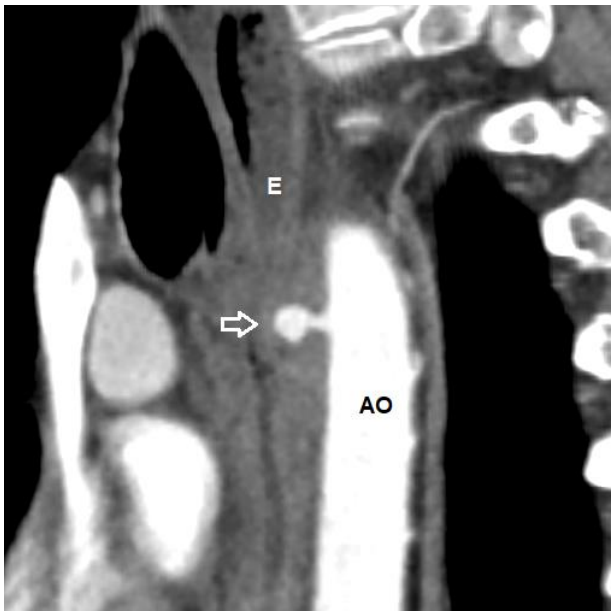
AEF is a rare condition, classified as primary or secondary (post-surgical) and in many cases clinical confirmation is made post-mortem due to its high lethality rate [1,6]. The classic symptomatic triad of AEF, described by Chiari, consists of chest pain, sentinel bleeding (days before), followed by massive hemorrhage [1]. However, not all patients present this triad as in our case, where the patient did not feel pain. Most of cases in the literature were diagnosed through upper digestive endoscopy and computed tomography, which have been described as good diagnostic options [1, 2, 7]. Esophageal malignancy is the third most common cause of AEF due to the infiltrative and ulcerative nature of the tumor [5,6,8]. However, there are a small number of cases in which radiotherapy may be the main causal factor for the development of AEF [1, 9, 10].

Radiotherapy is considered an independent risk factor for the formation of AEF and its effects depend on the dose, the patient's condition, and the irradiated field [3–5]. The effects of radiation vary in different tissues, with small vessels suffering from endothelial cell sloughing, fibrosis of the intimal layer with thrombosis and occlusion, while large

vessels are not directly affected by the effects of radiation. However, the vasa vasorum, a small network of capillaries that supplies nutrients to the vessel walls, can thrombose, leading to necrosis and perforation of the underlying vascular wall [2, 6, 9]. Currently, most esophageal tumors are treated with multimodal regimens and the incidence of AEF remains restricted to rare cases, with the effects of radiation remaining uncertain on fistula formation, especially in the presence of an infiltrative and ulcerative tumor in the vicinity of the aorta [4, 5].

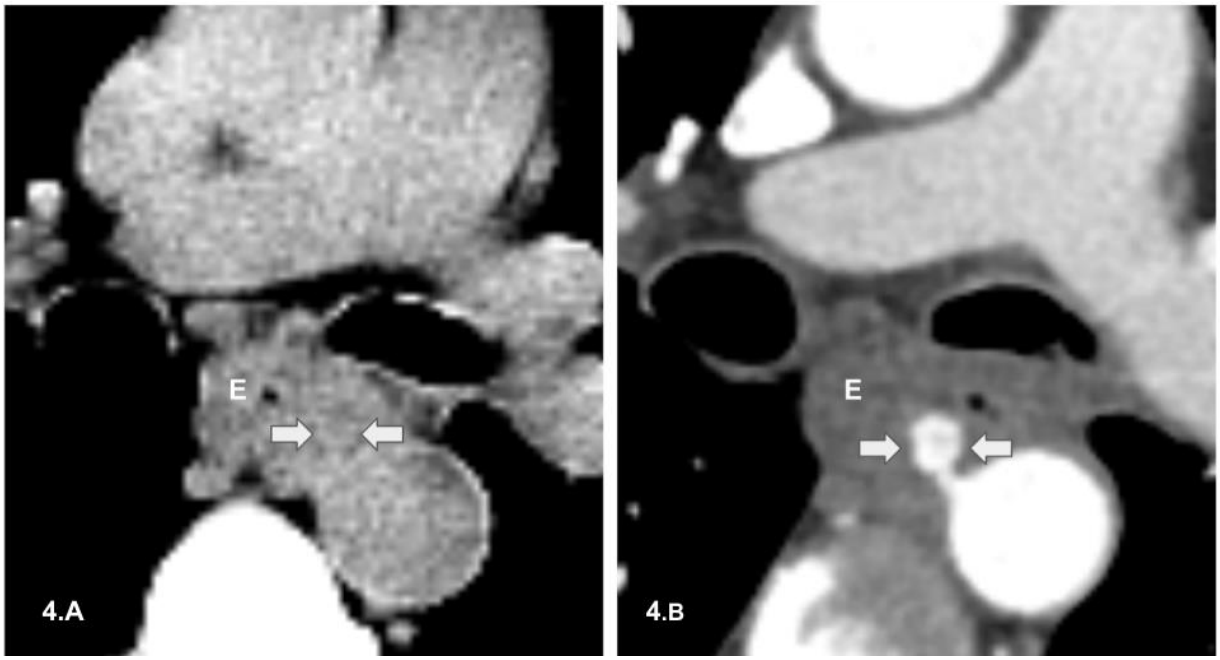
Figure 2. Chest angiotomography - axial section with a thickness of 1 mm before and after the use of intravenous contrast solution: pseudoaneurysm of the descending aorta projecting into the muscular wall of the esophagus, without acute hemorrhage (arrows), E - infracarinal esophagus, A - descending thoracic aorta.

Figure 3. Chest angiotomography - sagittal reformatted image. Pseudoaneurysm of the descending aorta projecting into the muscular wall of the esophagus (arrow), E - esophagus, AO - descending thoracic aorta.



In this case, the pseudoaneurysm arose from direct invasion of the thoracic aorta wall by the tumor, a diagnosis made after reviewing computed tomography images performed four months before the start of radiotherapy (Figure 4.A and 4.B). Unfortunately, this examination was performed without the use of intravenous contrast as radiotherapy planning, but revealed density changes in the area, suggestive of pseudoaneurysm formation. In this patient, radiotherapy may have been the determining factor for the rupture of the pseudoaneurysm and its fistulization with the esophageal lumen.

Figure 4. A. Computed tomography of the chest without contrast obtained 4 months before the start of radiotherapy. B. After radiotherapy: observe the presence of a rounded nodular image with a density like that of the thoracic aorta and which fistulized into the lumen of the esophagus, causing hypovolemic shock.



Treatment of AEF must be immediate and invasive management is the only option. There are no reported cases in which conservative management has been successful. Between the two reported options for invasive treatment, open surgical repair of reconstruction is associated with a higher risk of morbidity and mortality, while endovascular treatment using a stent to seal the fistula gap is considered a safer immediate alternative, particularly in hemodynamically unstable patients, with favorable short-term outcomes [2, 7, 9, 11, 12]. However, potential complications include a high rate of infection and recurrence as likely occurred in our case [6, 11].

The case report type of study has limitations regarding the extrapolation of population data and does not have the power to establish cause and effect. However, the sequence of facts described here and documented with images gives us the chance to clarify the relationship between a tumor and the wall of a large artery. As it is an immediate and almost always fatal outcome, it cannot be investigated with a prospective study or clinical trial. Some types of study could be used to increase the number of cases under analysis and make population inferences, including a retrospective cohort, case-control study or even a meta-analysis of published cases. Perhaps this way it will be possible to establish a cut-off point between the maximum period between the vascular study and the start of radiotherapy sessions as well as investigations between radiotherapy sessions.

Therefore, it is necessary to establish a protocol for carrying out a vascular study such as CT angiography immediately before the start of radiotherapy, especially when

there are neoplastic lesions in close contact with large vessels and possible vascular invasion. At this stage, the medical team must be able to detect the formation of a pseudoaneurysm and treat it before radiotherapy sessions, preventing a future rupture. This recommendation was not found in the literature and the standardization of diagnostic exams before and during radiotherapy sessions can potentially save lives.

Funding: None.

Research Ethics Committee Approval: We declare that the patient approved the study by signing an informed consent form and the study followed the ethical guidelines established by the Declaration of Helsinki.

Acknowledgments: None.

Conflicts of Interest: None.

Supplementary Materials: None.

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