

CASE REPORT

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Rare case of calcified fibrin sheath causing central venous occlusion, mimicking a retained catheter fragment

Sze Yuen Lee^{*} , Yamunadevi Arunasalam and Sri Idayu Mohamad

Abstract

Background Long-term placements of central venous catheters (CVCs) are known to cause various complications, among which include fibrin sheath formation, causing catheter dysfunction, infection and thrombosis. Post-catheter removal, these sheaths may sometimes be retained in the vein; however, are rarely calcified. When retained sheaths are calcified, they may cause diagnostic confusion on imaging, as they may mimic a retained catheter fragment.

Case presentation We report a case of a patient with end-stage renal disease and a history of multiple prior CVC insertions presenting with symptoms of central venous occlusion post-revascularization of his stenosed arteriovenous fistula. CT venography revealed a linear tubular hyperdensity within the right brachiocephalic vein and superior vena cava, which at first glance looked like a retained catheter fragment. However, further scrutiny revealed a retained calcified intravascular fibrin sheath as the cause of occlusion.

Conclusions Awareness of the radiological discriminating features of calcified fibrin sheaths are important to prevent misdiagnosis and unnecessary interventions as these sheaths may mimic retained catheter fragments. Calcified fibrin sheaths should also be considered as a differential diagnosis of radio-opaque intravascular structures or venous calcifications post-CVC removal.

Keywords Central venous obstruction, Fibrin sheath, Vascular calcification, Renal dialysis, Central venous catheters, Thrombosis, Case report

Background

Central venous catheters (CVC) are commonly used for various purposes, including drug administration, parenteral nutrition, fluid resuscitation, long-term venous access, blood taking, central venous pressure monitoring as well as hemodialysis. They are commonly inserted in the internal jugular, subclavian and femoral veins. Fibrin sheaths, also known as catheter-related sheaths, sleeves or cast are a common complication, with varying reported incidence ranging from 10 to 56% in radiology

case series to as high as 100% in experimental studies [1–3]. These sheaths may cause various complications such as infection, venous thrombosis and even pulmonary embolism [1, 4]. Retained fibrin sheaths post-catheter removal are less common and are rarely calcified. Krausz et al. [4] reported that retained fibrin sheaths were present in 13.6% of 147 patients studied, of which 45% were calcified. When calcified, they may mimic a retained catheter fragment, causing diagnostic confusion and unnecessary interventions. We report a case of a retained calcified intravascular fibrin sheath causing central venous obstruction, mimicking a retained catheter fragment, which was diagnosed 5 years after removal of a long-term tunneled dialysis catheter in an adult with end-stage chronic renal disease.

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Case presentation

A 33-year-old male with underlying renal failure was referred from a dialysis center to our district hospital due to acute shortness of breath 3 h post-hemodialysis. He also complained of progressive neck and facial swelling for the past 2 weeks following a revision done to his arteriovenous fistula (AVF).

He had been on dialysis for the past 8 years since being diagnosed with end-stage renal failure secondary to nephrotic syndrome. Due to primary failure of his right brachiocephalic fistula, he was dialyzed via peritoneal dialysis. However, after multiple episodes of infection, he was subsequently converted to hemodialysis via a CVC, which was changed every 2 weeks. He then had a long-term tunneled CVC insertion into his right internal jugular vein (IJV) for hemodialysis, which lasted for 2 years and was removed 5 years ago. Since then, he was dialyzed via a left brachiobasilic fistula (BBF). Due to slow flow and reduced thrill in the fistula for about 1 month prior to presentation, he was referred to a private AVF center for assessment. An ultrasound done during that time noted severe juxta-anastomosis intimal hyperplasia. Exploration of the basilic vein at left distal arm was done with shortening of the vein and placement of a 3-cm (polytetrafluoroethylene) PTFE patch graft juxta anastomosis 2 weeks prior to presentation. No fistulogram was done at that time.

Initial chest radiograph done on admission showed no significant lung pathology apart from mild blunting of the left costophrenic angle likely due to mild left pleural effusion. Due to his complaint of progressive facial and neck swelling, central venous occlusion was suspected and he was referred to our radiology department for a thoracic CT venogram as our center

did not have fluoroscopic services to perform a diagnostic catheter venogram. A CT scan of the thorax on a 16-slice scanner was done in the unenhanced, first pass and delayed venous phase with standard image reconstruction algorithms.

On the initial unenhanced CT scan, a linear tubular hyperdensity measuring approximately 6.2 cm in length was seen within the right brachiocephalic vein extending to the superior vena cava (SVC), which at first glance was concerning for a retained catheter fragment (Fig. 1). Subsequent contrasted phases showed stenosis and non-opacification of bilateral brachiocephalic veins near the confluence extending to the upper SVC (Fig. 2). The lower SVC was reconstituted from collaterals via right superior intercostal and azygos veins, which was better seen on the first-pass phase (Fig. 3). The rest of the SVC and superior cavoatrial junction were patent with no filling defects or stenosis. The inferior vena cava (IVC) was also patent and normal in caliber. On further scrutiny, in view of the discontinuous nature of the calcifications, a diagnosis of a calcified fibrin sheath causing chronic central venous stenosis was made. Retrospective examination of the initial chest radiograph showed a faint calcified tubular structure in the right paratracheal region (Fig. 4) corresponding to the calcified fibrin sheath.

The patient was then referred to the vascular surgery team at the nearest tertiary hospital in which ligation of the revascularized AV fistula was performed. Currently, the patient is being dialyzed via a long-term tunneled femoral dialysis catheter and remains well.

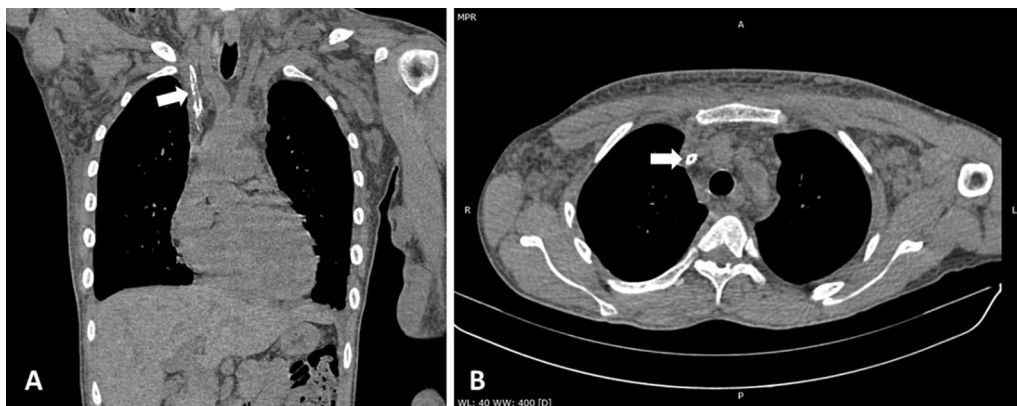


Fig. 1 Unenhanced coronal and axial CT scan of the thorax showing a hyperdense linear tubular structure (white arrows) measuring approximately 6.2 cm in length within the right brachiocephalic vein extending to the superior vena cava (SVC), which at first glance was concerning for a retained catheter fragment. However, the discontinuous and interrupted appearance of this hyperdense structure, with a visible lumen suggested a calcified fibrin sheath instead

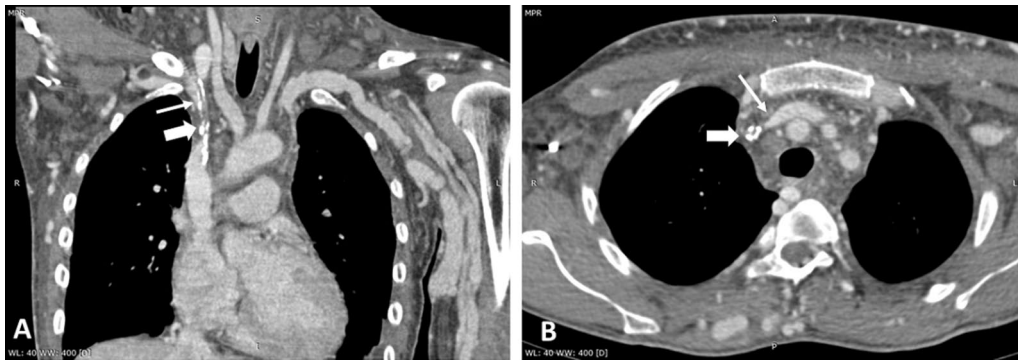


Fig. 2 Contrasted delayed-phase CT venogram in coronal and axial view showing stenosis and non-opacification of bilateral brachiocephalic veins (thin white arrows) near the confluence extending to the upper SVC. The retained calcified sheath (thick white arrows) showed no contrast opacification within



Fig. 3 First-pass phase of CT Thorax in axial and coronal view showing the dilated collateral veins, namely the azygos veins (thick white arrows) and the right superior intercostal veins (thin white arrows)

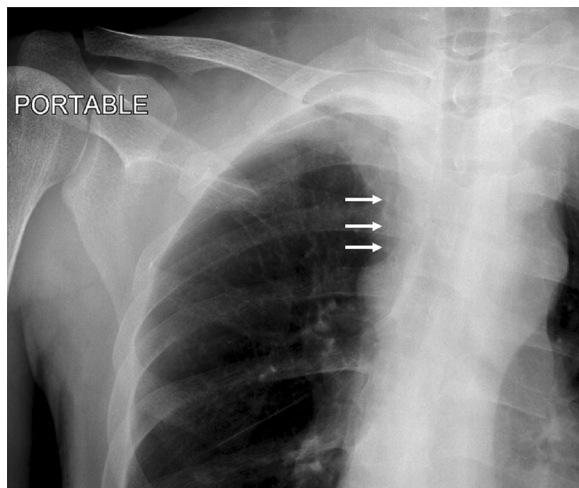


Fig. 4 Retrospective examination of the initial chest radiograph showed a faint calcified tubular structure in the right paratracheal region (white arrows) corresponding to the calcified fibrin sheath

Discussion

Fibrin sheaths are a well-known complication of central venous catheters, with the earliest descriptions dating back to 1964 [2]. These sheaths can form as early as 24 h post-insertion and can encase the entire length of the catheter by 5–7 days [1, 3]. They usually arise at two sites of endothelial injury, namely at the venous entry site of the catheter as well as at the catheter tip, where there is contact with the venous wall [1]. Forauer et al. [5] described two microscopic patterns of histological changes, in which focal intimal injury and pericatheter thrombus formation were seen in short-term catheters, while smooth muscle proliferation, venous wall thickening with focal catheter attachment to the vessel wall, organization of thrombus and collagen formation were observed in long-term catheters.

These sheaths can be retained post-catheter removal and are sometimes calcified as reported by Krausz et al.

They found that 21% of these fibrin sheaths remnants were in patients with end-stage renal disease, likely due to the larger diameter of the dialysis catheters, which increased the degree of traumatic contact with the venous wall as compared to the smaller caliber of CVCs and peripherally inserted central catheters (PICC) used for other purposes. These dialysis catheters were also typically left in situ for longer periods, predisposing to sheath formation [4]. In a study of tunneled dialysis catheter exchange and de novo placements by Shanaah et al. [6], they noted that fibrin sheath incidence was 47%. Matusik et al. [7] found 17 case reports of CVC-related fibrin sheaths, of which eight cases were in hemodialysis patients with retained calcified fibrin sheaths, some initially misinterpreted as a retained catheter tip. In this case, our patient had a history of multiple CVC insertions and exchanges as well as a long-term tunneled dialysis catheter which was left in situ for 2 years, increasing his risk of fibrin sheath formation and retention.

Imaging findings of fibrin sheaths on fluoroscopic catheter venography have been well described [8] and well known among radiologists. This is due to the common complication of fibrin sheaths causing catheter dysfunction, thus requiring catheter venography for further evaluation. However, not many may be familiar with the CT imaging features of retained catheter sheaths, causing diagnostic confusion especially when calcified as they may mimic a retained catheter fragments due to the hyperdense tubular configuration of the sheaths as seen in several case reports [9–11]. Commonly reported CT findings of calcified retained sheaths are an irregularly shaped discontinuous or interrupted tubular structure, similar to or nearing bone density with a patent lumen within, as well as with a diameter greater than expected for a CVC [4, 7, 10, 11]. In some cases, contrast material was seen to pass through the lumen [12]. The contrast material may sometimes be isodense to the sheath, making them inconspicuous on contrast-enhanced CT scan, and are therefore better visualized on non-contrasted scans [4, 7]. On the other hand, a fractured catheter would be seen as a continuous tubular structure, and due to the spatial resolution of CT scanners, the lumen of a retained catheter fragment may not be well-visualized, especially when the lumen is less than 2 mm. Therefore, Baciarello et al. recommended examining the presence or absence of a lumen as a criterion for differentiating a catheter from a sheath [11].

In our patient, the hyperdense tubular structure that was seen on the initial unenhanced CT thorax within the right brachiocephalic vein and SVC gave the impression of a retained catheter fragment adherent to the vessel wall at first glance (Fig. 1). However, on closer inspection, the hyperdense tubular structure was noted to have an

average density of 600–800HU and appeared to be discontinuous and interrupted, with a visible lumen measuring 2.7 mm in maximum diameter. The overall outer diameter of this tubular structure measured approximately 10 mm, which is much larger than expected in comparison with the diameter of a 16-French permanent tunneled dialysis catheter. No contrast was seen within the lumen, and there was also concomitant stenosis and occlusion involving bilateral brachiocephalic veins near the confluence and the upper SVC. All these features therefore lead to the diagnosis of a retained calcified fibrin sheath in our patient. Retrospective analysis of the chest radiograph taken on admission showed evidence of the sheath at the right paratracheal region (Fig. 4).

It is unknown whether the fibrin sheath in our patient was calcified prior to catheter removal or whether the calcifications developed after being retained, as there was no previous CT done and we were unable to obtain his previous chest radiographs for comparison, due to the last catheter being removed 5 years ago. From previous case reports, calcifications of the sheaths were noted in hemodialysis patients with increased levels of serum calcium, phosphate and parathyroid hormones [3, 13]. Vascular calcifications may also be due to inflammatory cytokines, metabolic or genetic etiology [7]. Krausz et al. [4] observed calcification of a previously non-calcified sheath in one patient, proving that the sheath may become calcified after being retained. In our patient, his recent blood parameters were within normal range; however, we were unable to assess his old records to ascertain whether he had any previous derangement in his calcium and phosphate metabolism.

Central venous stenosis is a common problem among hemodialysis patients. They are more commonly associated with CVCs inserted into the subclavian vein as compared to the internal jugular veins and, however, may also occur without a history of previous catheter placement in up to 40% of patients [14, 15]. In patients with prior CVC insertions, endothelial injury and mechanical irritation by the catheters predispose to luminal narrowing of the central veins causing stenosis and occlusion [5, 16]. Retained fibrin sheaths with associated venous occlusions have been reported in cases on CT as well as on autopsy [1, 4, 5]. Our patient had a history of repeated cannulation to his right internal jugular vein likely causing recurrent vessel trauma with recurrent sheath formation, venous wall thickening and subsequent central venous occlusion. It is likely that this occlusion did not occur acutely, in view of the presence of well-established collateral channels in this patient. However, his acute symptoms of progressive facial and neck swelling which occurred only after revision of his AVF were likely attributed to the sudden increased in blood flow from the AVF,

increasing blood flow through the collateral system. As the obstruction was at the level of the brachiocephalic and preazygos SVC, he had collateral drainage via the right superior intercostal veins and azygos veins.

Management of central venous stenosis may be conservative in asymptomatic cases. For patients who are symptomatic, depending on the degree of stenosis, balloon angioplasty with or without stenting may be performed. In our case, the degree of stenosis was severe and our patient was symptomatic; however, due to the sheath being calcified and adherent to the wall, balloon angioplasty was not a feasible option. He was referred for ligation of his left brachial AVF to relieve the symptoms of neck and facial swelling. At the time of this report, he is on regular hemodialysis via a long-term tunneled femoral catheter and remains well.

Conclusions

Knowledge of the radiological discriminating features of retained calcified fibrin sheaths is important in differentiating them from fractured catheter tips to prevent diagnostic confusion and unnecessary interventions. Retained calcified fibrin sheaths should be considered as one of the long-term complications of hemodialysis CVCs and also as a potential cause of SVC obstruction. They should also be included in the differential diagnosis of radio-opaque intravascular structures or venous calcifications in hemodialysis patients with a history of previous CVC placement.

Abbreviations

AVF	Arteriovenous fistula
BBF	Brachioabasilic fistula
CT	Computed tomography
CVC	Central venous catheter
IJV	Internal jugular vein
IVC	Inferior vena cava
PTFE	Polytetrafluoroethylene
SVC	Superior vena cava

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Author contributions

SYL is the main author in writing and compiling the case report. YA helped in writing and obtaining patient's clinical data. SIM helped in writing and supervision. All authors have read and approved the manuscript.

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Availability of data and materials

Not applicable.

Declarations

Ethics approval and consent to participate

Patient gave informed consent to use their data and images in this case write up.

Consent for publication

Written informed consent was obtained from the patient for publication of this case report and accompanying images.

Competing interests

The authors declare that they have no competing interests.

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