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Case report

Ischiorectal fossa metastasis from colon cancer: Case report of a rare entity and review of literature☆

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ABSTRACT

Introduction and importance: Colorectal cancer is one of the most common cancers both nationally and internationally. It commonly metastases to local lymph nodes, liver and lungs, with few reported cases of rare sites of metastasis such as adrenal glands, breast and skin.

Case presentation: We report a 55-year-old-female admitted as case of large bowel obstruction and unintentional weight loss. Computed tomography scan of chest, abdomen and pelvis (CT CAP) showed sigmoid colon circumferential thickening with three lesions in the right hemi-liver. A laparoscopic diverting ileostomy followed by a colonoscopy showed a sigmoidal mass consistent with adenocarcinoma on histopathology. Hence, she received neoadjuvant chemotherapy followed by hepatectomy for the liver metastasis. Post-operatively CT CAP showed a newly developed right ischiorectal fossa (IRF) nodule along with newly developed porta hepatis lymph node. PET scan showed uptake in these two new lesions. Therefore, the patient underwent resection of the primary tumor, porta hepatis lymph node and right ischiorectal fossa nodule excision. The histopathology of the primary tumor came as moderately differentiated adenocarcinoma with both ischiorectal lesion and the porta hepatis nodule being positive for metastatic disease.

Clinical discussion & conclusion: Ischiorectal fossa tumors are extremely rare with the majority being benign in origin. Nevertheless, the possibility of metastasis is there with no clear explanation regarding the pathway of how the metastatic cells can reach the IRF. Pre-operative diagnosis is important to determine the appropriate approach particularly if the mass is thought to be malignant. Further larger studies are needed to understand the pathway of metastasis to IRF.

1. Introduction

Colorectal cancer is the most common cancer among Saudi males and the third most common among Saudi females [1]. It commonly metastases to local lymph nodes, liver and lungs, with few reported cases in the literature of rare sites of metastasis to such as adrenal glands, breast and skin [2,3]. However, to the best of our knowledge there have been no reported cases in the English literature regarding colon cancer metastasizing to ischiorectal fossa. This is the first case of ischiorectal fossa metastasis from colon cancer. This case has been reported in line with the SCARE criteria [4].

2. Case report

We report a case of 55 years old medically free Saudi female, who presented to another facility complaining of left-sided colicky non-radiating abdominal pain, associated obstipation for one day duration. Also, she noticed unintentional weight loss and altered bowel habit over 6 months. She was not on any medications and she had no family history of any malignancy. Upon presentation computed tomography scan of the abdomen showed circumferential thickening involving the distal descending colon spanning for 4 cm distally with the mural thickness up to 1.3 cm associated with pericolic speculation and multiple lymph nodes. The liver demonstrated three lesions in the right liver lobe; the first in segment VI measuring 4 cm, the second in segment VIII measuring 2 cm and the third in segment IVB measuring 3 cm all of which were consistent with metastatic disease with no intra or extrahepatic biliary duct dilatation. In the referring hospital, a laparoscopic diverting ileostomy was performed. Then, she was referred to our institute for definitive treatment.

☆ The manuscript has been read and approved by all the authors. We confirm that this work is original and has not been published elsewhere nor is it currently under consideration for publication elsewhere.

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Fig. 1. PET scan of abdomen showing FDG avid in the porta hepatis lymph node.

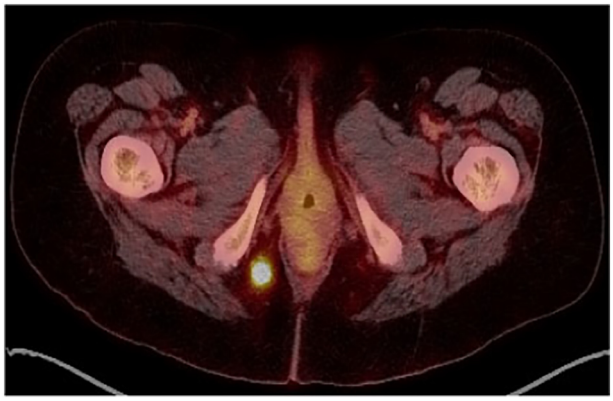

Fig. 2. PET scan of pelvis showing FDG avid in the right ischiorectal fossa nodule.

In our institute, a complete colonoscopy revealed a sigmoidal mass, that was biopsied and was consistent with adenocarcinoma. Her carcinoembryonic antigen (CEA) level was 1061 μg/L. The case was discussed in multidisciplinary tumor board and the decision was to start with neoadjuvant chemotherapy of 6 cycles of FOLFOX followed by segmental liver resection followed by primary tumor resection. Following liver resection, Computed tomography scan of chest, abdomen and pelvis (CT CAP) with IV contrast showed a newly developed well-defined centrally necrotizing nodule at the right Ischiorectal fossa measuring 2.5 cm in diameter along with a newly developed porta hepatis lymph node with stable primary tumor. An 18 fluorodeoxyglucose (FDG) PET/CT scan showed uptake in these two new lesions (Figs. 1–2).

In light of her new findings, the patient underwent resection of the primary tumor with ileocolic anastomosis as well as a perianal excision of the ischiorectal fossa nodule by colorectal surgery consultant, along with porta hepatis lymph node excision by hepatobiliary surgery consultant in July 2020 (Fig. 3). The ischiorectal fossa nodule showed necrosis within the nodule (Fig. 4A–B) and was positive for both CDX2 and CK20 immunohistochemical staining (Fig. 4C–D) while being negative for both CK7 and PAX8 immunohistochemical staining (Fig. 4E–F) consistent with metastatic adenocarcinoma. The final histopathology of the colon specimen came as moderately to poorly differentiated colonic adenocarcinoma with the ischiorectal nodule and the porta hepatis both being positive for metastatic disease. The patient was referred to medical oncology for further management and was found to have metastatic disease to both brain and lungs and passed away after 6 months from her surgery.

3. Discussion

Colorectal cancer (CRC) commonly metastasizes to local draining lymph nodes, liver, lungs, peritoneum, ovaries, central nervous system, bone, kidneys. In addition, it can metastasize to rare sites as hilar lymph nodes, mediastinal lymph nodes, axial lymph nodes, breast, adrenal glands, skin and muscles [2,3,5].

However, a review of the literature using the key-words "ischiorectal fossa tumors", "ischiorectal fossa metastasis" and "colon cancer" in the following Databases 1) PubMed 2) PubMed Central 3) Web of Science including all English literature till March 2021, showed that there have been no reported cases of colon cancer metastasis to the ischiorectal fossa in the English literature prior to our case.

The ischiorectal fossa (IRF) constitutes a large part of the anorectal space. It is pyramidal in shape, bounded by levator ani and the external anal sphincter muscles muscle medially, the obturator internus muscle and obturator fascia laterally, the superficial and deep transverse perineal muscles anteriorly, the lower border of gluteus maximus muscle and the sacrotuberous ligament posteriorly, the levator ani muscles superiorly, and skin of the perineum inferiorly [6]. Tumors in IRF are generally rare and they are more commonly to be benign rather than malignant [6,7]. Primary tumors can originate from different structures

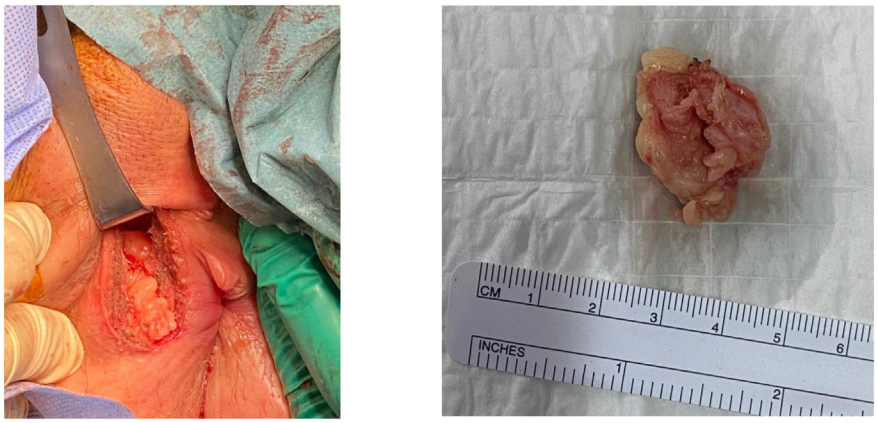

Fig. 3. Intra-operative pictures of: (A) the right ischiorectal fossa after nodule excision (B) right ischiorectal fossa nodule.

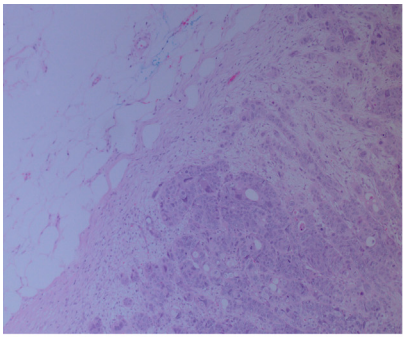

Figure.4-A Microscopic picture of the nodule using H & E staining

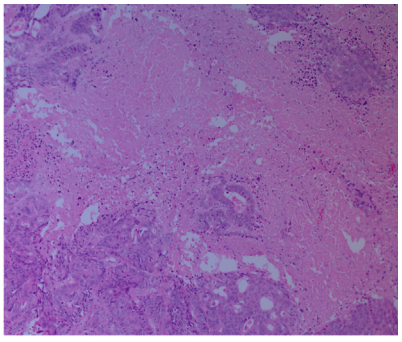

Figure.4-B Microscopic picture of the nodule using H & E staining showing tumor necrosis

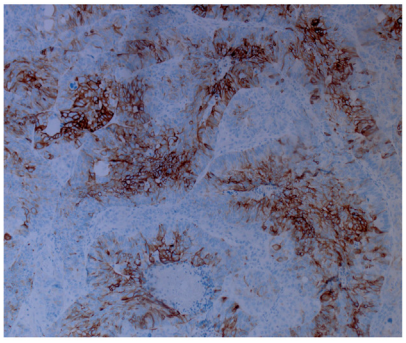

Figure.4-C Microscopic picture of the nodule showing positivity for CK20 immunohistochemical staining

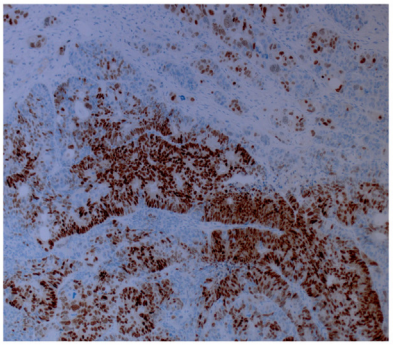

Figure.4-D Microscopic picture of the nodule showing positivity for CDX2 immunohistochemical staining

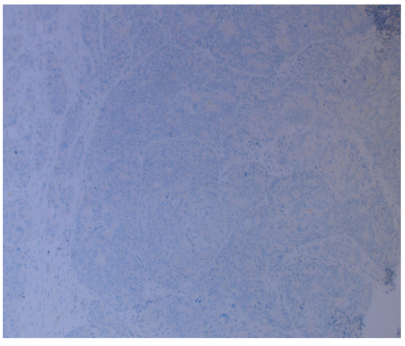

Figure.4-E Microscopic picture of the nodule showing negativity for PAX8 immunohistochemical staining

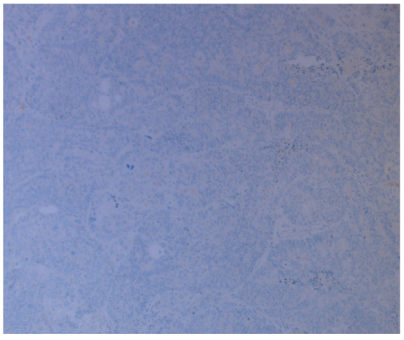

Figure.4-F Microscopic picture of the nodule showing negativity for CK7 immunohistochemical staining

Fig. 4. Microscopic pictures of the right Ischiorectal fossa nodule with different staining confirming it's the colonic origin. A. Microscopic picture of the nodule using H & E staining. B. Microscopic picture of the nodule using H & E staining showing tumor necrosis. C. Microscopic picture of the nodule showing positivity for CK20 immunohistochemical staining. D. Microscopic picture of the nodule showing positivity for CDX2 immunohistochemical staining. E. Microscopic picture of the nodule showing negativity for PAX8 immunohistochemical staining. F. Microscopic picture of the nodule showing negativity for CK7 immunohistochemical staining.

within the fossa like vessels, nerves, fat, skin, and muscles or as a direct extension from other primary tumors originating from the adjacent pelvic organs [6,8]. However, a distant metastatic spread from primary lesions to the ischiorectal fossa is extremely rare and has been reported only few times in the literature from primary lung cancer, ultra-low rectal cancer, prostate cancer, melanoma, gastro-intestinal stromal tumor and chordoma [3,6,9].

Colon cancer usually metastasizes through the lymphatics channels which parallel the arterial distribution, and almost all lymphatic drainage from colorectal cancer eventually ends in the thoracic duct via

cisterna chyli [5]. However, this mechanism of metastasis does not explain the pathway of metastasis in our case. Xeufeng Guo et al. conducted a study with the aim to investigate the lymph node metastasis of mesorectal and ischiorectal fossa in ultra-low rectal cancer. Twenty-three specimens were examined where 415 lymph nodes were detected, of which only two cases with ischiorectal fossa lymph node metastasis and 1 case with micro-metastasis were found [9].

Uzun et al. conducted a retrospective study reviewing patients who were found to have enlarged gluteal or Ischiorectal lymph nodes during the period between 2011 and 2015. Twenty patients were included, of these, there was only 1 case of cancer-related lymphadenopathy (prostate cancer) while the remaining were secondary to inflammatory/infectious causes [10]. Prostatic cancer usually spreads to pelvic lymph nodes in the obturator fossa, all along external and internal iliac vessels, and to the sacrum interior space, despite this known lymphatic spread pattern of prostate cancer, Fang et al. reported another case of a biopsy-proven metastatic prostate adenocarcinoma to IRF [11].

With regards to imaging modalities, magnetic resonance imaging (MRI) is the best tool to detect and assess IRF masses, even though it might show nonspecific features of the primary lesion like; heterogeneity with moderate-high signal intensity on T2-weighted images and moderate to marked enhancement [13] . In addition, trans-anal ultrasonography is another imaging modality that can be of use particularly intra-operatively to localize lesions that are smaller than 5 cm [13]. If the diagnosis was not certain by imaging alone or if there was a potential role for neoadjuvant therapy, a biopsy for confirmation of diagnosis would be of importance [7,13] . Taking into consideration, even if the result of the needle or incisional biopsy were negative, the possibility of malignancy can't be ruled out [15].

In the case of malignant IRF masses, complete surgical excision is crucial to provide the best possible prognosis yet there are no clear guidelines for it [13]. Furthermore, accessing the IRF can be challenging, as small tumors might not be palpable and depending on imaging alone can be misleading as the location of the mass might change relatively to the patient position on the operating table. On the other hand, large tumors might require a combined anterio-posterior approach with resection of any involved pelvic organ or even partial removal of the sacrum [15].

Lithotomy position would be helpful in identifying completely infralevator tumors, as the effect of the pelvic floor would push the mass toward the skin [6]. In our case, the tumor was palpable per rectally, so she was placed in a lithotomy position which also facilitated the excision without the need of changing the patient position after the resection of the primary tumor.

4. Conclusion

Ischiorectal fossa tumors are extremely rare with the vast majority being benign in origin. Nevertheless, the possibility of metastasis is there with no clear explanation regarding the pathway of how the metastatic cells can reach the IRF. Pre-operative diagnosis is important to determine the appropriate approach particularly if the mass is thought to be malignant. Further larger studies are needed to understand the pathway of metastasis to IRF from colon cancer.

Abbreviations

CT CAP	computed tomography scan of chest, abdomen and pelvis (CT CAP)
IRF	ischiorectal fossa
PET	positron emission tomography
CEA	carcinoembryonic antigen
FDG	fluorodeoxyglucose
CRC	colorectal cancer
MRI	magnetic resonance imaging

Availability of data and materials

Data sharing is not applicable to this article as no datasets were generated or analysed during the current study.

Ethics approval and consent to participate

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

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Guarantor

Abdullah Saleh AlQattan and Mohammed Tahtouh.

Research registration number

This is the first reported case of metastasis of colon cancer to the ischiorectal fossa.
UIN: Researchregistry6727.

CRediT authorship contribution statement

AZ: Writing manuscript, review and editing, data collection, literature review
AQ: Writing manuscript, review and editing, data collection, literature review
AA: Data collection, literature review
TS: Supervision, literature review
MT: Supervision, literature review.

Declaration of competing interest

None declared.

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