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FACIAL VENOUS TUMOUR THROMBUS FROM SUBMANDIBULAR GLAND MUCOEPIDERMOID CARCINOMA: AN ATYPICAL TUMOUR SPREAD IN HEAD AND NECK CANCER

Mariam Malik[1], Rana Bilal Idrees[1], Zeeshan Rashid Mirza[1], Sharifa Sikandar[1], Saba Nawaz[1], Maham Khalid[1], Muhammad Hamid Chaudhary[2]

[1] Radiology, INMOL Cancer Hospital, Lahore, Pakistan
[2] Cardiovascular Surgery, CPEIC, Multan, Pakistan

Corresponding author's e-mail: dr.hamid632@gmail.com

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ABSTRACT

Background: Tumour thrombus of the facial vein is an exceedingly rare complication arising from mucoepidermoid carcinoma of the salivary glands. Early detection is pivotal for appropriate management, as delays can lead to metastatic disease, worsening the prognosis.

Case description: We present a case involving a 76-year-old male with a history of mucoepidermoid carcinoma of the right submandibular gland, previously treated with surgical resection and radiotherapy. The patient, a long-term worker in a rubber factory, presented with a painless, firm swelling in the right cheek, persisting for three months. Contrast-enhanced computed tomography (CECT) showed distended facial vein with enhancing thrombus confirmed by sonographic correlation demonstrating intralesional vascularity. Cannon ball pulmonary nodules were also noted. Radiological findings led to a core biopsy, confirming tumor thrombosis of the facial vein due to mucoepidermoid carcinoma. However, the patient declined a biopsy of the pulmonary nodules, and has been referred to oncology for further management.

Conclusions: This case highlights the critical importance of considering venous tumour thrombus in patients with previous salivary gland malignancies presenting with new or persistent facial swellings. It emphasises the role of advanced imaging techniques in the early identification of this rare entity. Additionally, it stresses the need for healthcare providers to engage in thorough discussions with patients about the potential consequences of forgoing recommended treatments, reinforcing the need for vigilance in monitoring such patients.

KEYWORDS

Mucoepidermoid carcinoma, facial veins, venous thrombosis, submandibular gland neoplasms, salivary gland neoplasms

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LEARNING POINTS

- Tumours of head and neck may cause thrombosis of veins by direct invasion resulting in a tumour thrombus, or indirectly by exerting a mass effect and vein compression.
- These can be distinguished by contrast-enhanced computed tomography (CECT) or magnetic resonance imaging (MRI).
- Doppler ultrasound may show patchy neovascularisation in a tumour thrombus, which would be absent if thrombosis was caused by compression.

INTRODUCTION

Tumours of the head and neck may cause thrombosis of veins by direct invasion resulting in a tumour thrombus, or indirectly by exerting mass effect and compression of the vein leading to stasis and consequently a bland thrombus. The distinction between the two may be made on contrast-enhanced cross-sectional studies: contrast-enhanced computed tomography (CECT) or magnetic resonance imaging (MRI), where a tumour thrombus would show post-contrast enhancement while bland thrombus would remain non-enhancing. Doppler ultrasound shows patchy neovascularisation in a tumour thrombus, which would be absent in the case of a bland thrombus. The management strategy of a tumour and a bland thrombus would differ, therefore differentiation between the two is essential.

Tumour thrombus in neck veins is infrequently seen and when present has most often been observed in thyroid malignancies[1]. Its incidence in salivary gland tumours is extremely rare and to date, no exact information on the exact number of reported cases is available. Similarly, no exact data are available with regards to age distribution or gender predilection; however, the published case reports of salivary gland tumours with associated tumour thrombus are mostly of older patients and the most frequent salivary gland reported is parotid. The tumour thrombus may be apparent clinically or radiologically, and is a poor prognostic indicator[2] but because of its rare occurrence, the clinical presentation as well as management and complications have not been clearly established.

The salivary gland malignancies comprise 2% to 5% of head and neck cancers[3]. Mucoepidermoid carcinomas are the commonest malignant tumours of salivary glands in all age groups. These can have a range of presentation from having no symptoms to being metastatic or locally aggressive. The venous drainage of the submandibular gland is in the facial and sublingual veins, which subsequently drain into the internal jugular vein. Hence direct venous invasion can manifest as a tumour thrombus in these venous structures and should be kept in differential diagnosis in patients with known submandibular gland carcinoma.

CASE DESCRIPTION

We report a 79-year-old male patient who has a long-standing history of working in a rubber factory. The patient was treated for a mucoepidermoid carcinoma of the right submandibular gland four years ago, which was surgically resected followed by 33 sessions of radiotherapy. The patient also complained of a firm, non-tender swelling in the right cheek since the time of presentation, which has remained unaddressed. This has been progressively increasing in size. On examination the swelling was hard and mobile. Contrast-enhanced MRI could not be performed as the patient is claustrophobic, therefore contrast-enhanced computed tomography (CT) face and neck was advised for work-up of locoregional residual or recurrent disease process.

A retrospective review was made of preoperative CECT face and neck performed three years previously, which revealed a mass replacing the right submandibular gland consistent with the biopsy-proven mucoepidermoid carcinoma. An additional note was made of an enhancing tubular structure adjacent to right submandibular gland, not reported by the radiologist; however, detailed evaluation was not possible as only limited hard copy sections are available *(Fig. 1)*.

The current post-surgical CECT of face, neck and chest demonstrated a surgically absent right submandibular gland *(Fig. 2A)*. There is a tortuous intensely enhancing tubular

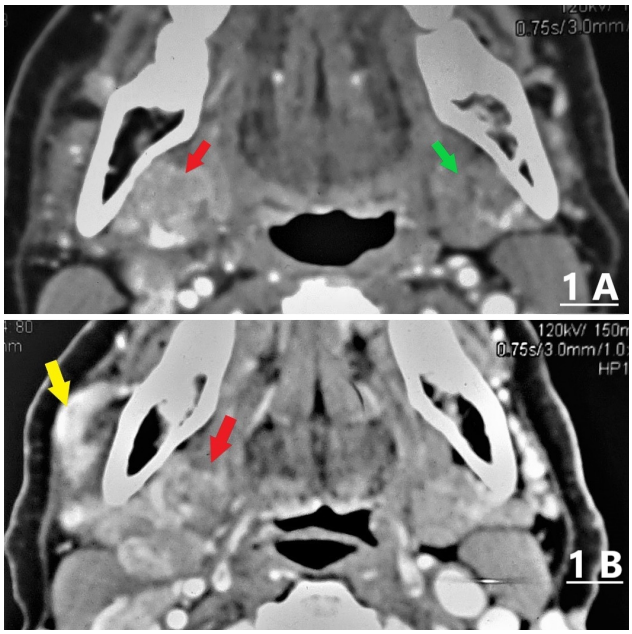

Figure 1. Presurgical CECT axial images. Figure 1A demonstrates that the right submandibular gland has been replaced by a heterogeneously enhancing mass (red arrow). There is a normal looking left submandibular gland (green arrow). Figure 1B demonstrates right submandibular gland mass (red arrow) and tumour thrombosis of the right facial vein (yellow arrow). This was missed by the reporting radiologist.

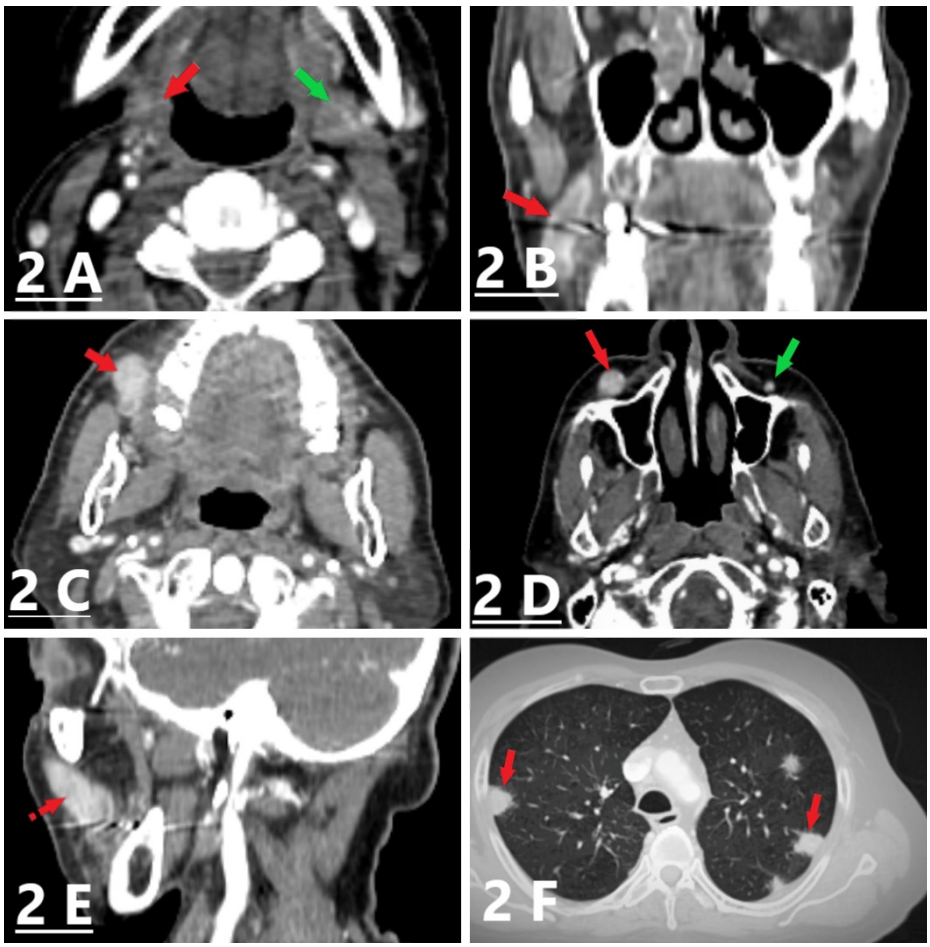

Figure 2. Post-surgical CECT images. Normal left submandibular gland (A, green arrow). The right submandibular gland has been removed surgically (red arrow); B) Coronal, and C, D) axial CECT images; E) Sagittal CECT section demonstrating the tumour thrombus distended tortuous right facial vein, shown by a red arrow; D) Section showing normal calibre contrast opacified left facial vein for comparison (green arrow); F) Post-surgical axial CECT image demonstrating the multiple masses in the lungs bilaterally (red arrow), highly suspicious for pulmonary metastasis.

structure extending from the surgical bed following an upward trajectory within the subcutaneous buccal fat *(Fig. 2B and C)*. This passes over the ramus of the mandible and anterior to the right half of the alveolar arch of the maxilla, lying anteromedial to the right masseter muscle. The lesion extends up to the level of medial angle of the right eye. Considering its tubular tortuous appearance, described course and comparison with the contralateral facial vein, findings are suspicious for tumour thrombosis of right facial vein *(Fig. 2B, C, D and E)*. Sections through the chest reveal multiple variable size soft tissue density nodules and masses bilaterally, highly suspicious for pulmonary metastasis *(Fig. 2F)*.

Correlative ultrasound shows a hypoechoic soft tissue mass contained within the enlarged facial vein and colour Doppler reveals intense intralesional vascularity, suggestive of extensive tumour thrombosis *(Fig. 3)*. Based upon the radiological findings, the treating physician advised core biopsy of the facial vein thrombus. Core biopsy was performed under ultrasound guidance and histopathology revealed a tumour thrombosis of mucoepidermoid carcinoma *(Fig. 4)*. For the confirmation of pulmonary metastasis, the patient was also advised to undergo the CT guided biopsy of the lung masses; however, he refused any further intervention.

DISCUSSION

Direct extension of a head and neck tumour into a neighbouring vein is rare and very few cases of its occurrence have been published to date. Among the tumours that have been reported to invade into the neck veins are mucoepidermoid carcinoma, paraganglioma, myoepithelial carcinoma and squamous cell carcinoma.

Fluorodeoxyglucose positron emission tomography (FDG PET) can show increased avidity in a neck vein with or without a tumour thrombus, therefore increased uptake itself is not a reliable indicator for tumour thrombus. Modalities such as ultrasound Doppler, contrast-enhanced CT and MRI may aid the identification of the thrombus as well as the extent of tumour extension and provide clues towards it being malignant by demonstrating the presence of neovascularity on ultrasound Doppler and post-contrast enhancement in cross-sectional imaging. For definite diagnosis, histopathology remains the gold standard. The differentiation between bland versus tumour thrombus is crucial in surgical planning and radiation field selection.

Tumour thrombus formation is favoured by biological factors such as platelet activation, uncontrolled platelet degranulation, modified thrombosis/ fibrinolysis mechanisms, vascular endothelial growth factors, platelet-derived growth factors and epidermal growth factors[2]. All of these are present in head and neck cancers. Follicular carcinoma of the thyroid is the commonest head and malignancy involved in the development of tumour thrombus at an estimated rate of 1.3%[2], which may be attributed to its tendency for haematogenous spread. The rate of tumour thrombosis by other malignancies such as the salivary

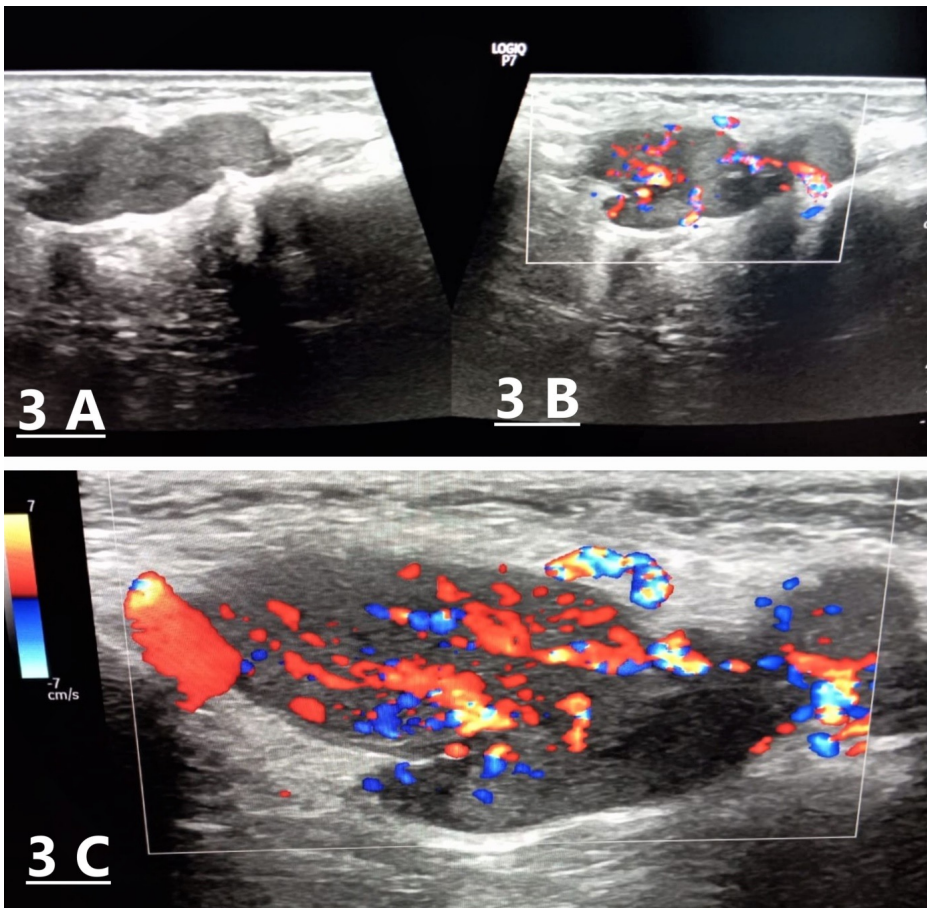

Figure 3. A) Greyscale ultrasound image showing lobulated tubular structure containing heterogeneous hypoechoic thrombus grossly distending the right facial vein with resultant occlusion; B) The colour Doppler image shows intense vascularity within the thrombus as shown by coloured areas; C) The focused colour Doppler ultrasound image through the right facial vein demonstrates vivid intrathrombus vascularity. No free flow is otherwise present in the background right facial vein (not shown in the figure).

gland tumours have not been documented owing to the rare incidence. Smoking, viral infections, ionising radiations and occupational exposure of products such as rubber or asbestos are all predisposing factors for development of mucoepidermoid carcinomas. The histopathological grading provides an insight into the malignant potential of these tumours with high-grade tumours being the most aggressive. Mucoepidermoid carcinomas typically metastasise by the lymphatics in preference to the haematogenous pathway, hence surgical management is directed towards neck node dissection. Haematogenous spread by direct invasion is extremely rare and is an independent risk factor for metastasis, therefore its presence prompts urgent surgical resection and chemotherapy. The Radiation Therapy Oncology Group (RTOG) 1008A is also investigating the role of combined chemotherapy and radiotherapy with radiotherapy alone for treatment of tumours of salivary glands[4]. If the treatment is delayed, prognosis is worsened by development of distant metastasis[2].

Our case highlights the importance of keeping in mind the possibility of this rare occurrence as early detection improves the treatment outcome and hence the prognosis. Given the venous drainage of the submandibular gland into the facial vein, the thrombosis of this vein ipsilateral to the site of prior resected primary mucoepidermoid tumour raises strong suspicion for malignancy invasion and consequently resulting in tumour thrombus formation. The finding is further augmented by the presence of an enlarged right facial vein distended with thrombus, intra thrombus vascularity on colour Doppler and post-contrast enhancement of the thrombus on CT scan. Furthermore, the presence of multiple nodules and masses in lung parenchyma

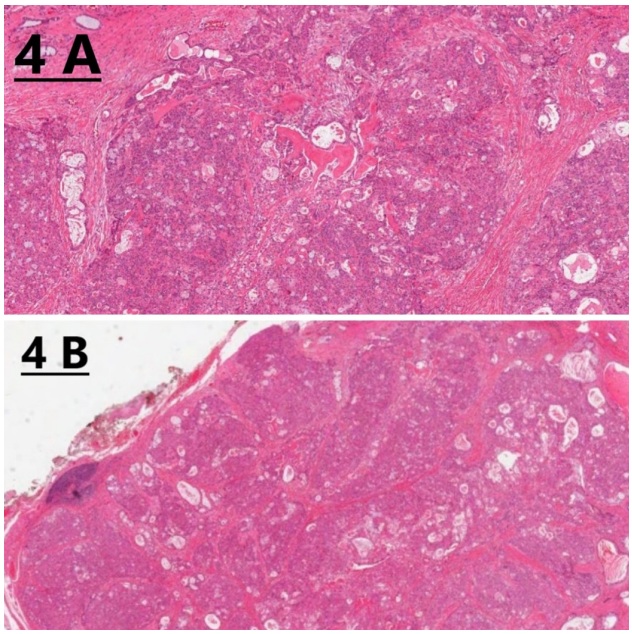

Figure 4. Histopathology of the core biopsy specimen obtained from the facial vein thrombus. Sections examined reveal a glandular growth pattern with varying proportions of solid nests of epidermoid cells, intermediate cells and mucocytes. The mucus cells are embedded in epidermoid cell nest or lining cystic spaces. Intermediate cells are found within epidermoid cell nests or form separate nests. Luminal pools of mucin are also seen.

bilaterally raises a strong suspicion of pulmonary metastasis, which unfortunately could not be confirmed by histopathological analysis secondary to patient's refusal for further intervention. Hence after the confirmation of tumour thrombosis by core biopsy performed under ultrasound guidance, the patient was referred to oncology for further management.

CONCLUSION

Tumour thrombus of the facial vein as a complication of salivary gland mucoepidermoid carcinoma is a rare entity. Recognition and knowledge of this phenomenon is essential for timely recognition and treatment. Untreated cases can progress to metastatic disease, which worsens the prognosis. In the absence of metastatic disease, treatment is mainly by surgical resection of the involved vein.

REFERENCES

1. Yap D, Dragan A, Weller A, Tatla T. Rare case of venous tumour thrombus from acinic cell carcinoma of the parotid gland. *Ann R Coll Surg Engl* 2020;**102**:e161–e166.
2. Dekhou A, Rehman R, Parzen JS, Quinn TJ, Zhang PL, Rontal M, et al. Primary parotid tumor thrombosis: immunohistologic features and awareness of metastatic potential. *Cureus* 2021;**13**:e16174.
3. Flavell-Birch A, Brasch HD, Tan ST. Parotid mucoepidermoid carcinoma with extensive intravenous metastasis: a case report. *AJOPS* 2021;**4**:71–74.
4. Radiation Therapy Oncology Group. RTOG 1008 protocol information: a randomized phase II/phase III study of adjuvant concurrent radiation and chemotherapy versus radiation alone in resected high-risk malignant salivary gland tumors. Published November 5, 2015.