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Medically Refractory Ulcerative Colitis in Third Trimester Pregnancy, Treated with Diverting Loop Ileostomy and Turnbull Transverse Colostomy

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Background	A 35-year-old woman with long-standing ulcerative colitis (UC) developed severe medically-refractory symptoms in her third trimester of pregnancy. Despite best medical efforts, the patient lost weight during her last trimester and required opioids for pain control. Given her large gravid uterus she underwent a Turnbull "blow hole" colostomy and diverting loop ileostomy to minimize uterine manipulation and anesthetic exposure and mitigate risks of preterm labor. Her postoperative course was complicated by recurrent loop ileostomy prolapse requiring local revision. She eventually delivered a healthy boy at 36 weeks via scheduled cesarean section. Six months following delivery, she underwent restorative proctocolectomy with a diverted ileal pouch anal anastomosis (IPAA) with loop ileostomy reversal four months later.
Summary	UC can rarely require surgery during pregnancy whereby presenting a surgical dilemma by risking the well-being of mother and fetus. Turnbull colostomy and loop ileostomy are temporizing options for the management of surgical colitis during late term pregnancy as a bridge to eventual postpartum restorative proctocolectomy and IPAA.
Conclusion	Coordinated care between gastroenterology and surgical teams may help delay surgery until the postpartum state, however some UC patients may require surgery during pregnancy. Temporizing measures such as the Turnbull colostomy with loop ileostomy may avoid fetal stressors associated with colectomy and should remain part of the surgeon's armamentarium for difficult situations.
Keywords	Ulcerative colitis, Turnbull Colostomy, inflammatory bowel disease during pregnancy

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Case Description

A 35-year-old woman with a 10-year history of ulcerative colitis (UC) was admitted to the hospital for an acute UC exacerbation in her 29th week of pregnancy. In the two years before her admission she failed multiple biologic agents and immunomodulators before experiencing one year of symptom control with adalimumab. Approximately four weeks prior to admission she experienced increasingly frequent bowel motions despite the addition of prednisone and additional adalimumab dosing. Serum adalimumab levels were therapeutic and anti-adalimumab antibodies were absent. At admission, the patient noted 15 bloody bowel movements daily, a 16-pound one-month weight loss, and intractable pelvic pain requiring opioid analgesics.

Intravenous methylprednisone was initiated upon admission but failed to improve her symptoms. Extensive stool testing for infectious colitis was negative. Limited carbon dioxide flexible sigmoidoscopy performed on the sixth hospital day demonstrated friable erythematous mucosa from anal verge to the distal sigmoid colon and biopsies revealed cryptitis and crypt abscess consistent with chronic ulcerative colitis with activity. Cytomegalovirus (CMV) immunostains and serum CMV titers were negative.

Fetal monitoring throughout her hospitalization showed no fetal distress and fetal weight was estimated to be 1641 grams (73rd percentile) via ultrasound. Following six days of IV steroid therapy, cyclosporine salvage therapy was cautiously trialed following detailed discussions with the patient's care teams. The patient's symptoms failed to improve despite six days of cyclosporine infusion and the patient lost an additional 3 kg of mass. Multidisciplinary discussions were repeated and given persistent severe symptoms, ongoing weight loss, and persistent opioid requirements, surgical intervention was deemed necessary. Neonatal intensive care unit, obstetric anesthesiology, and wound, ostomy, and continence nursing consultations were sought as surgery was planned. Two upper abdominal stoma sites were marked and the patient received preoperative stoma education.

She underwent Turnbull transverse "blow hole" colostomy and loop ileostomy with a general anesthetic and epidural with continuous fetal monitoring on the 14th hospital day (31st week of pregnancy). Maternal fetal medicine

and the neonatal intensive care unit were present if fetal stress required immediate cesarean. A 10 cm supra-umbilical vertical midline incision was made just large enough to accommodate the surgeon's hand. The inspected colon revealed erythematous pancolitis without signs of ischemia and the small bowel showed no evidence of Crohn disease. A loop of ileum 40 cm proximal to the ileocecal valve was delivered over a rod through a previously marked muscle-splitting stoma trephine in the right upper quadrant. The loop ileostomy was intentionally made more proximal than usual so as not to interfere with an eventual ileoanal pouch. A separate 3 cm subxiphoid incision was made through the linea alba. Continuous running suture was used to create a watertight seal between the transverse colon serosa and peritoneum. Following midline wound closure, the loop ileostomy was matured. A vertical colotomy opened the blowhole colostomy and the open colon edge was matured flush circumferentially with the dermis with continuous suture (Figure 1 and Figure 2). Total anesthetic time, which included surgery, positioning, urinary catheterization, fetal monitoring and extubation was approximately 60 minutes.



Figure 1. Turnbull blow hole colostomy and loop ileostomy, approximately 30 days postpartum. The efferent limb of the loop ileostomy has been sutured close to temporize against recurrent prolapse.



Figure 2. Close up view of the Turnbull loop colostomy, approximately 30 days postpartum.

Mother and fetus tolerated the surgery without complications. Cyclosporine and adalimumab were discontinued and intravenous steroids were transitioned to an oral steroid taper. The patient was discharged home on the 11th postoperative day after resolution of postoperative ileus. Her postoperative course was complicated by recurrent prolapse of the efferent limb of the loop ileostomy that required multiple local reductions with and without anesthetic.

The patient underwent elective cesarean section of a healthy boy at the 36th week of gestation with reduction of recurrent loop ileostomy prolapse. Six weeks post-partum, she again developed recurrent prolapse of the efferent ileostomy limb requiring local conversion to an end ileostomy with closure of the efferent ileostomy limb. At three postpartum months, she underwent total proctocolectomy with ileal pouch anal anastomosis (IPAA) and diverting loop ileostomy with eventual loop ileostomy reversal four months after creation.

Discussion

UC is a disease process that affects a young patient population with peak incidence during reproductive years. An estimated 30–50 percent of female patients with UC will experience an exacerbation while pregnant or in early postpartum period. Recent advances in pharmaceuticals, particularly biologic agents, provide physicians with more medical options, but a small fraction of patients will develop medically refractory disease, hemorrhage, perforation, toxic colitis, or sepsis that requires surgery in the gravid state. While perforation, massive hemorrhage, and toxic megacolon are typically steadfast surgical indications, declaring a patient refractory to medicine requires multidisciplinary input to exhaust all options deemed safe for the mother and fetus.

If possible, non-urgent surgery should be performed in the second trimester when preterm contractions and spontaneous abortion are least likely. However, recent consensus guidelines state a pregnant woman should never be denied indicated surgery, regardless of trimester.² Surgery during pregnancy for UC has historically been a challenging issue with high risk for mother and fetus.³ Preterm delivery, low birth weight, and "small for gestational age" are adverse fetal outcomes associated with active UC during conception or pregnancy.4 A systematic review of 4,473 gravid patients undergoing non-obstetric surgery revealed an overall miscarriage rate of 5.8 percent, but approached 11 percent for women in the first trimester. Premature delivery occurred in 8.2 percent of patients and non-obstetric surgery induced delivery in 3.5 percent of patients, and overall fetal loss rate was 2.5 percent in this cohort of mostly dated studies involving acute appendectomy and cholecystectomy.5 Since maternal gestational weight gain is a strong predictor of adverse pregnancy outcomes in inflammatory bowel disease (IBD)⁶ the entire surgical care team must show equipoise when deciding on performing surgery during pregnancy.

Restorative proctocolectomy with IPAA (e.g., "J-pouch") is the ultimate goal for most UC patients requiring surgery, but pelvic surgery during pregnancy is typically avoided given bleeding risks arising from an engorged pelvic venous plexus and the gravid uterus which hinders visualization. Moreover, a staged approach to restorative proctocolectomy is typically employed in urgent and emergent settings or in the setting of potent immunosuppressives to minimize postoperative septic complications. Considering the additional nuances associated with pregnancy, surgical options for the gravid UC patient include subtotal colecto-

my with end ileostomy, partial colectomy with colostomy and mucous fistula, loop ileostomy, and Turnbull colostomy with diverting ileostomy via laparoscopy or open approaches.

Turnbull first described combined diverting loop ileostomy and decompressive "blow hole" transverse colostomy in 1971 to treat severely ill UC patients with toxic megacolon⁸ which was later adapted for pregnant UC patients requiring colectomy.⁹ The "blow hole" colostomy decompresses the colon to avoid immediate resection, and loop ileostomy diversion allows colonic inflammation to gradually improve. Following fetal delivery and maternal convalescence, restorative proctocolectomy can later be completed with lower risk to mother and child under more favorable conditions.

Spartan studies guide optimal surgical management for pregnant UC patients requiring surgery. Wilson et al. reported 38 cases of UC requiring surgical intervention during pregnancy. Sixty percent of patients underwent surgery during the second trimester, and approximately 40 percent of patients had surgery in conjunction with cesarean section.3 Dozois and colleagues reported five pregnant women with fulminant UC who required surgery in the second trimester (3 of 5), first trimester (1 of 5) and third trimester (1 of 5).10 All patients underwent subtotal colectomy and end ileostomy with zero maternofetal mortality and the majority of patients underwent eventual IPAA. The authors proposed using the Turnbull procedure for severely ill and unstable pregnant patients while using subtotal colectomy with ileostomy for all others. Killeen and colleagues later published a systematic review of 56 pregnant UC patients requiring surgery, detailing an overall maternal mortality rate of 19 percent; however, the maternal mortality rate in patients after 1980 was nil.⁷ The authors noted a fetal mortality of 35 percent, and overall rates of preterm labor approached 50 percent, while all third trimester patients universally experienced preterm labor. While the fetus in the index case was viable at 29 weeks, the authors favored a staged approach utilizing the Turnbull procedure in hopes to minimize the risk of preterm labor.

Conclusion

Ulcerative colitis exacerbations are commonly encountered during pregnancy and demand coordinated care between gastroenterologists, surgeons, and obstetricians for optimal maternofetal outcomes. Urgent surgery may rarely be required in gravid UC patients and the surgeon

must conscientiously balance the immediate well-being of mother and child with a long term surgical strategy to produce a suitable functional gastrointestinal outcome for the mother. Staged surgical approaches, such as the Turnbull blowhole procedure, are helpful adjuncts for high risk UC patients requiring surgery under critical conditions. Elective IPAA procedures can be performed at a later date, when surgery is less perilous.

Lessons Learned

Timeliness of surgical interventions for ulcerative colitis coupled with prudent surgical decision minimizes maternofetal risk during pregnancy. Utilization of staged and temporizing surgeries assures the best long term outcomes for gravid mothers with UC and their fetuses.

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