Clinical report

Esophageal carcinoma in second-trimester pregnancy in a resource-limited setting: Case report

Abdirahman Ibrahim Said\textsuperscript{a,c}, Ismail Hussein Ismail\textsuperscript{a,c}, Ayanle Mohamed Mohamed\textsuperscript{a,c}, Halimo Barkhad Mohamed\textsuperscript{a,c}, Abdissamad Mohamoud Ali\textsuperscript{a,c}, Don Eliseo III Lucero-Prisno\textsuperscript{d} and Hassan Abdirahman Elmi\textsuperscript{b,e,⁎}

\textsuperscript{a} College of Health Science, School of Medicine, Amoud University, Borama, Somalia
\textsuperscript{b} Department of Biology, Amoud University, Borama, Somalia
\textsuperscript{c} Internal Medicine Department, Borama Regional Hospital, Borama, Somalia
\textsuperscript{d} Faculty of Public Health and Policy, London School of Hygiene and Tropical Medicine, London, United Kingdom
\textsuperscript{e} Department of Zoology, Faculty of Science, Charles University, Prague, Czech Republic

\textbf{Abstract}

Esophageal carcinoma during pregnancy is an extremely rare condition that presents unique challenges in terms of diagnosis and management. In this case study, we report on a 34-year-old pregnant woman from Somalia/Somaliland who presented with dysphagia and weight loss. She had previously been treated for gastroesophageal reflux disease, but her symptoms worsened over time. Upon examination, the patient exhibited cachexia, palpable cervical nodes, and basal crepitation upon admission. A CT scan of the neck revealed esophageal carcinoma with metastasis, while an ultrasonography confirmed the presence of a 24-week fetus. Due to the limited availability of chemotherapy and cancer treatment facilities, palliative care was initiated, unfortunately resulting in the unfortunate demise of both the patient and the fetus. This case highlights the significant impact of resource scarcity on disease management, as well as the ethical complexities involved in making decisions regarding palliative care for pregnant patients with cancer. It emphasizes the importance of timely diagnosis and the implementation of comprehensive, multidisciplinary approaches in addressing such rare occurrences.

© 2024 The Author(s). Published by Elsevier España, S.L.U. This is an open access article under the CC BY-NC license (http://creativecommons.org/licenses/by-nc/4.0/).

\textbf{Keywords:}
Esophageal cancer
Second-trimester pregnancy
Somalia/Somaliland
Palliative care

\textbf{Carcinoma de esófago en el embarazo del segundo trimestre en un entorno de recursos limitados: informe de caso}

El carcinoma de esófago durante el embarazo es una afección extremadamente rara que presenta desafíos únicos en términos de diagnóstico y tratamiento. En este estudio de caso, informamos sobre una mujer embarazada de 34 años de Somalia/Somalilandia que presentó disfagia y pérdida de peso. Anteriormente había sido tratada por enfermedad por refluego gastroesofágico, pero sus síntomas empeoraron con el tiempo. Al ser examinado, el paciente presentaba caquexia, ganglios cervicales palpables y crepitación basal al ingreso. La tomografía computarizada de cuello reveló carcinoma de esófago con metástasis, mientras que la ecografía confirmó la presencia de un feto de 24 semanas. Debido a la limitada disponibilidad de instalaciones de quimioterapia y tratamiento del cáncer, se iniciaron los cuidados paliativos, lo que desafortunadamente resultó en la desafortunada muerte tanto de la paciente como del feto. Este caso pone de relieve el impacto significativo de la escasez de recursos en el manejo de la enfermedad, así como las complejidades éticas involucradas en la toma de decisiones con respecto a los cuidados paliativos para pacientes embarazadas con cáncer. Enfatiza la importancia del diagnóstico oportuno y la implementación de enfoques integrales y multidisciplinarios para abordar estos casos raros.

© 2024 The Author(s). Publicado por Elsevier España, S.L.U. Este es un artículo Open Access bajo la licencia CC BY-NC (http://creativecommons.org/licenses/by-nc/4.0/).

\textsuperscript{⁎} Corresponding author.
\textit{E-mail address: rabiic23@amoud.edu.so} (H.A. Elmi).
Background

The diagnosis of cancer during pregnancy presents a complex medical and ethical challenge for both patients and healthcare providers. Cancer Associated with Pregnancy (CAP) is a relatively rare occurrence, estimated to occur in approximately 1 in 1000 pregnancies in developed nations, and even less frequently in developing countries due to the younger age of pregnant women. Among pregnancy-associated cancers, gastrointestinal malignancies are notably uncommon, with colorectal cancer (CRC) being the most prevalent. Other gastrointestinal cancers, such as hepatoma, are also more commonly encountered than esophageal cancer, making the latter exceptionally rare in pregnant individuals. When cancer is diagnosed during the first and second trimesters of gestation, both the patient and physicians face a difficult decision regarding whether to continue or terminate the pregnancy. Given the limited resources available, particularly in regions like Somaliland, we are presenting this case to shed light on the complexities of such settings.

Case presentation

History

Demographic data (DD): A 34-year-old female gravida 2 para 1 from Garba Dadar, was admitted to Borama Referral Hospital (BRH) in Borama, the capital city of the Awdal region, Somalia/Somaliland.

Chief complaint (CC): The patient presented with a 2-year history of difficulty swallowing.

History of present illness (HPI): She had been in good health until approximately 2 years ago when she started experiencing dysphagia, which gradually worsened from solid to liquid foods. At the same time, she experienced a significant weight loss of 20 kg over the past 2 years, with a noticeable acceleration in the last 6 months. Evening fever was also reported. In addition, the patient complained of progressive dyspnea but did not have orthopnea or paroxysmal nocturnal dyspnea (PND). She presented with a productive cough accompanied by streaked blood sputum, pleuritic chest pain, and halitosis. Furthermore, the hoarseness of voice, which had notably worsened in recent months, was observed.

Past medical history (PMHx): The patient had a long-standing history of gastroesophageal reflux disease (GERD) and had been on chronic omeprazole therapy. She had previous hospitalizations at BRH.

Social history and family history: There is no family history of cancer. She is a non-smoker, residing in a rural area, and working as a housewife. She has 1 healthy child, aged 2 years. Recently, she traveled to Ethiopia.

Physical examination (PE): Upon thorough examination, it was observed that the patient presented with cachexia and experienced episodes of hypotension, while the remaining vital signs appeared to be within the normal range. Palpation of the patient’s anterior cervical nodes revealed palpable nodes that exhibited rigidity, swelling, lack of mobility, and immobility. Auscultation of the lungs identified bilateral basal crepitations. Furthermore, the abdominal, neurological, and musculoskeletal examinations yielded unremarkable findings. The overall progress is shown in Fig. 1.

Investigations: The computed tomography (CT) scan of the neck, conducted in the first trimester of the patient’s pregnancy, revealed the presence of proximal esophageal carcinoma with invasion of the laryngeal and proximal tracheal walls, as well as multiple cavitary cervical adenopathy. Upon admission, the complete blood count (CBC) showed a white blood cell count of 7.3*10^9/L, a hemoglobin level of 10.1 g/dL, a mean corpuscular volume (MCV) of 95 fl, and a creatinine level of 0.89 mg/dL. Subsequent ultrasonography performed during the patient’s hospitalization confirmed the presence of a single fetus at 24 weeks gestation, with a normal fetal biophysical profile. Although further investigation through endoscopy and biopsy was recommended, the patient declined these procedures due to financial constraints.

Treatment and management: Due to the patient’s refusal for further investigation and the presence of metastatic cancer, alongside the lack of chemotherapy and sufficient cancer infrastructure in the war-torn country, the implementation of palliative care was commenced. This encompassed nutritional support and the contemplation of palliative surgeries for dysphagia, which the patient declined. Consequently, the patient chose home care in a rural area instead of receiving intensive care at the hospital. Regrettably, both the patient and the fetus succumbed to the disease after 2 weeks.

Discussion

Esophageal cancer, which is well-known for its high global mortality rate, presents a substantial challenge in the field of oncology. Typically, this type of cancer is diagnosed in older individuals, and the occurrence of esophageal carcinoma in young people is rare, with incidence rates increasing with age. The association between cancer and pregnancy is even more uncommon, estimated to range from 0.07% to 0.1% of all malignant neoplasms. This rarity underscores the complexity of managing such cases, particularly given the unique challenges posed by the coexistence of cancer and pregnancy. The 2 main subtypes of esophageal cancer, squamous cell carcinoma and adenocarcinoma, collectively make up more than 95% of malignant esophageal tumors. The prevalence of each subtype varies based on epidemiological factors, with studies from South Africa indicating a higher prevalence of squamous cell carcinoma. Notably, significant risk factors for esophageal squamous cell carcinoma include smoking, alcohol use, and achalasia. Conversely, major risk factors associated with esophageal adenocarcinoma include gastroesophageal reflux disease, obesity, and smoking. Upper endoscopy with biopsy and histopathological confirmation serves as the gold standard for diagnosis, while CT imaging plays a crucial role in staging esophageal cancer. Notably, the utility of CT scans becomes increasingly significant as the cancer progresses, aiding in the detection of various pathological features such as invasion into adjacent mediastinal structures. Advanced staging of the disease correlates with a worse prognosis, highlighting the importance of accurate staging through imaging modalities like CT scans. Furthermore, CT scans are instrumental in identifying T4 stage tumors, characterized by the loss of fat planes between the tumor and adjacent structures, with high sensitivity and specificity. Additionally, CT imaging facilitates the detection of aortic and tracheobronchial invasion with nearly 100% sensitivity. Therefore, as esophageal cancer advances, CT scans become even more
useful in guiding treatment decisions and predicting patient outcomes.\textsuperscript{8} Esophagectomy continues to be the primary treatment for early-stage esophageal cancer. For locally advanced cancers, a multimodal approach involving neoadjuvant chemotherapy or combined chemoradiotherapy (CRT) followed by surgery is recommended.\textsuperscript{9} Various chemotherapy regimens, including 5-fluorouracil (5-FU), cisplatin, and mitomycin C (MMC), have demonstrated clinical efficacy and are commonly used in these treatment protocols.\textsuperscript{10} One significant challenge in diagnosing and managing esophageal cancer during pregnancy pertains to the tendency of its symptoms to mimic those related to pregnancy itself. As a result, the diagnosis of esophageal cancer is often delayed, leading to the disease being discovered at more advanced stages.\textsuperscript{1} This delay in diagnosis can have profound implications for both the mother and the fetus, affecting the available treatment options and the overall prognosis. In the specific case presented, the patient’s realization of her pregnancy was delayed due to the masking effect of pregnancy symptoms caused by the disease. The experience of being pregnant while dealing with cancer presents various challenges and emotional turbulence for expectant mothers.\textsuperscript{1} This emotional turmoil can significantly impact decision-making when it comes to choosing treatment options. As illustrated in the case at hand, some patients opt for palliative care for themselves and their fetuses. These decisions pose profound ethical dilemmas for healthcare providers, highlighting the necessity of sensitive and nuanced approaches to patient care within these complex situations. Moreover, this case emphasizes the importance of developing comprehensive cancer care infrastructures that can cater to the distinct needs of pregnant cancer patients, particularly in regions with fragile healthcare systems like Somalia/Somaliland. At the core of this infrastructure lies the establishment of multidisciplinary teams encompassing oncologists, obstetricians, psychologists, and social workers. These teams play pivotal roles in providing holistic care and support for these patients.\textsuperscript{3} Additionally, increasing awareness and providing education among healthcare professionals regarding the diagnosis and management of cancer during pregnancy is crucial in facilitating early detection and improving outcomes.

**Conclusion**

The infrequent manifestation of esophageal cancer in pregnant women poses considerable obstacles in terms of diagnosis, treatment, and decision-making processes. This case study emphasizes the emotional and ethical intricacies associated with the provision of health care for pregnant individuals with cancer, thereby emphasizing the requirement for comprehensive cancercare systems and multidisciplinary approaches to adequately support these patients.

**Funding**

Not applicable.

**Authors’ contributions**

Abdirahman Ibrahim Said, Ismail Hussein Ismail, Ayanle Mohamed Mohamed, Halimo Barkhad Mohamed, and Abdisamad Mohamoud Ali contributed to history taking and provided care for the patient throughout her hospital stay. Additionally, Hassan Abdirahman Elmi and Abdirahman Ibrahim Said contributed to the development of the manuscript, writing, and editing.

**Ethical approval**

The study adheres to the ethical standards required for research at Borama Regional Hospital. The ethical approval is issued under registration number BRH22/2024.

**Declaration of competing interest**

The authors affirm that there are no conflicts of interest about the publication of this article.

**Acknowledgments**

We would like to extend our sincere thanks to Mohamed Ali Ibrahim, Abdirashid Ismail H. Muhumed, Zakariye Mohamed Egeh, Suheyb Mubarak Osman, Arab Farah Mohamoud, Khalid Abdi Ismail, Nasra Abdisamad Osman, Abdillahi Mohamed Hassan for their providing valuable insights and assistance. Also, we would like to extend our gratitude to Drs. Safia Farah, Dr. Mohamed Abdirahman Osman, and Dr. Muna Abdillahi for their hard work.

**References**