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A Rare Malignant Paradox In The Oesophagus: Case Report Of Carcinosarcoma With Pathological Insights

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ABSTRACT

Oesophageal carcinosarcoma is a rare biphasic malignant tumour composed of both epithelial and mesenchymal components. A 64-year-old Indian woman presented with progressive dysphagia, weight loss, and decreased appetite for six months. Imaging revealed a well-defined, homogeneously enhancing submucosal lesion along the posterior oesophageal wall, suggestive of a benign lesion such as leiomyoma. Upper gastrointestinal endoscopy identified a large submucosal mass with surface ulceration. biopsy from the ulcer showed atypical pleomorphic cells suggestive of carcinoma. The patient underwent video-assisted thoracoscopic hybrid McKeown esophagectomy with two-field lymphadenectomy. Gross examination revealed a 5 x 3 x 3 cm polypoid mass confined to the submucosa. Histopathological evaluation demonstrated a biphasic tumour with solid nests of squamous epithelial cells showing comedo-type necrosis and a pleomorphic spindle cell component. Immunohistochemistry confirmed the diagnosis of carcinosarcoma, with strong cytokeratin and p63 positivity in the epithelial component and strong vimentin positivity in the mesenchymal component. The tumour was staged as pT1b N0 Mx, with all margins free of tumour and uninvolved lymph nodes. This case highlights the need for thorough histological and immunohistochemical assessment of oesophageal submucosal lesions, even when radiological features suggest a benign process. Early surgical management combined with adjuvant chemotherapy may improve outcomes in such rare and aggressive tumours.

Keywords: *Oesophageal carcinosarcoma, Biphasic tumour, Immunohistochemistry*

1. INTRODUCTION

CASE REPORT

A 64-year-old Indian woman presented with dysphagia, weight loss, and decreased appetite for 6 months. A CT scan of the thorax revealed a well-defined, nearly homogeneously enhancing oval lesion with a broad base on the posterior wall of the oesophageal lumen. The lesion measured 4.7 x 2.4 x 1.4 cm and extended from the inferior margin of the D3 vertebra to the superior margin of the D6 vertebra. An air crescent was identified along the anterior margin of the lesion. There was no evidence of obstruction or proximal dilation of the oesophagus and infiltration into the adjacent adipose tissue, indicating a benign submucosal oesophageal lesion, such as a leiomyoma (Fig.1).

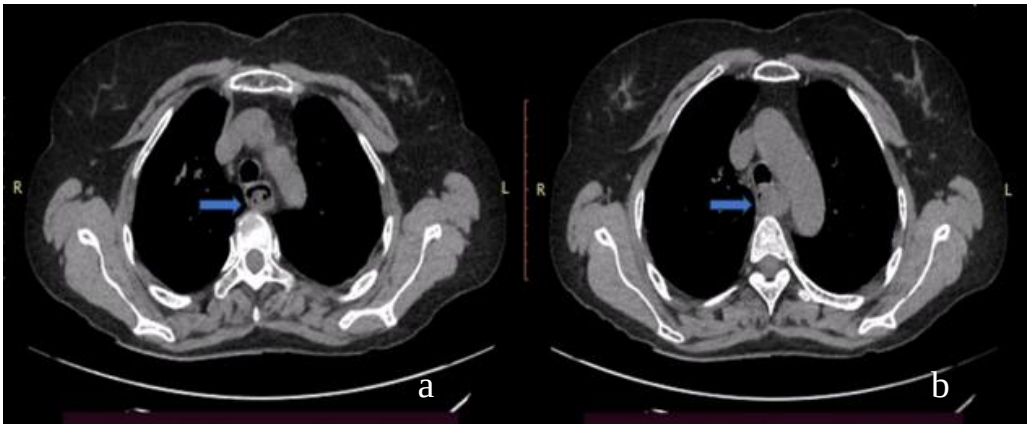
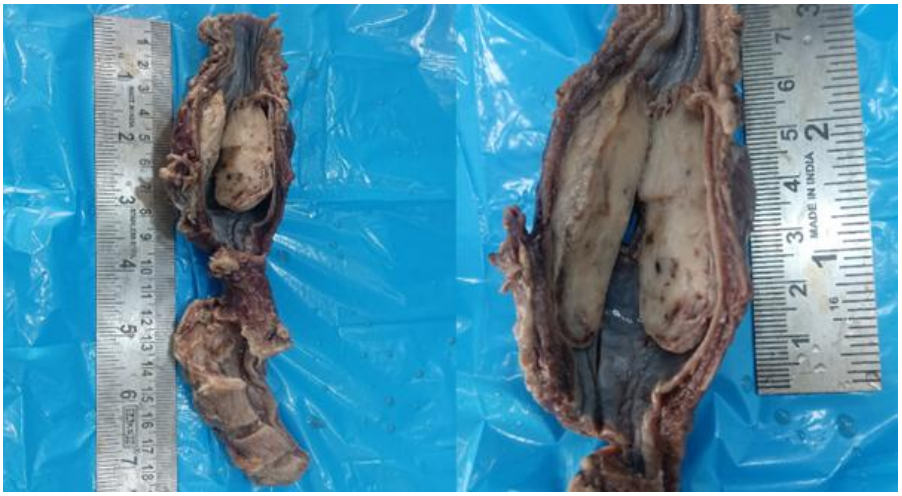


Fig.1: a, b.) CT thorax shows a well-defined, homogeneously enhancing oval lesion on the posterior oesophageal wall, measuring 4.7 × 2.4 × 1.4 cm (blue arrow).

Upper gastrointestinal (UGI) endoscopy with biopsy indicated a large submucosal lesion exhibiting surface ulceration, measuring 7 x 4 x 4 cm, extending from 23 to 27 cm from the incisors. Biopsy taken from the ulcer shows atypical cells with pleomorphic, hyperchromatic nuclei and moderate to abundant eosinophilic cytoplasm. Few binucleated and multinucleated cells are seen, which are suggestive of carcinoma. The patient subsequently underwent a video-assisted thoracoscopic hybrid McKeown esophagectomy accompanied by a two-field lymphadenectomy.

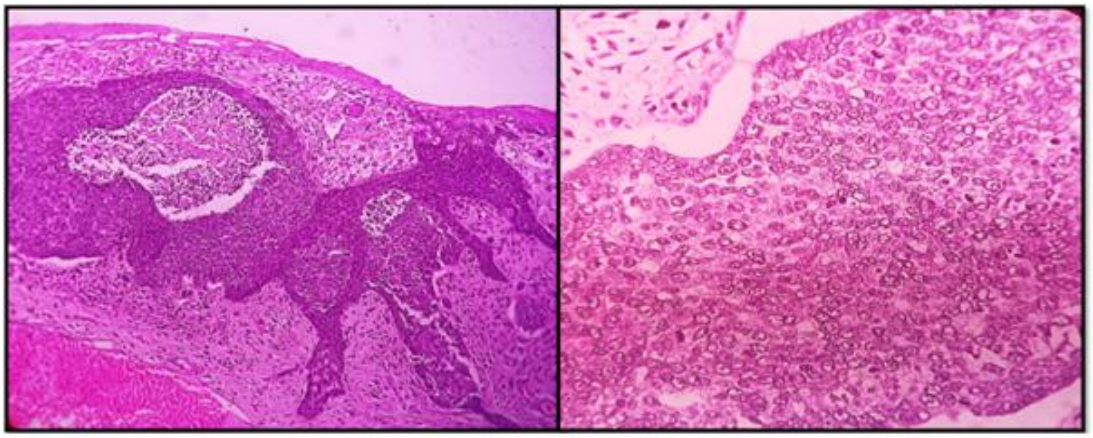
Gross examination of the surgical specimen revealed a polypoid mass measuring 5 x 3 x 3 cm located in the middle oesophagus with a homogeneous grey-white firm cut surface. The tumour appeared confined to the submucosa (Fig.2). On exploration, two lymph nodes were identified.



a **b**

Fig.2: Gross features: a.) Polypoid mass in the middle oesophagus; b.) The cut surface reveals a homogeneous grey-white appearance, confined to the submucosa.

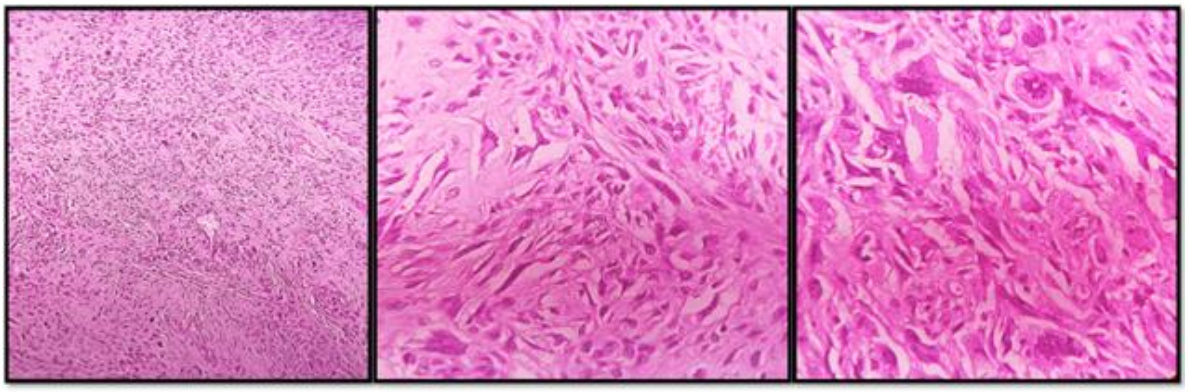
Histopathological examination using Haematoxylin and Eosin (H&E) demonstrated an infiltrative tumour extending into the submucosa with a biphasic morphology. The epithelial component was arranged in solid nests and anastomosing trabeculae, accompanied by comedo-type necrosis. The epithelial cells exhibited moderate pleomorphism with round to ovoid vesicular nuclei, scant cytoplasm, and numerous mitotic figures (Fig.3).



a **b**

Fig.3: a.) The epithelial component was arranged in solid nests and anastomosing trabeculae with comedo-type necrosis (H&E,100x); b.) The cells showed moderate pleomorphism, round to ovoid vesicular nuclei, and scant cytoplasm (H&E,400x).

The stromal component showed marked pleomorphism with spindle-shaped cells, elongated hyperchromatic to vesicular nuclei, and collagen deposition. Numerous bizarre nuclei and bi-/multinucleated tumour giant cells were also observed (Fig.4). This component also exhibited regions of necrosis, increased vascularity, and foci of extravasated red blood cells, resembling an angiosarcoma pattern. No perineural invasion or lymphovascular invasion was observed. The muscularis and serosa were free of tumour involvement. All surgical margins were free of tumour. The two identified lymph nodes showed reactive changes but no evidence of tumour.



a **b** **c**

Fig.4: a.) Cellular neoplasm composed predominantly of spindle cells arranged in fascicles(H&E,100x); b.) Spindle cells with marked nuclear pleomorphism, hyperchromasia, and occasional mitoses (H&E,400x); C.) Cellular atypia with bizarre, pleomorphic tumour giant cells (H&E,400x).

The differential diagnoses included carcinosarcoma, basaloid squamous cell carcinoma, and angiosarcoma. Immunohistochemical analysis demonstrated strong cytokeratin positivity in the epithelial component, confirming squamous differentiation (Fig.5), and strong vimentin positivity in the spindle cell component, indicating mesenchymal differentiation typical of carcinosarcoma (Fig.6). Moderate p63 positivity further supported squamous differentiation, while mild nuclear positivity for p53 in 20% of tumour cells suggested a possible p53 mutation, commonly found in sarcomatoid carcinomas (Fig.5). Negative staining for S100 and CD34 ruled out neural or melanocytic differentiation and angiosarcoma, respectively (Fig.6).

a **b** **c**

Fig.5: Immunohistochemistry studies a.) Strong cytoplasmic positivity for cytokeratin in the epithelial component, confirming squamous differentiation (CK,100x); b.) (p63, 100x); c.) Focal nuclear positivity for p53 (p53, 100x)

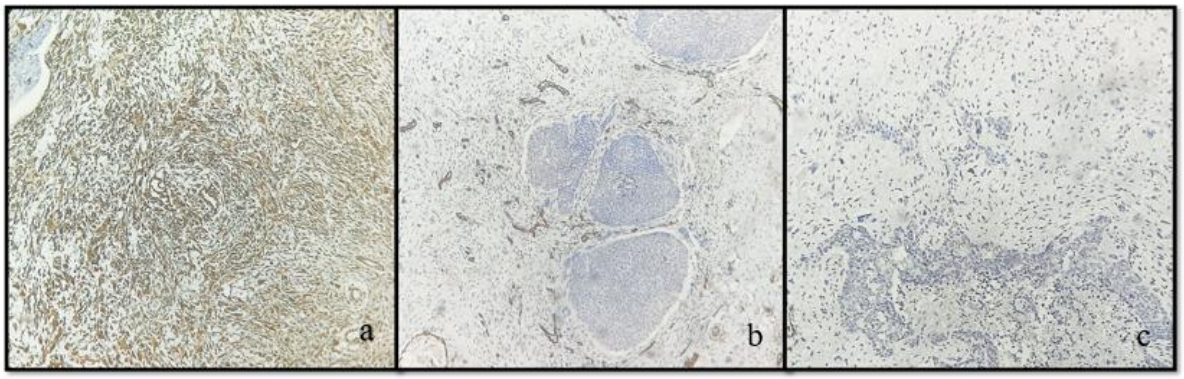


Fig.6: Immunohistochemistry studies a.) Strong cytoplasmic positivity for vimentin (Vimentin,100x); b.) Negative CD34 staining (CD34,100x); c.) Negative staining for S100 (S100, 100x)

Based on histopathological and immunohistochemical findings, a definitive diagnosis of oesophageal carcinosarcoma was established. The pathological stage was determined to be pT1b N0 Mx, according to the 8th edition of the American Joint Committee on Cancer (AJCC) staging manual. Postoperatively, the patient received three cycles of chemotherapy and remains under vigilant observation.

2. DISCUSSION

Carcinosarcoma, initially characterized by Virchow in 1865, is an uncommon and malignant neoplasm consisting of squamous epithelial and sarcomatous components. Oesophageal carcinosarcoma, referred to as pseudo-sarcoma or spindle cell carcinoma, is a mixed neoplasm characterized by the coexistence or spatial separation of carcinomatous and sarcomatous components within the tumour (1). Various hypotheses have been proposed regarding its development. The collision theory suggests that two separate stem cells independently undergo malignant transformation. The metaplastic theory suggests that both components originate from a common progenitor cell (2), whereas the third theory suggests that the sarcomatous component develops as a reactive response to carcinoma (3).

Carcinosarcoma constitutes a minor proportion of oesophageal cancers, varying from 0.5% to 2.8%, with increased incidence among middle-aged to elderly populations. It primarily affects the middle and lower segments of the oesophagus, possibly due to the prevalence of smooth muscle in these areas. The clinical presentation resembles that of other oesophageal malignancies, with prevalent symptoms such as dysphagia, weight loss, odynophagia, and anorexia (4,5). In contrast to conventional oesophageal carcinomas that demonstrate wall thickening, carcinosarcoma frequently manifests as a polypoid or protruding intraluminal mass, which can be a key feature for its identification (6).

Histologically, carcinosarcoma exhibits a biphasic architecture, comprising a squamous epithelial carcinoma component and a sarcomatous stroma consisting of spindle cells. Immunohistochemical analysis is essential for differentiating the dual components, with squamous regions generally exhibiting positivity for cytokeratin and p63, whereas the sarcomatous regions are vimentin-positive (5,7,9).

Oesophageal carcinosarcoma generally exhibits reduced local invasion compared to conventional squamous cell carcinoma, yet it may be more suitable for surgical resection even in advanced stages. Certain reports indicate a comparatively more favourable prognosis in relation to squamous cell carcinoma (4,10). Iyomasa et al. documented a 5-year survival rate of 27%, with results comparable to poorly differentiated squamous cell carcinoma; But carcinosarcoma shows more tendency for recurrence, mostly related to hematogenous metastases (11). This highlights the ongoing discussion regarding its harmful potential, as while local invasiveness may be reduced, the likelihood of distant spread remains significant (9).

Hashimoto et al. conducted a study revealing that among patients who underwent esophagectomy, overall survival rates were 73% at three years and 61.9% at five years, indicating that although many patients may survive for several years, long-term survival rates diminish over time (12). These findings highlight the necessity for long-term monitoring and the significant risk of recurrence, thereby underscoring the importance of rigorous post-operative surveillance.

The recent study by Yamauchi et al. elucidates the difficulties encountered in the treatment of oesophageal carcinosarcoma, especially regarding chemoradiotherapy. This case report describes an uncommon occurrence of rapidly proliferating carcinosarcoma subsequent to definitive chemoradiotherapy, which initially demonstrated a favourable response but later exhibited rapid tumour progression. This differential response is likely due to varying sensitivity of the carcinoma and sarcomatous components to chemoradiotherapy, with the carcinoma component exhibiting a favourable response while the sarcomatous component demonstrated resistance. An analysis of nine supplementary cases of oesophageal carcinosarcoma subjected to chemoradiotherapy indicated that although chemoradiotherapy may be effective for the squamous cell carcinoma component, its effectiveness diminishes for the sarcomatous component (13–17).

This underscores a considerable challenge in therapy, as the sarcomatous element is typically more resistant to chemoradiotherapy, resulting in rapid tumour proliferation and recurrence following initial intervention. Thus, the authors propose that surgical resection, frequently coupled with regional lymph node dissection, continues to be the most reliable curative strategy for cases exhibiting significant sarcomatous components (9).

3. CONCLUSION

Oesophageal carcinosarcoma is a rare and aggressive malignancy that necessitates thorough diagnostic evaluation due to its biphasic nature. Early detection through histological and immunohistochemical analysis is critical for accurate diagnosis and treatment. While surgical resection remains the most effective approach, adjuvant chemotherapy may enhance patient outcomes. Due to the high risk of recurrence and distant metastasis, long-term follow-up is crucial.

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