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CASE REPORT Open Access

A rare enterouterine fistula between uterine leiomyoma and sigmoid colon in a nulliparous woman: a case report

M. Maivizhi Shruthi[1*], Biswajit Sahoo[1], Saubhagya Kumar Jena[2], Pruthwiraj Sethi[2], Subarna Mitra[2], Manoj Kumar Nayak[1], Shilpy Jha[3], Nerbadyswari Deep Bag[1] and Laveti Sai Sarvazna[2]

Abstract

Background The commonest benign neoplasm encountered in women of reproductive age group is uterine leiomyoma which usually presents with abnormal uterine bleeding, chronic pelvic pain, urinary disturbances, urinary retention, constipation, and dyspareunia. Uterine leiomyoma presenting with bowel fistulization is a rare presentation. To our knowledge and based on a thorough literature search, no previous reports of uterine leiomyoma showing spontaneous fistulous communication with the sigmoid colon have been reported.

Case presentation A nulliparous woman in her early 30s presented with complaints of lower abdominal pain, abdominal fullness, pus discharge from the vagina, and difficulty in respiration. On contrast-enhanced computed tomography (CECT), a large heterogeneously enhancing mass was seen in the rectouterine space showing air-fluid levels, with a fistulous communication with the adjacent sigmoid colon. Histopathological examination proved the lesion to be a uterine leiomyoma.

Conclusion The chance of fistulization with the bowel should be considered whenever a uterine mass with an air-fluid level is encountered.

Keywords Uterine leiomyoma, Bowel fistulization, Sigmoid colon

Background

Uterine leiomyoma is the commonest benign neoplasm encountered in women of reproductive age. It commonly presents with abnormal uterine bleeding, chronic pelvic pain, urinary disturbances, urinary retention, constipation, and dyspareunia [1]. Uterine leiomyoma presenting with bowel fistulization is a rare presentation. Although few cases of uterocutaneous fistula and ileouterine fistula have been reported prior, uterosigmoid fistula, due to leiomyoma, has never been reported. We present a unique case of uterine leiomyoma showing fistulous communication with the sigmoid colon. Spontaneous fistulous communication of leiomyoma without known risk factors for fistula formation, like prior surgery or uterine artery embolization, is extremely rare.

Case presentation

A nulliparous woman in her early 30s presented to the gynecology department with complaints of lower abdominal pain, abdominal fullness, pus discharge from the vagina, loss of appetite, and difficulty in respiration for 1 month. She has been married for 10 years and was nulliparous. Menstrual history revealed regular menstrual

*Correspondence:
M. Maivizhi Shruthi
maivizhishruthi17@gmail.com
[1] Department of Radiodiagnosis, All India Institute of Medical Sciences, Bhubaneswar, Odisha 751019, India
[2] Department of Obstetrics and Gynaecology, All India Institute of Medical Sciences, Bhubaneswar, Odisha, India
[3] Department of Pathology, All India Institute of Medical Sciences, Bhubaneswar, Odisha, India

cycles for every 28–30 days, lasting for 3 to 4 days. On perabdominal examination, a large mass was noted in the lower abdomen measuring approximately 15×18 cm which was soft in its consistency with the lower pole of the mass not palpable. Per speculum examination showed pus discharge from the vagina obscuring the cervix's view. Initial ultrasound examination showed a large heterogeneous solid mass measuring 12cmx16cm in the pelvis, displacing and compressing the uterus anteriorly. Bilateral ovaries were not separately visualized. For further evaluation of the mass, CECT was done, which revealed a sizeable ill-defined mass in the rectouterine space with possible origin from the posterior wall of the uterus or cervix measuring approximately 13×14x14cm. The lesion was displacing the uterine body anterosuperiorly (Fig. 1). A significant portion of the mass was necrotic and showed air-fluid levels, with suspicious communication with the adjacent large bowel loops. Rectal positive contrast confirmed a fistulous communication with the sigmoid colon, causing an air-fluid level within the mass(Fig. 2). The mass was compressing the right lower ureter causing moderate hydroureteronephrosis. Succeeding CECT, colonoscopy showed a sizeable fistulous opening in the sigmoid colon at 20 cm from the anal verge with stool inside the cavity. During the hospital course, the patient developed deep vein thrombosis in the left common iliac, external iliac, common femoral, and popliteal veins. The inferior vena cava

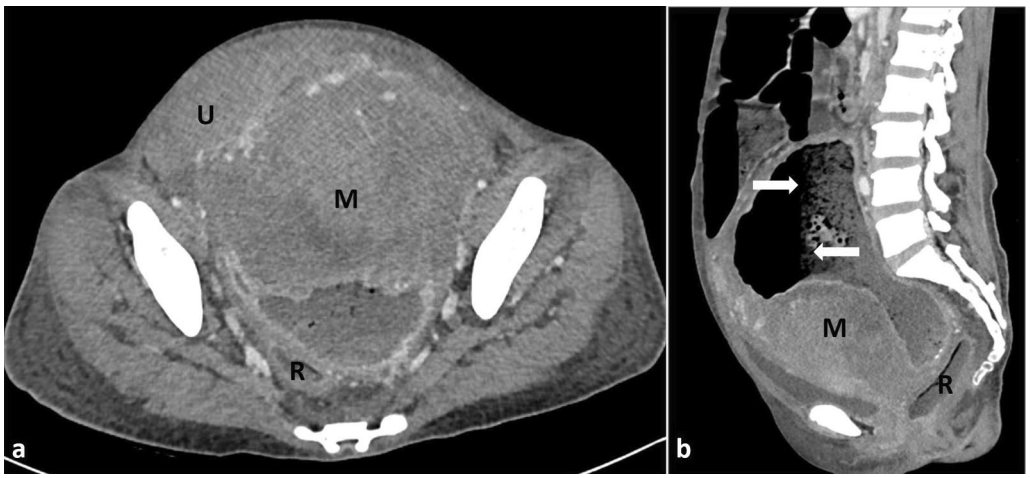

Fig. 1 CECT axial **a** and sagittal **b** images in a nulliparous woman in her early 30s, with complaints of lower abdominal pain, pus discharge from vagina showing a large mass (M) in the rectouterine space displacing the uterus (U) anteriorly and the rectum (R) posteriorly with evidence of air-fluid level (white arrows) within the mass

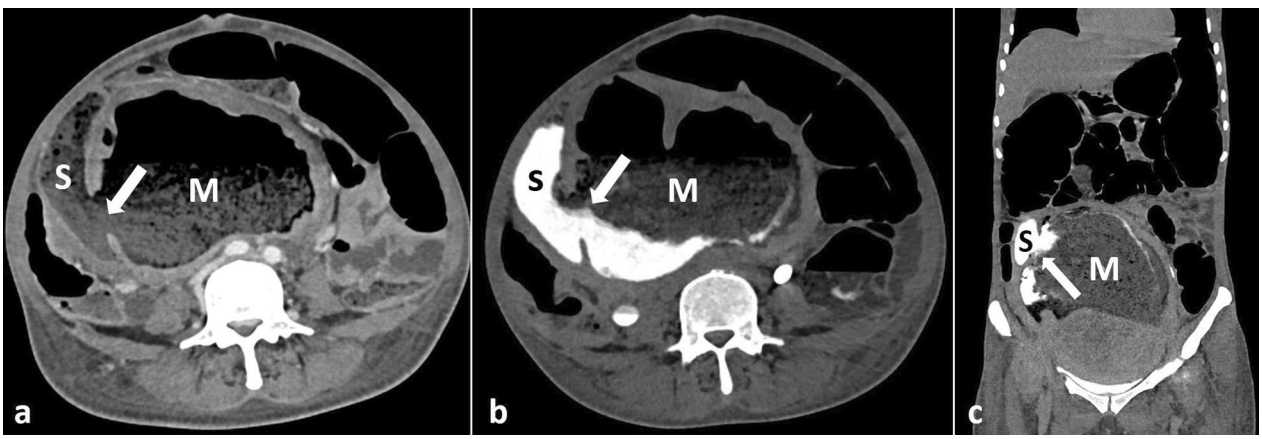

Fig. 2 CECT axial image without rectal contrast **a** showing a fistulous communication (white arrow) between the mass (M) and the adjacent sigmoid colon (S) with the presence of air-fluid level within the mass. CT axial **b** and coronal **c** images with rectal contrast showing contrast from the sigmoid colon (S) entering into uterine mass (M) depicting fistulous communication (white arrow) of uterine mass with the sigmoid colon

and right-sided venous system appear normal. On ultrasound guidance, a core needle biopsy was performed. Histopathological examination showed benign spindle cell neoplasm with tumor cells arranged in intersecting fascicles, indistinct cytoplasmic borders, eosinophilic to pale cytoplasm, and cigar-shaped nuclei consistent with leiomyoma. These tumor cells exhibit mild atypia and rare mitoses (Fig. 3). Immunohistochemistry showed strong and diffuse positivity for smooth muscle actin, and CD117 was negative in these tumor cells.

Discussion

Uterine leiomyoma is the commonest benign neoplasm encountered in women of reproductive age. The risk factor of fibroid includes early menarche, late menopause, obesity, nulliparity, and a family history of fibroids [1]. Complications of uterine fibroid can be categorized into urological and non-urological complications. Non-urological complications are torsion of large uterine leiomyoma, cystic degeneration, anemia, circulatory problems like thrombus formation, bowel disturbances such as constipation, and pregnancy-related complications like miscarriage or infertility. Urological complications include urethral or ureteral obstruction, renal failure, and hematuria. Urinary retention, a common complication in uterine fibroids, occurs due to compression of the bladder outlet caused by leiomyoma involving the broad ligament. Compression of ureters by leiomyoma occurring at the level of the pelvic brim causes obstructive uropathy, which can be unilateral or bilateral [2].

The most commonly occurring urogenital fistula includes vesicovaginal fistula and uterovaginal fistula. Strenuous and prolonged labor, cesarean section, and gynecological surgeries are the common preceding risk factors for urogenital fistula. Enterouterine fistula is an infrequent occurrence. The pathogenesis behind enterouterine fistula includes inflammatory processes like Crohn's disease, diverticulitis, peritonitis, spontaneous or iatrogenic uterine rupture, neoplasms involving uterus or bowel loops with subsequent invasion, and fistulous communication with each other. Fistula formation in cases of uterine leiomyoma can occur either due to prior history of surgery, which disrupts the integrity of the wall of the uterus and promotes adhesion and fistula formation, or prior history of uterine artery embolization, which can cause the leiomyoma to degenerate resulting in an inflammatory response succeeded by fistulization. A case of fistulous communication between uterine leiomyoma and the abdominal wall is reported prior by Anderson et al.[3]. Degenerated leiomyoma, which shows inflammatory changes and necrosis, can promote fistula formation. A case of fistula formation between degenerated leiomyoma of the uterus with the urinary bladder has been reported by Fridman et al.[4]. A case of uterine leiomyoma, which had undergone degenerative changes during pregnancy and was complicated by a cesarean section that showed fistulous communication with an ileal loop, has been reported [5].

The types of degeneration in leiomyoma are hyaline, cystic, calcific, septic, or myxomatous, and carneous or red degeneration. A case of degenerated uterine leiomyoma with air-fluid level has been reported where septic degeneration of fibroid had caused air-fluid levels [6]. Neoplasm of the large intestine causing sigmoid uterine fistula has been priorly reported. The uterus is a thick muscular organ, and extension of inflammation and succeeding fistulization is extremely rare. Carcinoma rectosigmoid-causing infiltration of the uterus and subsequent fistulization with the uterus has been reported [7]. In our case, there was a fistulous communication between the degenerated uterine leiomyoma and the adjacent sigmoid colon, which was proved by radiological and histopathological examinations.

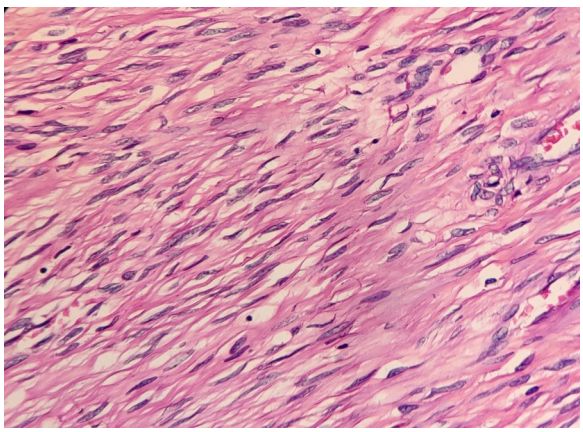

Fig. 3 Histopathological examination (Hematoxylin and eosin, 400X) showed benign spindle cell neoplasm with tumor cells arranged in intersecting fascicles, indistinct cytoplasmic borders, eosinophilic to pale cytoplasm, and cigar-shaped nuclei consistent with leiomyoma

Conclusions

To our knowledge and based on a thorough literature search, no previous reports of uterine leiomyoma showing spontaneous fistulous communication with the sigmoid colon have been reported. In conclusion, the chance of fistulization with the bowel should be considered whenever a uterine mass with an air-fluid level is encountered. Moreover, CECT and histopathological examination can be a helping hand in reaching the correct diagnosis.

Abbreviation
CECT Contrast-enhanced computed tomography

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Author contributions
MSM contributed to the conception, prepared the manuscript, and drafted and revised the article. BS and MKN contributed to the concept and design, manuscript preparation, article drafting, and final approval. SKJ, PS, SM, and LSS contributed to data collection and other patient information, drafting the article. SJ made the final pathological diagnosis, helped draft the article, and reviewed it. NDB contributed to revising the article, supervision, and final approval. All authors have read and approved the manuscript.

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Availability of data and materials
The imaging data are available in departmental PACS in DICOM format. Other patient data are with the medical records department.

Declarations

Ethics approval and consent to participate
Written informed consent for publication of their clinical details and/or clinical images was obtained from the patient. A copy of the consent form is available for review by the Editor of this journal. Ethics approval has been waived for case reports with written informed consent by Institutional Ethics Committee, AIIMS Bhubaneswar.

Consent for publication
Written informed consent for publication of their clinical details and/or clinical images was obtained from the patient. A copy of the consent form is available for review by the Editor of this journal.

Competing interests
The authors declare that they have no competing interests.

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