

Case report

Hysterotomy Scar Abdomino-Uterine Fistulas: A Post-Cesarean Finding

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Introduction

Uterine fistula is a rare complication of cesarean section. While there are descriptions of vesicouterine, uterocutaneous, and ureterouterine fistulas after cesarean section, there are no reports in the literature of abdomino-uterine or utero-cervical fistulas arising from hysterotomy scars in patients with prior cesarean sections [1, 2, 3]. A case series of 15 patients with prior remote histories of cesarean section presented with chronic pelvic pain and specific abdominal point tenderness to palpation over the cesarean skin incision. All 15 patients demonstrated laparoscopic evidence of densely adherent uterine adhesions to the anterior abdominal wall and intraoperative or gross specimen evidence of abdomino-uterine fistulas.

Methods

An institutional review board (IRB)-approved retrospective chart review was performed on this collection of patients as a case series. These patients received care between August 2012 and May 2013, and their charts were reviewed during this time period. All available laparoscopic and hysteroscopic images and operative findings in dictated reports by the primary surgeon were reviewed. Additionally, all pathologic findings including pre-operative endometrial biopsies and post-operative tissue specimens were reviewed.

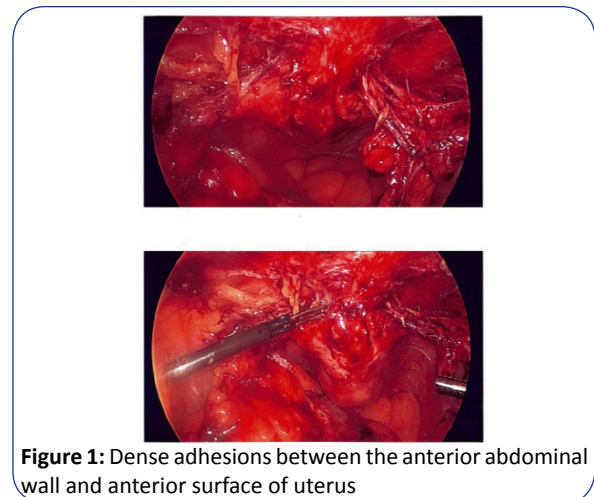
Case Presentations

A case series of 15 patients with prior remote histories of cesarean section by Pfannenstiel or vertical incision presented with chronic pelvic pain and specific, exquisite abdominal point tenderness to deep palpation over the cesarean skin incision. None of these 15 patients had undergone cesarean section performed by neither the lead surgeon nor any partner in his practice. In all cases the pain was refractory to prior medical and surgical interventions. Fourteen of the patients were offered and underwent laparoscopic supracervical hysterectomy (LSH), and one patient underwent laparoscopic trachelectomy. Nine patients underwent hysteroscopy prior to or concurrently with hysterectomy. All 15 patients underwent concurrent laparoscopic lysis of adhesions. These patients received care between August 2012 and May 2013.

The mean age was 40. The mean BMI was 32. A triad of dysmenorrhea, menorrhagia, and dyspareunia was described in 4 patients. Four patients had diagnoses of endometriosis. The failed

medical therapies were tramadol, leuprolide, and topiramate. One patient underwent serial superior hypogastric nerve blocks prior to hysterectomy for analgesia, which ultimately did not resolve the pain long-term. Prior failed surgical evaluations included dilation and curettage, numerous diagnostic and operative laparoscopies, and endometrial ablation. One patient had undergone a prior LSH from an outside provider.

All 15 patients demonstrated dense adhesions between the anterior abdominal wall and uterus (in the lower uterine segment area of the prior cesarean hysterotomy) (Figure 1).



Increased bleeding was subjectively encountered in all 15 cases during lysis of adhesions specifically between the uterus and

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abdominal wall. The estimated blood loss ranged from 25-60 milliliters per case. During laparoscopic dissection of the anterior abdominal wall adhesions off the lower uterine segment, an abdomino-uterine fistula was identified in all 15 cases (Figure 2) in the area of the prior cesarean hysterotomy. In 4/15 cases hysteroscopy demonstrated a fistulous tract visualized in the lower uterine segment or cervix (Figure 3).

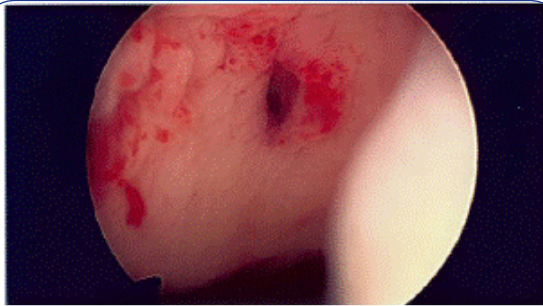


Figure 3: Hysteroscopic view of a fistula tract in the right anterolateral endometrial cavity

In 14/15 cases care was taken to excise the fistula completely, including repetitive excisions of the cervix in a disc-like fashion until no gross evidence of the fistula remained. The remaining one case of trachelectomy removed the fistula via complete cervical excision.

The patient status post prior supracervical LSH for pain attributed to endometriosis from an outside provider underwent laparoscopic trachelectomy with our department. Intraoperative inspection revealed a retained portion of the lower uterine segment to the cervix from the prior LSH. The excised cervix with lower uterine segment in situ grossly demonstrated the fistula pathway using a lacrimal dilator with the yellow and red arrows demonstrating dilator entry and exit, respectively (Figure 4).

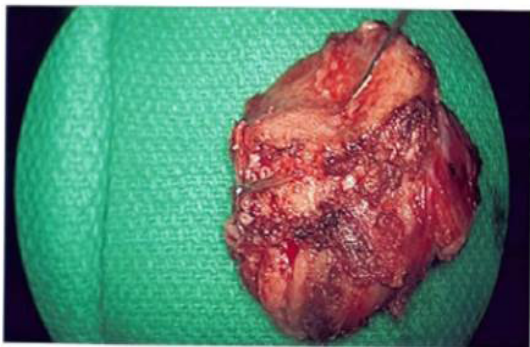


Image 4: Cervix excised during trachelectomy with yellow arrow demonstrating the entrance direction of the lacrimal dilator into the fistula tract and the red arrow demonstrating the exit of the dilator from the fistula tract

Unfortunately, given the surgical approach of LSH, 14/15 specimens were submitted to pathology morcellated (with the 1 exception being the trachelectomy), so pathology fistulograms were not able to be obtained to further describe the fistula tract anatomy. Pathologic

analyses were conducted in standard gross fashion by the affiliated hospital's pathology department. There were no pathology reports of malignancy or hyperplasia. Observationally, 15/15 of these patients experienced resolution of pelvic pain. Of note, during the same time period, the lead surgeon did not encounter any patients with this same clinical presentation (of specific point tenderness overlying the prior cesarean skin incision) who did not have the described laparoscopic findings. The lead surgeon conducts a busy gynecologic practice with a high percentage of women with chronic pelvic pain, approximately 25-30%. However, all of the patients who received care with this exact constellation of clinical symptoms were taken to the operating room and had similar laparoscopic findings suggestive of an abdomino-uterine fistula. Thus, no patients encountered to this point with this specific clinical presentation showed any absence of an abdomino-uterine fistula.

Discussion

Post-cesarean uterine fistulas are rare; vesicouterine fistulas after cesarean comprise 1-4% of all genitourinary fistulas [1]. There are 4 described management cases of uterocutaneous fistulas, and one case report describes a utero-uterine fistula as a complication in the context of cesarean section [1, 2, 3, 4, 5]. One of the earliest described vesicouterine fistulas was Youssef's syndrome, which characterized vesicouterine fistulas in patients with histories of cesarean sections presenting with amenorrhea or menoruia (cyclic hematuria) [6,7]. Vesicouterine fistulas after cesarean section can also present with urinary incontinence [8]. This is the first case series of abdomino-uterine and utero-cervical fistulas arising from hysterotomy scars in patients with prior cesarean sections.

This case series presents laproscopically-identified abdomino-uterine and cervico-uterine fistulas as an etiology for specific abdomino-pelvic point tenderness in the background of chronic pelvic pain unresponsive to medical therapy in women with remote prior cesarean delivery. Our practice found some clinical applications useful when encountering these patients intraoperatively. If the clinical history gave suspicion for abdomino-uterine fistulas in the context of pain, hysteroscopy prior to or concurrent with the definitive management of hysterectomy was performed. Using a 70° scope may provide the best advantage to view the fistulous tract from the endometrial perspective. A 30° or 0° hysteroscope may be inadequate to visualize these types of fistulas in their anterior & lateral location. In intraoperative setting during laparoscopic lysis adhesions between the uterus and anterior abdominal wall, it was helpful to anticipate the increased bleeding (especially of dark, endometriosis-colored blood) in these scarred planes and continue the dissection until appropriate mobilization of the adherent bladder was accomplished. Ensuring the uterocervical junction has been completely dissected is critical, as the goal is to encounter the fistula in entirety. Amputation of the cervix must be inferior to the uterocervical junction (even if that anatomic level is difficult to ascertain) to completely excise the fistula (if a supracervical hysterectomy is performed). Retrograde

filling of the bladder may assist in revealing the bladder outline and thus decrease risk of bladder injury.

These patients with specific clinical presentations represent an interesting population and foundation for future studies. Further prospective investigations of a larger number of patients could help better describe demographics, patient symptomatology, and risk associations. Outpatient imaging when given this clinical scenario could provide specific delineations of the exact fistula pathways and thus help define the ideal imaging modalities in diagnosing these fistulas pre-treatment. Further studies prospectively are needed to discover the best imaging modalities. Possibilities to augment any hysteroscopic findings of fistulas include computer tomography with fistulograms or magnetic resonance imaging. Whole specimen pathology studies with fistulograms could be conducted in research patients who are offered definitive surgical management. Studies combining detailed outpatient workups and further intraoperative inspections can spur data to support randomized controlled trials that eventually explore therapeutic options.

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