



## Mucin Secreting Poorly Differentiated Rectal Adenocarcinoma with Inguinal Metastasis: Is it Regional or Distant Metastasis? A Case Report and Review of Literature

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### Abstract

This report presents a case of solitary inguinal metastasis after treatment of rectal cancer by abdominoperineal resection. A 79-year-old Omani man was diagnosed with poorly differentiated mucinous adenocarcinoma of the rectum in March 2015. Metastatic involvement of inguinal lymph nodes (ILN) from rectal adenocarcinoma is unusual presentation, particularly without signs of distant spread to other organ sites. In the current time, ILN involvement, including solitary involvement, is classified as metastatic disease (M). Inguinal mass was totally excised; neoadjuvant radiation therapy and chemotherapy had been also carried out before. Pathological analysis showed poorly differentiated mucinous adenocarcinoma compatible with his rectal cancer. The interesting finding was that this case did not seem to belong to any traditional rectal cancer metastasis pathway. This case is representative and worthy of further study to explore whether there is another rectal cancer metastasis pathway.

**Keywords:** Mucin; Adenocarcinoma; Inguinal Lymph Nodes; Metastasis

### Background

Poorly differentiated mucinous adenocarcinoma of the rectum is considered as a distinctive subtype of rectal tumor and characterized by the presence of abundant mucin which compromise of approximately 50% of its volume [1]. This tumor is characterized by a lower incidence and poor prognosis compared with other rectal tumors according to previous studies results. In addition, this kind of tumor is characterized by features of aggressive behavior, high rate of metastasis and diagnosing at an advance stage [2,3]. There have been few studies on mucinous adenocarcinoma because there are only few cases diagnosed and reported [4].

Inguinal LNs metastasis from rectal carcinoma remains an unclear classification whether it is regional or distant metastasis. Some studies have reported that inguinal LNs metastasis in rectal carcinoma are considered as a non-regional metastasis in TNM classification [5]. However, adenocarcinoma of the lower rectum can metastasize to inguinal LNs in the same manner of the anal canal cancer in some occasions [6,7]. Furthermore, metastasis may occur as a result of locally advanced primary tumors, and that in these cases, non-surgical option is often preferred because the frequency of distant metastasis and poor prognosis [8,9]. In controversy, some studies found that LNs excision showed a good

prognosis; hence surgical treatment would be a reasonable option [10,11].

According to the 8<sup>th</sup> edition of the TNM classification, Inguinal LNs metastasis regards as a regional in anal carcinoma and distant in rectal carcinoma [5]. Although there are no adequate evidences to resolve this difference, survival of patients and other site distant metastasis is believed to be an appropriate evidence for determining regional versus distant metastasis. In this respect, the aim of this case report is to identify the long-term outcomes of the inguinal LNs metastasis from the rectal mucinous adenocarcinoma whether distant or regional. In this report, a 79 year-old patient with a mucin secreting adenocarcinoma of the rectum that had previously undergone a robotic abdominoperineal resection with end colostomy abroad without evidence of distant metastasis, but then recurred with left inguinal mass 4 years later. We will also perform a review of the literature and recent studies of inguinal LNs metastasis and the type of the metastasis in mucinous adenocarcinoma of rectal cancer.

### Case Presentation

A 79 year-old Omani male presented with a 10 months history of approximately 7.7 x 5.9 x 5.6 cm solitary lobulated soft tissue mass lesion in the left inguinal region, on the background of abdominoperineal resection and end colostomy of the low rectal tumor done in 2015 in India. The mass was non-tender, hard and irreducible with normal overlying skin. A biopsy of the mass revealed that the findings of the left inguinal lymph nodes were consistent with metastasis from a previously diagnosed mucin secreting adenocarcinoma; however, no lymph nodes were seen in these sections. With regards to the history of his condition, the patient presented with anal mass 8 years ago for which colonoscopy and excisional biopsy was done in 2015. Histopathology analysis of the rectal specimen reported a mucin secreting poorly differentiated adenocarcinoma of the rectum. As a result of that he underwent a robotic abdominoperineal resection with end colostomy in 2015. The final histological examination showed 20 lymph nodes taken were negative for malignancy. Preoperative CT-TAP showed no obvious local or distant metastasis as well as local invasion. He also underwent MRI perineum which reported no obvious anal invasion. Postoperatively, he underwent computed tomography scans of the chest, abdomen and pelvis (CT-TAP) regularly. The CT-TAP on March 2015 reported that there was no apparent liver and lung metastasis or obvious local invasion. Follow up PET CT scan in 2016 and 2017 reported that the overall scan findings were of stable disease with no evidence of local or distant metastasis. CEA was not elevated. Patient was recommended for adjuvant chemo radiotherapy and

agreed for oral capecitabine for 6 months which completed eighth cycles on November 2015.

In September 2018, colonoscopy through end-colostomy was done with no significant finding noted. Seven months later, colonoscopy was repeated with normal colon reported. The surveillance PET scan on October 2019 revealed a heterogeneous lobulated soft tissue mass in the left inguinal region suspecting metastasis for which histological evaluation was recommended. The patient underwent a left inguinal mass biopsy in October 2019 (Figure 1 and 2). The histopathological results showed features consistent with metastasis from the previously diagnosed mucin secreting adenocarcinoma for which the MDT decided to give him radiotherapy and reassessed then. The patient received radiotherapy to the pelvis and the left inguinal lymph node to a dose of 45 Gy from December 2019 to January 2020 concurrently with capecitabine.

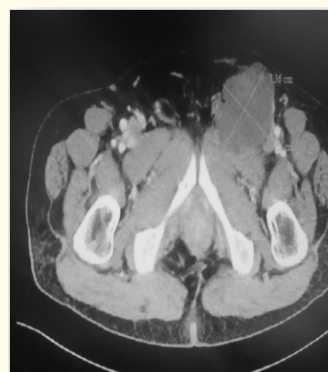


Figure 1: Computed tomography image of the left inguinal lymphadenopathy.

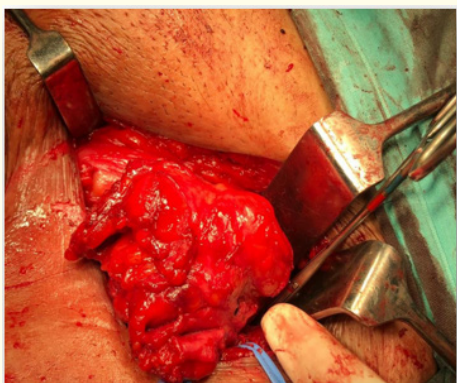


Figure 2: Computed tomography image of the left inguinal lymphadenopathy.

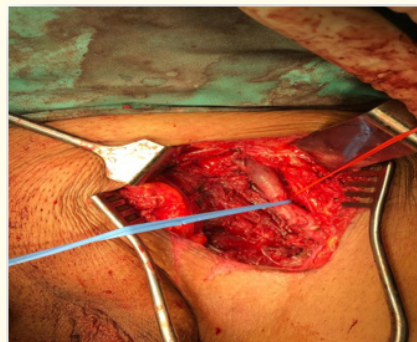
In July 2020, the patient was scheduled for left inguinal mass evaluation which reduced 15% in size post chemo radiotherapy. Excisional biopsy for the large left inguinal mass was decided as the patient was hemodynamically stable and apart from the mass there was no significant history or examination finding.

The patient underwent a left inguinal lymph nodes en-block dissection in July 2020. He was positioned supine and general anesthesia was given. The femoral triangle boundaries were identified and a vertical incision was made over the midpoint.

The great saphenous vein was adherent to the mass and thrombosed; hence, the vein was ligated. The mass had invaded the underlying muscle; therefore, it was dissected off the floor and all the feeding arteries were identified and ligated. The mass had extended below the inguinal ligament; therefore, the ligament was divided to enucleate the whole mass. One drain was placed after the whole mass excised and sent for histopathology. The patient recovered well postoperatively (Figure 3-5). One week later, histopathology examination was released reporting a nodular soft tissue mass (measures 8 x 6 x 5 cm) and weighs 14.1g, irregular surface, cystic appearance and filled with mucinous materials. The section microscopically revealed fibroconnective and skeletal muscle fibers with metastasis from a carcinoma comprising large pools of extracellular mucin. Tumor cells were positive for CK7, CK9 and CDX2 immunohistochemically. Of note, no lymph node tissue was seen in the section examined. The report concluded that the diagnosis is consistent with a metastasis from the previously diagnosed mucin secreting adenocarcinoma.



**Figure 3:** Solitary lobulated mass of the left inguinal region dissection.



**Figure 4:** Femoral vein identified and securely separated.



**Figure 5:** Completely excised left inguinal mass (8 x 6 x 5 cm).

### Discussion and Conclusion

Metastatic involvement of inguinal lymph nodes (ILN) from rectal adenocarcinoma is a rare presentation of rectal cancer, especially without signs of distant spread to the liver, lung or other organ sites [11]. Hence, it represents a unique group of patients who should be managed differently and may have a good prognosis. Currently, ILN involvement, including solitary involvement, is classified as metastatic disease (M). However, anecdotal reports suggest that such patients are a distinct entity and should be managed differently. We present a case of solitary left inguinal metastasis in a patient with a post abdominoperineal resection of rectal mucinous adenocarcinoma.

The most interesting aspect of the present case is the atypical lymphatic route the rectal carcinoma followed to metastasize to the inguinal lymph nodes. Usually, there are three pathways of metastasis of rectal cancer including blood, lymphatic, direct spread

and planting. For lymphatic pathway, metastasis Tumors in the rectum can spread by two different routes. The lymphatic drainage of tumors of the upper rectum reaches the inferior mesenteric artery lymph nodes via the superior rectal arteries. Metastases of tumors originating from the lower rectum reach the internal iliac nodes by following the pathway of the middle and the inferior rectal arteries and then the common iliac and the para-aortic nodes. Tumors of the anal region spread to the superficial inguinal lymph nodes ascending along the femoral vessels to the deep inguinal nodes and along the iliac vessels to the para-aortic nodes [12,13].

In the present case, the most likely hypothesis is that the tumor metastasized to the deep inguinal lymph nodes through a lymphatic pathway along the left inferior epigastric artery being its origin encompassed by the tumor [14]. Furthermore, after abdominoperineal resection for carcinoma of the rectum with interruption of the normal pathway of lymphatic drainage, recurrent disease may find an alternative retrograde route to the superficial and deep inguinal nodes [12,15]. A similar lymphatic pathway could be possible there was another case reported by Hakeem, *et al.* found that a similar lymphatic pathway might possibly occur as in a case of right inguinal lymph nodes metastasis from an adenocarcinoma of the cecum with infiltration into the adjoining peritoneal fat [16].

To conclude, the presence of inguinal metastasis in a patient with rectal carcinoma can be considered regional rather than distant metastasis due to several reason; firstly, the good prognosis of patient who underwent LN inguinal dissection with curative outcome. Secondly, lack of evidence of distant metastasis or local spread. Thirdly, solitary LN involvement rather that group. However, further studies are required to clarify the anatomical explanation of this unusual pathway of metastasis.

### Consent

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

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