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Case Report

Phrenic nerve palsy as a complication of superior vena caval stenting

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ABSTRACT

Superior vena cava obstruction typically results from either primary pulmonary malignancies, lymphoma, or fibrosis related to central catheters. Endovascular stenting of superior vena caval obstruction is a common first approach, due to the rapid clinical improvement typically seen. The commonest complications are recurrence of obstruction and stent migration. We present herein the case of a phrenic nerve palsy secondary to endovascular stenting in a patient with superior vena cava obstruction due to primary small cell lung cancer.

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Introduction

We present a case of phrenic nerve palsy as an unusual complication of superior vena caval (SVC) stent placement. SVC obstruction is a syndrome characterized by cervico-facial, upper limb and thoracic oedema, dyspnoea, dysphagia, cough, collateral thoracic circulation, hoarseness, and occasionally, cerebral oedema manifested by cognitive deficits and visual disturbance [1–3]. The commonest causes are primary pulmonary malignancies, classically small cell lung cancer (SCLC) [4]. Non-Hodgkin's lymphoma, and less commonly Hodgkin's lymphoma are other causes [4]. Benign causes include fibro-

sis around the SVC following long term central catheters [5]. Given the common causes of SVC obstruction are malignant, the life expectancy of patients is typically around 6-7 months. The previously described management of SVC obstruction, where cerebral oedema is not a concern, includes chemotherapy, radiotherapy, and SVC stenting. Endovascular stenting is an excellent choice of initial management, as it does not preclude the option of chemotherapy or radiotherapy, provides the most rapid resolution of SVC obstruction, has a low complication rate, and is well tolerated by patients [6]. The efficacy of stenting approaches 95%, whereas chemotherapy and radiotherapy figures are up to 85% and may be lower depending on the particular pathology and patient characteristics [4].

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The most commonly reported complications of SVC stents are: recurrence of venous obstruction by either thrombosis or tumour invasion or compression, stent migration, and fracture of stent wires [3]. The overall rate of major complications is 4%, and death is 2% [7]. Stent migration and wire fracture are now uncommon with modern stents. Unusual complications reported include fever, cellulitis at the access site, pain during balloon inflation, and a single previously reported case of transient hemidiaphragm elevation after stent placement [8]. This is the only other reported case of phrenic nerve injury following SVC stent placement we are aware of. In the previous case, the stent was inserted after radiotherapy for bronchial carcinoma, and the chest radiograph finding of elevated right hemidiaphragm was shown to be resolved on repeat imaging 24 hours after the initial radiograph. It is not reported whether the patient experienced symptoms suggestive of the complication, and the initial radiograph was only taken 24 hours after the initial procedure [8]. The authors of this case, report the finding likely related to compression of the phrenic nerve against surrounding tumour, as opposed to damage caused directly by venoplasty and stent placement.

Various regimens of anticoagulation and antiplatelet therapies before, during and after stent placement have been described, including no anticoagulation, low dose heparin infusions, heparin followed by months-long anticoagulation, and antiplatelet therapy only. No authoritative consensus on the use or nonuse, and the particular regimens of the same, have been suggested to date.

Case report

A 78-year-old woman initially presented to the emergency department after 10 days of symmetrical facial and neck swelling. She had a tooth extracted 7 days into the initial 10 days of swelling, as she thought it may have been related to a dental abscess. This made no difference to the swelling. The tooth extraction examination did not have any findings suggestive of an abscess or infection. She did not have systemic signs or symptoms of infection, she was afebrile (36.2), without rigors, airway or respiratory function compromise, and had a normal white cell count and CRP. Her swelling was not responsive to an outpatient course of fexofenadine (Telfast, Sanofi-aventis, Macquarie Park, NSW, Australia), or a course of oral amoxicillin (APO-amoxycillin, Apotex PTY LTD, Macquarie Park, NSW, Australia)

She was admitted under the plastic surgery department and treated with amoxicillin, metronidazole, and gentamicin. A computed tomography (CT) of her facial bones showed a 6mm lucency within the maxilla posteromedial to the recent maxillary second right incisor (26) tooth extraction with the impression of a 2 mm direct communication with the underlying right maxillary antrum, as well as generalized right maxillary mucosal thickening. Generalized cellulitis within the soft tissues of the face, without focal abscess or collection was also seen on this scan. An orthopantomogram showed a tooth 16 periapical cyst which was thought to be potentially related to her symptoms, though was not removed due to the risk of oro-antral fistula. She was discharged after 3 days of IV antibiotics,

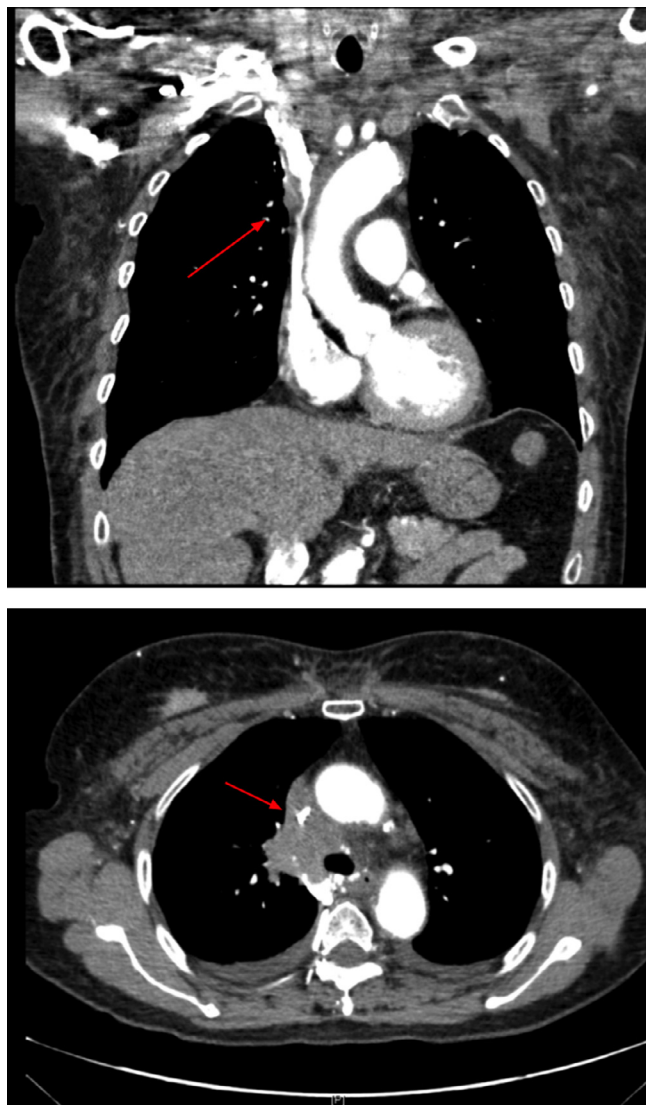


Fig. 1 – CT-coronal view and axial views demonstrating SVC compression by the surrounding mediastinal tumour.

on oral antibiotics. The facial swelling was unchanged at this point.

She represented after 6 days, with worsening facial, neck, and now bilateral upper limb, and upper thoracic swelling with associated dyspnoea, and reduced neck mobility. She also reported a new cough, and dysphagia without odynophagia, to liquids but not solids. On examination at this point, she had areas of distended superficial veins on the upper thoracic wall.

A right supraclavicular mass with mediastinal invasion was seen on chest CT, most suggestive of primary small cell bronchogenic malignancy, or less likely, lymphoma. There was severe SVC narrowing and complete compression of the azygous arch with features of SVC syndrome (see Fig. 1 below). A small nonocclusive thrombus in the right subclavian vein was also seen. There was associated right hilar lymphadenopathy.

The patient proceeded to endobronchial ultrasound, and fine needle aspirate cytology from this procedure later revealed primary small cell lung carcinoma. Later on the same

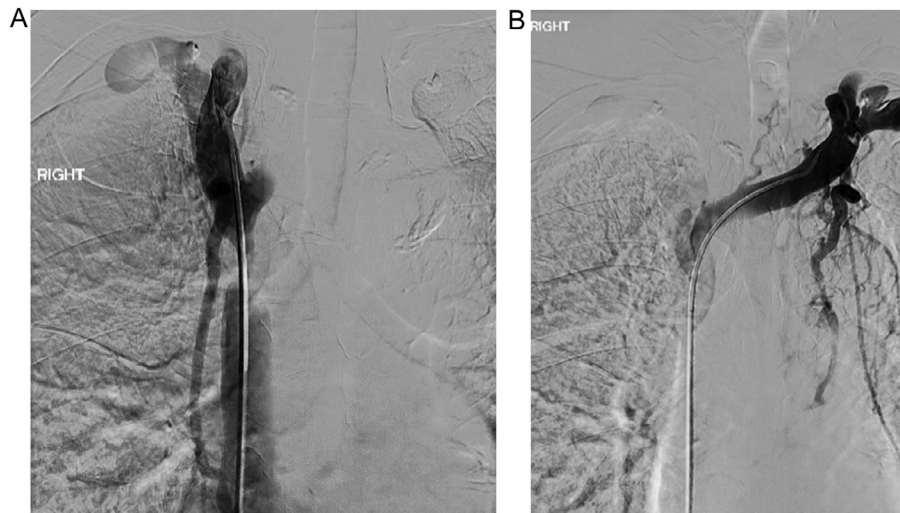


Fig. 2 – A (left) and B (right)—venography demonstrating flow limiting high grade stenosis in the upper and mid SVC with reflex and flow into collateral veins.

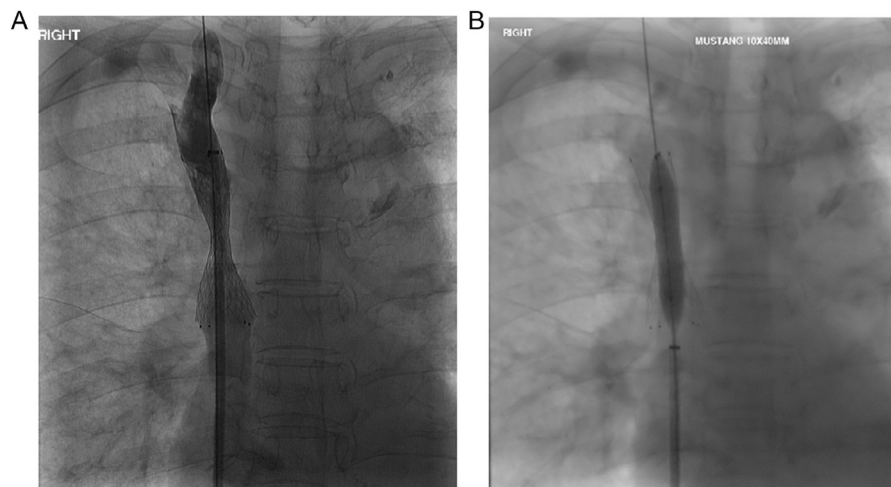


Fig. 3 – A (left) and B (right). Immediate venogram postdeployment of the sinus SL stent. A balloon plasty is performed on the right, in order to widen the narrowest obstructing point.

day, the patient was taken to the angiography suite with a view to placing an SVC stent across the obstruction. The stenosis was crossed with a 150 cm Bentson guidewire (Boston Scientific, Marlborough, MA) and a 5-French, 80 cm long, balloon dilatation catheter (Cook Medical, Bloomington, IN) via 10-French, 40 cm Introducer set (Cook Medical, Bloomington, IN) sheath in the right common femoral vein. Venography performed from both brachiocephalic veins confirmed the 84% stenosis of the upper to mid SVC (see Fig. 2a and b). Retrograde flow from the stenosis was evident with multiple collateral veins. A 100 cm long 10-French sheath (Optimed, Ettlingen, Baden-Wurttemberg, Germany) was advanced across the stenosis. A 20 × 60 mm sinus SL stent (Optimed, Ettlingen, Baden-Wurttemberg, Germany) was then deployed. Following deployment, the vein was conservatively venoplastied twice to an end diameter of 12 mm (see Fig. 3). Although the waist of the stenosis persisted, venography demonstrated markedly

improved antegrade flow (Fig. 4), therefore the procedure was terminated. A Heparin infusion (Heparin Sodium, Pfizer, West Ryde, Australia) at 500 units/h was commenced and the patient was sent to recovery.

During the postprocedure period, the patient became hypertensive (systolic > 200 mm Hg), and experienced dyspnoea, nausea and pain in her jaw, shoulder, and neck. She was also mildly dyspnoeic. A chest radiograph at this time showed an elevated right hemidiaphragm compared with a chest radiograph performed during the emergency admission the day prior (Figs. 5 and 6).

The elevated right hemidiaphragm and associated clinical findings of dyspnoea and pain, were presumably related to a phrenic nerve palsy likely secondary to the plasty performed as part of the SVC filter placement. This provisional diagnosis was supported by a chest radiograph without diaphragmatic eventration less than 24 hours prior, and a new finding



Fig. 4 – Postballoon dilatation, the stent remains with a waist, and good flow is demonstrated across the previously near-obstructed point.

imaging finding which also coincided with the patient's clinical syndrome. The right phrenic nerve enters the thoracic inlet between the right subclavian artery and brachiocephalic vein, before marginating the lateral border of the SVC, making it highly vulnerable in anatomic position, to extrinsic compression from SVC venoplasty against the CT demonstrated tumour encasing it. The radiograph performed in recovery did not show other clear causes for the sudden onset dyspnoea, and cardiac monitoring patterns were not suggestive of an acute coronary syndrome. Her hypertension and pain were treated with clonidine (APO-clonidine, Apotex PTY LTD, Macquarie Park, NSW, Australia) and fentanyl (Fentanyl, Teva Pharma, Macquarie Park, NSW, Australia), respectively. The patient was admitted to a monitored bed in the HDU. She

remained stable and was saturating at 97% on 4 L oxygen via nasal prongs.

Subsequent CT study of her brain and abdomen and an Fludeoxyglucose Positron Emission Tomography (FDG PET) scan, did not demonstrate any evidence of metastatic disease, and she was commenced on carboplatin and etoposide for the new diagnosis of SCLC. She remained well, hemodynamically stable and had ongoing clinical improvement of the facial, upper limb, and thoracic oedema and dyspnoea.

A repeat chest radiograph on day 1 postprocedure showed persistent elevation of the right hemidiaphragm, though this did not manifest as any obvious clinical finding. It also persisted on the FDG PET acquired a month later.

She unfortunately experienced heparin induced thrombotic thrombocytopenic syndrome secondary to the heparin she received as part of her management. This was incidentally found in her pathology investigations. On 3 day postdischarge follow-up, her clinical syndrome related to SVC syndrome had resolved to premorbid levels, and she remained well from a respiratory perspective.

Discussion

Management of SVC obstruction in patients with malignancy as a cause can be achieved with stenting, chemotherapy, radiotherapy, or a combination of these. SVC stenting is a highly effective and rapid method achieving relief of what is a distressing syndrome, in patients with a relatively short life expectancy, and thus has been suggested as an initial step in management [6]. In the present case, we experienced a rare complication of the procedure, a neuropraxia resulting from either the stretching of the nerve or compression between the stent, and the tumour.

Given the ongoing debate regarding stenting as a first option, the possibility of awaiting a cell diagnosis after endobronchial ultrasound and fine needle aspiration, with a view to chemotherapy as a first step, was discussed during the case. In the present case, there was evidence of thrombosis superior to the obstruction on initial imaging (thrombus within the right subclavian vein). The patient also had onset of dysp-

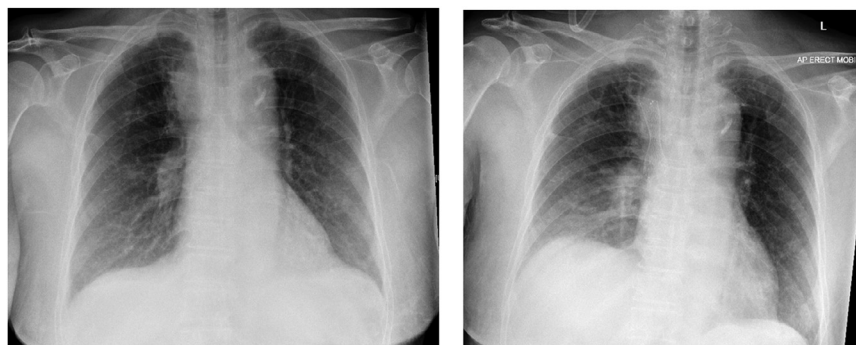


Fig. 5 and 6 – On the left is the chest radiograph at presentation, prior to SVC stent deployment. On the right in the study acquired around the time the patient became dyspnoeic and experienced new pain, postprocedure. The stent can be seen in place and has not migrated, The right hemidiaphragm is now elevated.

noea and hoarseness on presentation, which were concerning for early signs of airway compromise. There was no concern for cerebral oedema, and hence options such as hyperventilation, mannitol, and other supportive measures were not implemented. Given that chemotherapy can typically provide symptom relief in 7–15 days [4], stenting has probably been the most sensible option in this case, and the patient did experience rapid and persisting relief of the syndrome as expected. She was also able to commence chemotherapy without any delay.

In the previously described phrenic nerve palsy related to stenting, the hemidiaphragm elevation resolved on chest radiograph 24 hours after being demonstrated. In the current case, the palsy presented itself as a syndrome of neck and jaw pain, dyspnoea (though hard to distinguish from the confounding cause), and a subsequent chest radiograph demonstrating the associated anatomy. The follow-up chest radiograph and FDG PET showed persistent elevation of the diaphragm, however the patient's symptomatology resolved.

The distribution of shoulder, neck, and jaw pain, may have been due to extrinsic compression of the phrenic nerve. The phrenic nerve contains motor (around 70% of fibers), and sensory fibers to the central diaphragm, pericardium, and pleura. Although abdominal diaphragmatic irritation is well known to cause C4 dermatomal distribution pain, irritation, or damage to thoracic structures such as the pericardium and pleura are less well established to demonstrate shoulder tip pain.

We reaffirm the notion that SVC stenting is a reasonable first management option in patients presenting with SVC obstruction. Our case demonstrates that the technique is highly effective in providing relief of the acute and subacute results of obstruction, and that definitive tissue diagnosis and initiation of therapy need not be delayed. Further discussion surrounding the use of anticoagulants and antiplatelets is warranted, given that previously reported complications of the procedure have been related in part to the use of these, and we have experienced the same in the current case. To our knowledge, no formal retrospective or prospective evaluation

has been completed looking specifically at outcomes in cohorts who have received anticoagulation and those who have not.

Supplementary materials

Supplementary material associated with this article can be found, in the online version, at doi:[10.1016/j.radcr.2019.03.033](https://doi.org/10.1016/j.radcr.2019.03.033).

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