

Case Report

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Pinch-off syndrome from a chemoport catheter successfully managed with endovascular retrieval



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ARTICLE INFO	A B S T R A C T
Keywords: Pinch-off syndrome Oncology Interventional radiologist Case report	Introduction and importance: A totally implantable venous access device (TIVAD), also referred to as 'chemoport', is frequently used for oncology patients. Chemoport insertion via the subclavian vein access may compress the catheter between the first rib and the clavicle, resulting in pinch-off syndrome (POS). The sequela includes catheter transection and subsequent embolization. It is a rare complication with incidence reported to be 1.1–5.0% and can lead to a devastating outcomes. <i>Case presentation</i> : 50-year-old male had his chemoport inserted for adjuvant chemotherapy 3 years ago. During the removal, remaining half of the distal catheter was not found. There was no difficulties during the removal. Chest xray revealed that the fractured catheter had embolized to the right ventricle. Further history taking, he did experienced occasional palpitation and chest discomfort for the past six months. Electrocardiogram and cardiac enzymes were normal. Urgent removal of the fractured catheter via the percutaneous endovascular approach, under fluoroscopic guidance by an experience interventional radiologist was done. The procedure was successful without any complication. Patient made an uneventful recovery. He was discharged the following day, and was well during his 3 rd month follow up. <i>Conclusion</i> : Early detection and preventive measures can be done to prevent pinch-off syndrome. Unrecognized POS can result in fatal complications such as cardiac arrhythmia and septic embolization. Retrieval via the percutaneous endovascular approach provide excellent outcome in the case of embolization.

1. Introduction

A totally implantable venous access device (TIVAD) plays a crucial role for oncology patients in long-term chemotherapy, fluid or medication administration and parental nutrition. It is typically inserted through the subclavian or internal jugular vein. It may potentially be associated with complications such as pneumothorax, bleeding, cardiac arrhythmia, venous thrombosis or bloodstream infection [1]. In terms of a chemoport being inserted via the subclavian route, compression of the catheter in the space anterior to the first rib and posterior to the clavicle can result in pinch-off syndrome (POS). POS can result in transection and embolization of the chemoport. It is a rare complication, with the incidence reported to be around 1.1-5.0% [2]. Clinical presentation of POS is highly variable, covering a wide spectrum of clinical conditions, which include asymptomatic fracture of the catheter, catheter dysfunction, embolization of a catheter fragment leading to palpitation, dyspnea, cough, thoracic pain or septicaemia [3]. Here, we report a case of spontaneous fracture and migration of a chemoport catheter due to POS, which was subsequently successfully retrieved by the percutaneous endovascular approach, in collaboration with an interventional radiologist. Its technique and difficulties during percutaneous endovascular approach was rarely mentioned in other case reports. In this case, technique and difficulties encountered were discussed. We also emphasize early detection and preventive measures for POS. This manuscript has been reported in line with SCARE's 2020 Criteria [4].

2. Case presentation

A 50-year-old male was diagnosed with Stage III descending colon carcinoma in 2017. He had no drug allergy history or family history of

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malignancy. He is a non smoker and non alcoholic drinker. A left hemicolectomy was done, after which he was planned for adjuvant chemotherapy. A chemoport was inserted through his right subclavian vein by a surgeon, without complication. Post operatively, the location of the chemoport was confirmed by a chest radiography (Fig. 1). Scheduled chemotherapy was completed in 2018, and subsequent follow ups showed no evidence of tumour recurrence. The patient was keen for removal of the chemoport after three years. During the removal, it was found that only the port and half of the catheter (10cm) was in situ, but the remaining half of the distal catheter (10cm) was not found (Fig. 2). There was no excessive pull or dense adhesion during the removal. With a high index of suspicion for catheter fracture, a posteroanterior view chest x-ray was done immediately. The chest x-ray revealed that the fractured catheter had embolized to the right ventricle (Fig. 1). After investigating the patient's history, he mentioned that he experienced occasional palpitation and chest discomfort for the past six months, but he did not seek any medical attention. His vital signs were stable. Physical examination was unremarkable. Electrocardiogram and all blood investigations including cardiac enzymes were normal. The patient was referred to an interventional radiologist for removal of the fractured catheter via the percutaneous endovascular approach, under fluoroscopic guidance.

The patient was positioned supine, with continuous monitoring of cardiac and vital signs. A preliminary ultrasound was done by an interventional radiologist and cardiologist in a well equipped tertiary medical center. Senior interventional radiologist with more than 5 years of experiences in particular specialty field performed this procedure. A preliminary fluoroscopy showed the catheter fragment within the right ventricle. Under aseptic technique and sonographic guidance, local anesthesia was given to the right side of the neck. The right internal jugular vein (IJV) was accessed using 16G IV cannula, followed by insertion of a guidewire and 8Fr x 23cm Prelude sheath introducer (Merit Medical Systems, Inc., South Jordan, Utah, USA). En Snare Endovascular Snare system (Merit Medical Systems, Inc., South Jordan, Utah, USA), which consisted of a 6Fr catheter and 120 cm length 0.055" 20mm loop snare, was initially used. However, difficulty was encountered in attempts to loop onto the catheter fragment within the right ventricle, likely due to the stiffness of the catheter and snare system. Subsequently, a modified 5Fr Cobra-1, 65 cm catheter with looped 0.018-inch x 150 cm Terumo guidewire was used. The guidewire advanced beyond the catheter fragment and maneuvered to surround the catheter. The catheter fragment was successfully snared and

removed, together with the sheath (Fig. 3). Immediate manual compression was applied onto the right IJV to secure haemostasis. The length of the migrated piece was 10cm; no thrombus was observed at the tip (Fig. 2).

During the procedure, there were a few episodes of ectopic ventricular beats, particularly during the attempts to snare the catheter fragment within the ventricle. However, the patient remained asymptomatic throughout, and made an uneventful recovery. He was discharged the following day, and was well during his 3rd month follow up. During the follow up, he denied any palpitation, chest discomfort or reduced effort tolerance. Electrocardiogram was normal. He can carry out his daily activities and works as usual.

3. Discussion

POS occurred only in patients with a chemoport inserted via the subclavian vein infraclavicular approach. This is attributable to the small gap between the clavicle and the first rib, thus leading to mechanical compression and shearing forces acting on the catheter. A catheter can be damaged and fractured. An objective grading of POS was proposed by Hinke et al. [5]: grade 0, no narrowing in the catheter's course; grade 1, an abrupt change in the course without luminal narrowing; grade 2, luminal narrowing as the catheter passes under the clavicle (true pinch off sign); and grade 3, complete catheter fracture.

Clinical presentation of POS is highly variable. Patients may present with pain, with or without swelling, at the catheter insertion site, or catheter dysfunction with difficulty in aspirating or flushing the chemoport. A high index of suspicion of POS should be made if there is a positional change of catheter function by raising the ipsilateral arm or rolling the ipsilateral shoulder. Embolization of a catheter fragment might result in cardiac arrhythmia, dyspnea, cough, thoracic pain or act as a source of septicaemia. Surprisingly, some patients may remain asymptomatic, and the embolized fragments are found incidentally by routine chest radiography [3]. The interval from the time of chemoport insertion to pinch off is difficult to ascertain. This is because in the majority of cases, patients remain asymptomatic, and it is discovered incidentally [3].

Suspected POS should be complemented with a chest radiography or fluoroscopic imaging. Chest radiography may reveal a catheter deviation, narrowing or fracture, or the location of embolized catheter fragment. However, a normal imaging does not fully rule out the diagnosis of POS. Injection of contrast into the chemoport is helpful to identify any



Fig. 1. (A) Chemoport was inserted via right subclavian vein access. (B) Chest radiography revealed the embolized catheter fragment at right ventricle (arrow).



Fig. 2. (A) Fractured catheter (B) The length of the migrated catheter was 10cm.



Fig. 3. (A) Modified snare system using Cobra catheter and Terumo guidewire manoeuvred to surround the catheter fragment. (B) Successful ensnaring of the catheter fragment which was removed simultaneously with the catheter and sheath.

leak, as well as the position and condition of the catheter [6].

Our patient was having on and off palpitation and chest discomfort for a six-month duration, but the symptoms quickly disappeared after a short rest. It did not affect his daily activities, hence he did not seek any medical attention. He had not been using his chemoport for the past three years. We only realized the fractured chemoport catheter during the process of removal. A chest radiography showed an embolized catheter fragment in his right ventricle.

For grade 2 or grade 3 POS, immediate removal of the chemoport is required. Minimal invasive technique via the percutaneous endovascular approach to retrieve the embolized catheter fragment can be achieved by an interventional radiologist or cardiologist. It is the gold standard approach. About 93.5% of cases involving an embolized catheter fragment can be retrieved successfully via the percutaneous endovascular approach. Only 2.3% of cases require thoracotomy [3]. For grade 0 or grade 1 POS, routine observation with complementary chest radiography should be done. In the case of our patient, it was grade 3 POS with an embolized catheter fragment in the right ventricle. With the presence of an interventional radiologist, percutaneous endovascular retrieval of the embolized catheter fragment via right the internal jugular vein was performed.

During this procedure, there was initial difficulty encountered in ensnaring the catheter fragment within the right ventricle. This may have been due to the stiffness of the catheter and snare system, causing difficulty in maneuvering the loop onto the catheter fragment. The contact of the relatively stiff snare loop against the ventricular wall also caused brief episodes of arrhythmia in the patient. In order to overcome this difficulty, a modified snare system using a Terumo guidewire and Cobra catheter, which were less stiff, were used. This system was also less 'abrasive' on the ventricular wall, and thus did not cause any further episodes of arrhythmia during the procedure. Easier manipulation of the catheter and loop also enabled successful ensnaring and retrieval of the catheter fragment.

Prevention is better than cure. POS is preventable. Insertion of a chemoport the via internal jugular vein access will definitely help to prevent POS, and has a lower rate of venous thrombosis, catheter malfunction or malposition. Besides, POS can be avoided if chemoport insertion via the subclavian vein access is placed more laterally, since medial positioning is associated with a narrower costoclavicular space [6]. In addition, early recognition and detection of POS might help to avoid devastating complication. Periodic evaluation of the chemoport is important for early detection of POS. Signs of catheter dysfunction such as difficulty in aspirating or flushing the chemoport or presence of other clinical symptoms of POS mandate a chest radiography or fluoroscopic imaging.

4. Conclusion

In conclusion, early recognition of POS is vital to prevent the catheter being fractured and embolized. Unrecognized POS can result in fatal complications such as cardiac arrhythmia and septic embolization. Preventive measures such as chemoport insertion via the internal jugular vein access, routine monitoring using chest radiography, and a high index of suspicion for POS in catheter dysfunction are highly recommended. In the case of an embolized catheter fragment such as ours, retrieval via the percutaneous endovascular approach remained a gold

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standard approach, and provided an excellent outcome.

4.1. Patient's perspective

Patient was thankful and appreciated all the efforts done by doctors throughout his admission. He also felt fortunate in which the fractured catheter didn't cause major morbidity towards him.

Ethical approval

None.

Sources of funding

None.

Authors' contributions

All authors were actively involved in direct patient care and have read and approved the final manuscript. CJS is the principal author. Salina was an interventional radiologist who performing the percutaneous endovascular retrieval of catheter fragment, and was assisted by Wong We Li. Tan JH, Lee Ee Peng and raymond was involved in data collection, proofread and revised the manuscript.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Declaration of competing interest

None.

Acknowledgement

This report did not require approval from an institutional ethical

review board. All authors agreed with the content of the final manuscript. The authors conformed to the provisions of the Declaration of Helsinki in 1995 (as revised in Brazil in 2013).

Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.amsu.2021.102294.

Research registration

None.

Guarantor

Chuah Jun Sen.

Provenance and peer review

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References

- C. Ramachandra, J.P. Kumar, C. Srinivas, Pinch off syndrome a rare complication of chemo port, Int. J. Health Sci. Res. 9 (2) (2019) 298–301.
- [2] S.Y. Ko, S.C. Park, J.K. Hwang, et al., Spontaneous fracture and migration of catheter of a totally implantable venous access port via internal jugular vein - a case report, J. Cardiothorac. Surg. 11 (2016) 50.
- [3] A. Surov, A. Wienke, J.M. Carter, D. Stoevesandt, C. Behrmann, R.P. Spielmann, K. Werdan, M. Buerke, Intravascular embolization of venous catheter–causes, clinical signs, and management: a systematic review, JPEN - J. Parenter. Enter. Nutr. 33 (6) (2009 Nov-Dec) 677–685.
- [4] R.A. Agha, T. Franchi, C. Sohrabi, G. Mathew, for the SCARE Group, The SCARE 2020 guideline: updating consensus surgical CAse REport (SCARE) guidelines, Int. J. Surg. 84 (2020) 226–230.
- [5] D.H. Hinke, D.A. Zandt-Stastny, L.R. Goodman, E.J. Quebbeman, E.A. Krzywda, D. A. Andris, Pinch-off syndrome: a complication of implantable subclavian venous access devices, Radiology 177 (2) (1990 Nov) 353–356.
- [6] H. Li, S. Jen, J.H. Keshavamurthy, G.H. Bowers, H.A. Vo, E. Rotem, Imaging evaluation of catheter integrity prevent potentially fatal complication of pinch-off syndrome: illustration of two cases, Quant. Imag. Med. Surg. 7 (3) (2017 Jun) 369–372.