CASE REPORT

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# Complication of tunneled dialysis catheter of chronic ulcer in the site of vascular access entry leading to suspected squamous cell carcinoma (SCC): a case report

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## ABSTRACT

**Introduction:** Catheter-related infection (CRI) is one of the most common vascular access complications in dialysis catheter-related procedures. However, repeated chronic infected ulcer that leads to suspected malignancy in a tunneled dialysis catheter (TDC) insertion site and its risk factor have not been reported.

**Case description:** A 37 years-old woman presented with a chronic exudative ulcer in the right breast at the entry site of a tunneled dialysis catheter. The past medical history included type 2 diabetes mellitus, chronic kidney disease, hepatitis C, and obesity. The patient had routine hemodialysis with TDC for the last 2 years and had changed the TDC access 3 times due to recurrent CRI. At first, the lesion appeared as a small pustule and enlarged recurrently. Later, the lesion progressed into an 82x80 mm raised crateriform exudative ulcer with a raw surface similar to Marjolin's ulcer squamous cell carcinoma. The patient was referred to oncology due to limited facilities in our hospital.

**Conclusion:** CRI is one of the most common long-term complications of TDC. In out of the case, this led to chronic infection and change in TDC, chronic inflammation then became Marjolin's Ulcer. Risk factors for suspected SCC in our patient were immunosuppressive state from diabetes, hepatitis C, CKD, and chronic inflammation from a repeated chronic infected ulcer. SCC can be considered a long-term complication of TDC. Early prevention of CRI and risk factor controlling should be considered to prevent such complications.

**Keywords:** tunneled cuffed catheter, catheter-related infection, chronic ulcer, squamous cell carcinoma.
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## INTRODUCTION

Tunneled dialysis catheter (TDC) is a modern vascular access procedure with many advantages. However, there are many complications if it is used in the long term.[1] Tunneled dialysis catheters (TDCs) are said to act as a bridge to permanent lines for hemodialysis, including arteriovenous fistula. Catheter-related infections (CRIs) are one of the most common vascular access complications of dialysis catheter-related surgery.[2–4] Patients with catheterized hemodialysis have a 2-3 times higher risk of hospitalization for infection and death than patients with arteriovenous fistulas or grafts.[5]

Delayed treatment CRI can lead to malignant lesions. Malignant lesions can result from chronic ulcers, burn scars, wounds, sinuses, and fistulas of various origins. Malignant lesions associated with chronic ulcers usually result from malignant transformation to well-differentiated squamous cell carcinoma (SCC).[6] On rare occasions, a repeated chronic ulcer had a morphology similar to Marjolin's ulcer (MU).[7]

Malignant tumors are very difficult and difficult to manage. Early diagnosis requires a high level of awareness from researchers, who rely primarily on experience and many modalities. Aggressive treatment strategies with an interdisciplinary approach that includes radiation therapy and surgical management are essential to prevent morbidity and mortality.

This case report presents a repeated chronic infected ulcer that leads to suspected malignancy in a TDC insertion site and its risk factor.

## CASE DESCRIPTION

A 37 years-old woman was referred from dermatology and internist outpatient clinic to surgery clinic with a chronic exudative ulcer in the right breast at the entry site of a tunneled dialysis catheter for almost two years and enlarged rapidly in just one month. At first, the lesion appeared as a small pustule that enlarged and ruptured into a foul-smelling purulent ulcer.

Past medical history included a 3-year history of type 2 diabetes mellitus (DM), with macrovascular complications such as nephropathy with chronic kidney disease. The patient had routine hemodialysis with TDC for the last 2 years and had changed the TDC access 3 times due to recurrent CRI. Although informed about the risks, the patient refused timely arteriovenous fistula placement for hemodialysis.

The initial hemodialysis was through a catheter double-lumen (CDL) inserted in the right internal jugular vein two years ago. At that time the patient had profuse vomiting, septic condition, hyperglycemia, and hyperuricemia due to CKD. For a summary regarding the progression of the patient's disease, see **Table 1**.

The CDL and TDC procedure was carried out at the referral hospital due to the absence of vascular surgeon in our hospital. But the patient had routine hemodialysis and check up to the internist of our hospital. Other medical problems included hypertension, obesity, and hepatitis C.

The general physical examination presented normal blood pressure of 127/81 mmHg, sinus tachycardia at 124 bpm, a body temperature of 36.8oC, and body weight at 86 Kg (height: 160 cm, BMI: 33,6 Kg/m2). The local physical examination showed the lesion progressed into an 82x80 mm raised crateriform exudative ulcer with a raw base on the right breast (see, **Figure 2**). Axillary lymph node examination was performed, and there was no lymph node enlargement.

Laboratory-test results revealed normocytic normochromic anemia of 8.0 g/dL (reference range (RR) 11.7 - 15.5 g/dL), white cell count 14.290 /uL (RR 3.800 - 10.600 /uL) with neutrophil predominance (76.0 %; RR 50-70 %), lymphocytopenia 14% (RR 25-40%), ureum 68 mg/dL (RR 13 - 43 mg/dL), creatinin 2,74 mg/dL (RR 0.51 - 0.95 mg/dL), random blood glucose 196 mg/dL (RR 70-160 mg/dL) and reactive anti HCV with ELISA value 26.88 (RR >= 1.00 is positive). Other laboratory parameters were unremarkable.

Marjolin's ulcer squamous cell carcinoma on the right breast was suspected, with differential diagnoses including; other Marjolin's ulcer malignancies (basal cell carcinoma and melanoma malign), invasive carcinoma mammae, diabetic ulcer, keratoacanthoma, and necrotic abscesses.

Previously in the dermatology clinic, the patient had been treated with NaCl 0,9% compress combined with gentamicin ointment every 6 hours for 2 weeks. According to the patient, the wound did not show any healing progression. Therefore, the patient was referred to oncology for further examinations and treatments such as specimen culture, pathologic anatomy examination from biopsy, or other modalities. This patient had been referred to an oncology surgeon for further examination such as biopsy or other modalities. However, the patient could not undergo a biopsy at the moment because of her chronic kidney disease and needs further multidisciplinary evaluation before the biopsy could be done.

**Table 1. Timeline of vascular access procedure history and lesion progression of the patient**

| Time | Procedure | Description |
|---|---|---|
| July, 2019 | Initial CDL | Initial access in the right internal jugular vein. |
| August, 2019 | Initial TDC | Replaced the access from CDL to TDC. TDC was inserted in the right subclavian vein with the skin entry at approximately 5 cm superior to the outer margin of the areola mamma dextra. |
| October, 2019 | 1st replacement | The first infection occurred at the site of the skin entry. There was a pustular lesion which turned into an ulcer. TDC replacement was performed in the healthy skin for the skin entry. The wound was treated with Gentamicin ointment for two weeks and healed partially. |
| July, 2020 | 2nd replacement | Another infection occurred at the same site and had a purulent discharge in TDC. Aside from TDC replacement, the wound was treated with Gentamicin ointment and did not heal. |
| December, 2020 | 3rd replacement | The lesion progressed to a raised crateriform exudative ulcer with a raw surface similar to Marjolin's ulcer squamous cell carcinoma. The vascular surgeon considered replacing the skin entry to the upper right clavicle. |

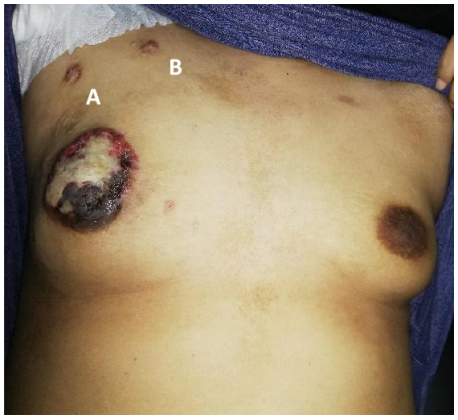


**Figure 1.** Lesion at the right breast. (A) cicatrix wound from skin site of entry in 2nd replacement of TDC; (B) cicatrix wound from venous entry incision

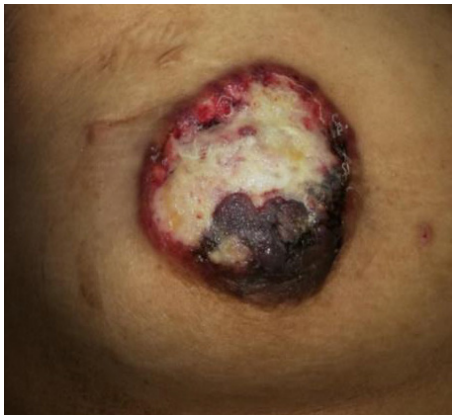

**Figure 2.** Raised crateriform exudative ulcer with a raw base similar to Marjolin's ulcer squamous cell carcinoma, there was no discharge from areola mamma.

## DISCUSSION

An untreated chronic ulcer was known for being a predecessor of malignancy in skin tissue.[6] But the malignancy is rarely reported as one of the long-term TDC complications. The incidence of malignant transformations is low, delaying the diagnosis of these patients and increasing morbidity and mortality.[7] Therefore, the importance of this case report is to show that SCC or other skin malignancies in entry site of TDC can be considered as one of the long-term TDC complications.

TDCs often serve as a bridge to permanent access to hemodialysis, including arteriovenous fistula. According to Castro *et al*, the most common and main reason for TDC's prolongation was access failure, followed by access maturity. Another important factor

was the difficulty of planning access creation. This may be due to delayed referrals, rapid deterioration of renal function, patient acceptance, surgical planning, patient education, system problems, comorbidities of patients, and simultaneous hospitalization for other problems.[1] In this case, the patient refused timely arteriovenous fistula placement for hemodialysis, despite being infected at the entry site. Her reason was she thought she just had temporary hemodialysis. She believed that she will not need hemodialysis in the future despite her comorbidity and progressive renal disease. This belief is tough to break and needs more patience and a subtle approach for the patient to change her mind.

The first CRI episode on this patient occurred 8 months after the initial TDC procedure. There are several factors to CRI risk factors, including skin staphylococcal colonization, catheter hub colonization, long-term use, diabetes, and patient-identified immune capacity.[4] Malignant tumors are known to tend to develop in areas of constant inflammation, such as chronic ulcers, where blood flow is inadequate. Virchow's hypothesis that chronic irritation is a factor in carcinoma development reinforces this notion. Another suggested theory that supports this concept is an increase in spontaneous mutations from prolonged inflammation and repeated attempts at healing.[7,8] The immunosuppressive condition due to type 2 DM, obesity, and hepatitis C in this patient interrupted the remodeling phase of the skin tissue.[8,9]

Malignant transformation occurs more frequently at the wound margins, where it is the point of rapid cellular turnover and is the site of malignant transformation in chronic ulcers.[7,8] As in **Figure 2**, the lesion showed increased crateriform at the wound margin and had MU features on the skin such as ulcerative and foul-smelling with rapid growth and rolled elevated margins which lead to suspected SCC.

A biopsy is a standard diagnostic test for analyzing the histological characteristics of MU. The biopsy can be in the form of an excision, incision or punch biopsy. To minimize false-negative results, samples should be collected from multiple sites, including the wound margin. However, the most important aspect of MU diagnosis is medical history. Studies have shown that monitoring wound changes and performing a biopsy can prevent MU. Some authors recommend an annual biopsy for chronic ulcers. Other studies recommend considering a biopsy of a wound that does not heal within 1-3 months.[7] Delayed biopsy in this patient was caused by the physician's unawareness of malignancy as one of the long-term TDC complications. The patient's immunosuppressive state is also a key point to the biopsy delay since metastasis might occur when the biopsy is performed without holistic assessment and multidisciplinary considerations.

There is one case report with SCC arising at the exit site of a TDC had been reported. It also presented the immunosuppressive condition, which is associated with organ transplantation. Although, the progression of SCC in the case was reportedly acute in a span of 3 months. Co-infection with human papillomavirus plays a role in the cause. Therefore, SCC can take an aggressive course for immunocompromised patients.[10] Managing the immunosuppressive condition such as controlling the blood glucose and losing of excessive weight from the patient may prevent the untreated wound ulcer that leads to malignancy such as MU SCC.

## CONCLUSION

SCC at entry site scar (albeit an unlikely event) may be added to the risks of TDC in patients on long-term immunosuppression. Early prevention of CRI, risk factor controlling, and early diagnoses for malignancy should be considered to prevent such complications. Further study needed to assess the correlation between TDC insertion and malignancy.

## DISCLOSURES

### Consent for Publication

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

### Funding

Provide statement regarding funding sources, grant or third-party support.

### Conflict of Interest

Provide statement regarding possible relationships between author(s) and any organizations or person that could influence the objectivity during the study, interpreting the result as well as during the writing of the manuscript.

### Author Contribution

Provide detailed contribution of each author for example. AA and BB involved in concepting, designing and supervising the manuscript. CC and DD conduct the study. AA and CC analyze the data. All authors prepare the manuscript and agree for this final version of manuscript to be submitted to this journal.

## REFERENCES

1. Castro V, Farber A, Zhang Y, Dicken Q, Mendez L, Levin SR, et al. Reasons for long-term tunneled dialysis catheter use and associated morbidity. J Vasc Surg. 2021;73(2):588–92. Available from: https://doi.org/10.1016/j.jvs.2020.06.121
2. Miller LM, Clark E, Dipchand C, Hiremath S, Kappel J, Kiaii M, et al. Hemodialysis tunneled catheter-related infections. Can J Kidney Heal Dis. 2016;3(1).
3. Poinen K, Quinn RR, Clarke A, Ravani P, Hiremath S, Miller LM, et al. Complications From Tunneled Hemodialysis Catheters: A Canadian Observational Cohort Study. Am J Kidney Dis. 2019;73(4):467–75. Available from: https://doi.org/10.1053/j.ajkd.2018.10.014
4. Beathard GA, Urbanes A. Infection associated with tunneled hemodialysis catheters. Semin Dial. 2008;21(6):528–38.
5. Dhingra RK, Young EW, Hulbert-Shearon TE, Leavey SF, Port FK. Type of vascular access and mortality in U.S. hemodialysis patients. Kidney Int. 2001;60(4):1443–51.
6. LM C, M C. Squamous cell carcinoma developed on neglected, mistreated and delayed diagnosed chronic venous leg ulcer. Arch Surg Clin Res. 2019;3(1):016–21.
7. Hobbs M, Campbell E, Braun KA, Wong L. Multiple synchronous acute Marjolin ulcer: A report of 2 cases and literature review. JAAD Case Reports. 2019;5(6):511–3. Available from: https://doi.org/10.1016/j.jdcr.2019.04.002
8. Som Nath N, Gilmore BF, McCann RK, Mosca PJ. Management of a cutaneous squamous cell carcinoma overlying an AV fistula. BMJ Case Rep. 2017;2017:2016–8.
9. Gonzalez ACDO, Andrade ZDA, Costa TF, Medrado ARAP. Wound healing - A literature review. An Bras Dermatol. 2016;91(5):614–20.
10. Hammes JS, Bestoso JT, Sharma A. Squamous cell carcinoma in situ arising at the exit site of a tunneled catheter. Am J Kidney Dis. 2004;44(3):43–6.