

A case report of metastatic esophageal squamous cell carcinoma during pregnancy

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Abstract

Esophageal cancer is extremely rare in pregnant women, accounting for only 0.07%–0.1% of all malignant neoplasms in pregnancy. It is rapidly progressive in nature and requires timely intervention. Our literature search yielded six case reports of esophageal cancers in pregnancy thus far. We herein report the case of a 30-year-old female, presenting at 32 weeks of gestation with complaints of hoarseness, dysphagia, and weight loss. The biopsy taken from within the esophagus, using an endoscope during an upper endoscopy procedure confirmed the diagnosis of squamous cell carcinoma of esophagus and Positron emission tomography-computed tomography revealed metastasis to parathyroid region. She was confirmed with metastatic squamous cell cancer positive for cytokeratin 5/6 and cytokeratin P40 immunohistochemistry. The Positron emission tomography-computed tomography of the chest showed a large hypermetabolic soft tissue mass in the mid-esophagus with significant proximal dilation. The head-and-neck computed tomography scan represented a necrotic nodal mass in the neck and circumferential mural thickening involving the mid-esophagus resulting in the proximal dilation confirming the primary tumor site. Two weeks after the delivery, the patient started a chemotherapy regimen consisting of nine cycles of Carboplatin and Paclitaxel. Postchemotherapeutic computed tomography showed remonstration of lesion in the thyroid gland and middle part of the esophagus. Thus, in pregnant patients with new onset hoarseness, dysphagia, or substantial weight loss, clinicians should keep esophageal cancer as part of their differentials. A thorough history, detailed physical examination, and imaging should be performed to determine esophageal cancer, as it can advance swiftly and has a poor prognosis if left untreated.

Keywords

Cancer, chemotherapy, esophagus, pregnant, case report

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Introduction

Cancer in pregnancy affects around one in every 1000 pregnancies, and these can arise from a variety of sources, including immune system alterations, exposure to toxins, genetic abnormalities, and hormonal fluctuations. Melanoma, ovarian, cervical, and breast cancers are a few frequent cancers that can develop during pregnancy; it is no longer a rare event.¹ Diagnostic difficulties complicate clinical progression and may cause delayed diagnosis and advanced illness manifestation. A multidisciplinary approach is frequently necessary to properly balance the health of the mother and the fetus in treatment decisions. The effects on the mother include mental distress, bodily discomfort, and a strong need for social support.

Esophageal cancer, on the other hand, is an exceedingly rare form of neoplasia and accounts for between 0.07% and 0.1% of all malignant neoplasms in pregnant women.² Yamada et al.³ reported only five cases of esophageal cancer

in pregnancy, indicating that it is highly unusual, as esophageal cancer primarily affects men in the seventh to eighth decade of their lives. Differentiating between the symptoms of esophageal cancer and those of pregnancy is the most challenging aspect of diagnosis due to the concealment of cancer symptoms.⁴ Despite a growing interest among surgeons and pathologists in the early detection of precancerous lesions, esophageal cancer is still diagnosed at an advanced stage. As a result, the reported prognosis and course of

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treatment is poor.⁵ Changes must be made to the standard surgical and chemotherapeutic management of cancer in pregnant women to lessen maternal and fetal risks.² We present the case of a 30-year-old pregnant patient with metastatic squamous cell esophageal carcinoma associated with lesions in thyroid and parathyroid regions. This case report has been reported following the SCARE Criteria.⁶

Case presentation

A 30-year-old pregnant patient, 1 gravida 0 para presented to the outpatient department at 32 weeks and 0 days of gestation with complaints of hoarseness, dysphagia, and weight loss that had been ongoing for the past 10 weeks. In addition, she experienced severe vomiting, difficulty breathing, and swelling on the left side of her neck, with a nonradiating localized pain at the substernal region from the initial 12 weeks of her pregnancy. She had a cholecystectomy 5 years ago and had no comorbidities or a family history of cancer. Moreover, she had no history of smoking, alcohol, or illicit drug use. She was taking prenatal vitamins and did not report any history of gastrointestinal bleeding, peptic ulcer disease, or gastro esophageal reflux disease. The patient was vitally stable and the general physical examination, which included assessments of the neurological system, gastrointestinal tract, respiratory system, and cardiovascular system, was unremarkable. Only on regional examination of the head and neck, there was a lump seen on left side of the neck. Her heart rate was 88 beats per minute, and her blood pressure was 100/70 mmHg.

An obstetrical ultrasound conducted at 30 weeks of gestation reported a singleton alive intrauterine pregnancy in cephalic presentation with a fetal length of 60 mm corresponding to a gestation age of 30 weeks and revealed normo-hydroaminos. A Doppler study was advised to rule out Intra-Uterine Growth Restriction.

Neck X-ray showed a mass in the esophageal region and a fine needle aspiration biopsy was conducted. A cytology report confirmed metastatic squamous cell carcinoma showing hyper chromatic, pleomorphic, and enlarged nuclei, with irregular nuclear membrane, prominent nucleoli, and moderate amount of cytoplasm. In addition, the focal keratinized pearl formation, acute inflammatory cells, and lymphocytes are also visible. The immunohistochemistry was positive for cytokeratin 5/6 and cytokeratin P40. The Positron emission tomography-computed tomography (PET CT) of the chest showed a large hyper metabolic soft tissue mass in the mid-esophagus (T4–T7) with significant proximal dilation (Figure 1), which is likely a primary tumor site, and a F-18 fluorodeoxyglucose (FDG) avid lymph node in the left perilesional area (regional metastasis), but no hyper metabolic metastases in the liver, spleen, or adrenal glands. Additionally, PET/CT of the head and neck revealed a centrally necrotic soft tissue mass in the lower left cervical region, displacing the thyroid and larynx to the right and extending up to the



Figure 1. The Positron emission tomography-computed tomography showed a large hyper metabolic soft tissue mass in the mid-esophagus (T4–T7) with significant proximal dilation.



Figure 2. Enhanced chest computed tomography coronal scan reveal a necrotic nodal mass displaced left lobe of the thyroid gland to the contralateral side and lateral displacement of the trachea and cervical esophagus.

prevertebral fascia, with no hyper metabolic abnormalities in the nasopharynx or oropharynx.

The thyroid ultrasound revealed a well-defined heterogeneous mass measuring 4.9×3.9 cm in the left parathyroid region with tiny specks of calcification. The head and neck computed tomography (CT) scan represented a necrotic nodal mass measuring up to $6.4 \times 7.5 \times 6.5$ cm, displacing the left lobe of the thyroid gland to the contralateral side and causing lateral displacement of the trachea and cervical esophagus. It also reported a circumferential mural thickening seen involving the mid-esophagus resulting in the proximal dilation confirming the primary tumor site (Figure 2).

The treatment plan after extensive collaboration among oncology, radiology, and gynecology experts regarding the patient's perspective and expectations was to start chemotherapy after Caesarean section as the patient was presented in her last trimester. Following careful consideration, a gastrostomy feeding tube (G-tube) insertion was performed in

the 8th month of pregnancy due to dysphagia and regurgitation, followed by an emergency Cesarean section at 32 weeks and 6 days of gestation, resulting in the delivery of a normal, healthy male infant weight 2.7 kg by cephalic presentation. There were no congenital abnormalities reported in the child, and the clinical course was uneventful. Two weeks after the delivery, the patient was advised to follow a chemotherapy regimen consisting of nine cycles of Carboplatin and Paclitaxel. She completed nine cycles of Carboplatin 150 mg/m² and Paclitaxel 100 mg/m², but no radiotherapy was done.

The postchemotherapy CT demonstrated a large heterogeneous lobulated lesion projecting from the lower lobe of the left thyroid gland, the size of the lesion reduced to 3.4 × 3.0 × 5.7 cm. It also revealed a circumferential mural thickening involving the mid part of esophagus. She has currently completed chemotherapy and is expected to undergo radiotherapy. Radical surgery was considered as a course of treatment but was advised against it.

Discussion

Esophageal cancer identified during pregnancy is extremely rare, and the primary challenge with diagnosis occurs due to esophageal cancer symptoms being frequently confused with pregnancy symptoms, leading to the diagnosis at a later stage.⁷ In our case, the patient was diagnosed at 32 weeks and 3 days with complaints of hoarseness, dysphagia, and weight loss. Moreover, female reproductive hormones have been found to play a preventative function against esophageal squamous cell carcinoma (ESCC) due to the presence of estrogen receptors in esophageal tissue and the tumor growth inhibitory effect of estrogen.⁷ In Western countries, men outnumber women concerning ESCC, and this may be due to men's higher rates of cigarette and alcohol consumption, both of which are considered ESCC risk factors.⁸ The patient presented above had no history of smoking, alcohol, or other drug misuse.

Given the significance of PET/CT in the diagnosis and surveillance of metastatic lesions, its use during pregnancy to confirm a nonmetastasized tumor is required for the continuation of the pregnancy.³ One of the main hazards associated with PET/CT imaging is the possible toxicity from exposure of ionizing radiation. Hence, to minimize fetal exposure, the radiation should not exceed 50 mGy (5 rad), below this threshold, the radiation does not cause any deterministic harmful effects including fetal growth restriction, congenital abnormalities, and microcephaly or pregnancy loss.⁹ To minimize the radiation exposure to the fetus, the patient underwent FDG-PET (2.7 mGY). Her pregnancy resulted in the delivery of a normal, healthy male infant weighing 2.7 kg.

The treatment of esophageal cancer during pregnancy adheres to similar guidelines as those for gastric cancer.¹⁰ Ueo et al.¹¹ defined standards for the treatment of pregnant women with gastric cancer, when diagnosed after the 30th

week of pregnancy, it is suggested that the child be delivered while still alive. In addition, Doosti-Irani et al.¹² stated that patients with esophageal SCC responded favorably when treated with neoadjuvant chemoradiotherapy, carboplatin-paclitaxel, radiotherapy, and surgery. The patient underwent an emergency C-section, giving birth to a healthy male infant and completing the nine cycles of chemotherapy resulting in the reduction of the lesion projecting from the left thyroid lesion.

A diagnosis of ESCC was made by histological assessment of the lesion; ESCC is treated with radiation therapy, chemotherapy, or surgical resection. The rare occurrence of ESCC in pregnant women has made it difficult to identify certain diagnostic markers in our investigation, such as tumor stage. Our patient's asymptomatic state and decreased lesion size following chemotherapy indicate that the chemotherapy cycles were successful. Radical surgery was being evaluated as a course of treatment but was not advised for a number of reasons. Complete removal is challenging due to the patient's advanced disease stage, which includes extensive local invasion and distant metastases. Given the patient's recent births and ongoing chemotherapy, the substantial risks of surgery—which include the potential to damage adjacent organs, infection, bleeding, and complications related to anesthesia—may exceed the benefits. Furthermore, scheduled radiation therapy following chemotherapy may reduce tumor size and eradicate cancer cells, hence obviating the necessity for surgery. A less intrusive treatment strategy can be more suitable, depending on the patient's age, general health, and personal preferences.

Given the advanced stage of the patient's cancer, an expandable metallic esophageal stent was recommended to palliate dysphagia and improve swallowing. This less-invasive procedure would enhance the patient's quality of life. The patient would continue with radiotherapy after chemotherapy to shrink the tumor and alleviate symptoms. Palliative care services were involved to manage symptoms, provide pain relief, and offer emotional support. Regular follow-up appointments to monitor progress and adjust the care plan as needed were done. Significant psychological discomfort can arise after an advanced cancer diagnosis, particularly in light of the hormonal changes that coincide with pregnancy. In order to teach coping mechanisms, offer emotional support, and promote contact with the medical staff, psychological counseling being a crucial step was suggested. In order to help the patient handle stress and take care of her newborn, this support was considered essential as she adjusts to her new position as a mother.

Pregnancy-related aggressive cancer management brings special difficulties. Younger individuals can frequently withstand harsh therapies better, but immunological tolerance brought on by pregnancy may slow the spread of cancer. It may be beneficial to combine aggressive radiation with sensitizing chemotherapy, but there are hazards to consider, such as radiation exposure to the fetus and serious side

effects for the mother. Because of the overlapping symptoms and diagnostic hazards, evaluating high-grade intestinal malignancies during pregnancy is difficult and necessitates a high index of suspicion along with a comprehensive screening strategy that includes a full history, physical examination, and suitable imaging.

In summary, a multidisciplinary strategy that takes into account the patient's age, immunological state, fetal safety, and therapy risks and benefits is necessary to treat severe malignancies during pregnancy. To create more effective diagnostic and therapeutic approaches for high-risk intestinal malignancies during pregnancy, more study is required.

Patient's final outcome

The patient reacted favorably to the chemotherapy treatment consisting of Paclitaxel and Carboplatin after being diagnosed. Positive treatment benefits were shown by a reduction in the size of the lesion on her left thyroid gland during a CT scan performed after chemotherapy. She will have radiation therapy after completing chemotherapy to further shrink the tumor and ease her discomfort. The patient has benefited from psychological counseling and has been tenacious and involved in her care throughout treatment. With frequent check-ins to assess and modify her care plan, she continues to be cooperative. The patient has demonstrated the effectiveness of a multidisciplinary approach by living a joyful life as a new mother and seeing considerable tumor decrease and symptom improvement overall.

Conclusion

Esophageal cancer in pregnancy is a rare entity with its symptoms being similar to events related to normal pregnancy which makes it essential for clinicians to thoroughly investigate and examine any patient exhibiting symptoms including a new onset of hoarseness, dysphagia, and significant weight loss. In addition, the FDG-PET for metastatic lesion diagnosis during pregnancy should be below the toxic fetal radiation dose threshold. The approach to chemotherapy, radiotherapy, and cancer surgery must be evaluated when treating pregnant patients especially in the last trimester to minimize the fetal and maternal risks.

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Author contributions

B.S. Involved in the conceptualization of the topic, data curation, formal analysis, investigation, methodology, and writing of the manuscript; M.T.A. Involved in the investigation, methodology, project administration, and writing of the manuscript; S.S. Involved in formal analysis, validating, reviewing, and editing of the manuscript; M.D. Involved in the supervision and editing of the manuscript.

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