



Captive uterus syndrome: An unrecognized complication of cesarean sections?



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ABSTRACT

Cesarean sections are a common surgical procedure at risk of complications including adhesions and chronic pelvic pain.

This case series presents 10 cases of patients presenting with chronic pelvic pain following a cesarean section which were found upon surgical exploration to have developed adhesions between the abdominal wall and the uterus. When they first consulted, patients were evaluated clinically with a questionnaire and physical exam, and with ultrasonography to evaluate pelvic anatomy when necessary. The evaluation was completed with diagnostic laparoscopy which revealed atypical post-cesarean adhesions which were treated by adhesion lysis or hysterectomy. Surgical protocols of the cesarean sections were retrieved and analysed for potential adhesion risk factors.

Patients presented with chronic pelvic pain that appeared in the early post-operative period. Physical exam revealed a subinvolted uterus with a high fixed cervix. Ultrasound examination revealed clues of adhesions manifested by points of traction and an irregular uterine border. No other diagnosis such as endometriosis, pelvic inflammatory disease, ovarian or bowel anomalies were identified during surgery. After laparoscopic adhesion lysis or hysterectomy, all patients who were treated noted a complete resolution of the pain that lasted during a follow-up of at least 5 years.

These findings suggest that adhesions that create traction and fix the uterus to the abdominal wall following caesarian section can be the cause of severe chronic pelvic pain. In the presence of such pain, clinicians should suspect the presence of adhesions and investigate and treat patients accordingly.

Introduction

According to the World Health Organisation statistics, the cesarean section rate has increased considerably worldwide since 1985. The ideal rate, estimated at 10–15%, has been largely exceeded in Europe and Canada with rates of 25% and 28.4% respectively [1–4].

Women undergoing cesarean section are exposed to several short and long-term complications. Short term complications of cesarean sections include anesthetic complications and surgical complications such as wound infection, hemorrhage, visceral injury, and venous thromboembolism [5,6].

In an American national survey, women undergoing cesarean section were found to be more likely to present postpartum pain, and up to 18% of them still reported pain 6 months postpartum [7]. Considering that the rate of chronic pelvic pain following cesarean delivery may be as high as 7%, a large number of women may be affected by such problems. [8].

Recent medical literature has taken an interest in early post cesarean section pelvic pain [9], while post-cesarean pelvic adhesions have also been studied in the broader context of post-surgical adhesions [10,11].

In light of these studies, the persistence of post-cesarean pain

appears to be the result of neuropathic pain while post-cesarean adhesions share the same etiologies as all post-operative adhesions. The natural evolution of peritubal, periovarian, and intestinal adhesions leads to chronic pelvic pain and infertility.

We noticed during clinical practice that certain patients presented with specific pain symptoms following cesarean section that were linked to the finding of adherences upon surgical exploration. The development of post-cesarean adhesions is in some cases correlated with the immediate post-operative course (fever, pelvic infection, sub-obstruction, hospital stay length), but no evidence of such an inflammatory or infectious process was present in our patients. The aim of the study was to determine whether the formation of these adherences was linked to a specific surgical method or peripartum circumstances, and what was the best course of treatment for this subset of women whose quality of life is impaired by severe pelvic pain [12].

Material and methods

10 cases of post cesarean section pain referred to a general obstetrics and gynecology practice in Montreal (Canada) over a period of ten years (2005–2014) are hereby presented, accompanied by some relevant information concerning the context of the diagnosis, surgical

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method at the time of cesarean section, peripartum circumstances, and patient management. Laparoscopic findings were also described concerning adhesions based on the review of surgical protocols. All patients were followed for a minimum of 5 years after diagnosis. Cases of chronic pelvic pain without prior surgery, endometriosis, and without any adhesions upon surgical exploration were excluded. In 7 out of the 10 cases presented, the surgical protocol of the cesarean section was available. All of the cesarean sections prior to diagnosis were performed in other hospital centers. Consent was obtained from all participating patients for publication of clinical details.

Results

The patients presented in this case series, were found to have what could be called “captive uterus syndrome”, a condition that associates pelvic pain with post-operative adhesions and is characterised by typical early onset lower abdominal pain that persists for several years and irradiates to the groin, is exacerbated by breastfeeding (increase in oxytocin is responsible for both the ejection of milk and contractions of the uterus that allow uterine involution), by certain movements and changes in posture (lying-sitting-standing), and accompanied by deep thrust dyspareunia at the resumption of sexual activity. None of the patients presented symptoms compatible with fibromyalgia, functional digestive disorders, or pelvic inflammatory disease.

Clinical examination, palpation, speculum examination, and digital vaginal examination reveal that the uterus is subinvolved and attached to the abdominal wall, the cervix is high and there is some cervical motion tenderness as a result of adhesions specific to cesarean sections. The anatomical relationships of the post-gravid uterus are disturbed, causing a direct apposition of the uterus with the anterior abdominal wall without an interposition of the intestines or omentum, these elements having been pushed back towards the posterior compartment during pregnancy. No vaginismus or vulvodynia was found upon physical examination and there were no signs of cervicitis or vaginitis.

The characteristics of the patients who presented with captive uterus syndrome are presented in Table 1.

Upon surgical exploration, the adhesions we found to have formed between the uterus and the abdominal wall were of three types (Fig. 1):

1. at the superior aspect with adhesions at the uterine fundus fixing it high on the anterior abdominal wall.
2. the second type corresponds to velamentous adhesions at the middle third of the uterus.
3. and lastly adhesions as severe as those affecting the superior aspect but at the inferior portion of the uterus.

Surgical exploration did not reveal any evidence of comorbid pelvic pathology that could result in chronic pelvic pain (ie: no endometriosis, cyst, hydrosalpinx, or leiomyoma were found). In all of the cases who underwent surgery, with lysis of the adhesions with or without hysterectomy, a complete and sustained remission of the pain was noted post-operatively. Only one patient declined hysterectomy and is still followed for pelvic pain.

Discussion and Hypothesis

This case series aims to identify a new clinical entity caused by adhesions between the uterus and abdominal wall that induce severe pelvic pain arising early after a cesarean section.

Previous studies have described the finding of adhesions between the uterus and abdominal wall in repeat cesarean sections, or when investigating secondary infertility, but have failed to describe specific symptoms allowing suspicion of this syndrome prior to surgical intervention [13–15]. When confronted with this specific clinical presentation and its particular pathophysiological context, after a thorough questionnaire and physical examination, the diagnosis for these

Table 1
Clinical Presentation and Characteristics of Patients Presenting with Captive Uterus Syndrome.

Age (years)	Delivery method and postpartum	Duration of pain	Other symptoms and pathologies	Diagnostic procedures	Treatment	Follow-up
27	Caesarean section, no surgical protocol available.	4 years	Dyspareunia since caesarean section and secondary infertility.	Laparoscopy	Laparoscopic Adhesion Lysis	Resolution of pain
28	Caesarean section for fetal distress non-closure of the peritoneum breastfeeding.	2 months	Dyspareunia	Laparoscopy	Laparoscopic Adhesion lysis	Resolution of pain. Subsequent vaginal delivery.
35	Elective caesarean section, previous myomectomy, closure with absorbable sutures.	5 years	Dyspareunia, secondary infertility and fibromyoma (discovered during pregnancy).	Iterative caesarean section and tubal ligation	Hysterectomy	Resolution of pain
36	Iterative caesarean section with adhesion lysis, myomectomy, prematurity and placental abruption.	2 months	Dyspareunia	Laparoscopy		Resolution of pain
37	Caesarean section for arrest of descent, non-closure of peritoneum.	11 months	Dyspareunia and subsequent pregnancy.	Iterative caesarean section	Hysterectomy	Resolution of pain
38	Iterative caesarean section, adhesion lysis, non-closure of peritoneum, breastfeeding.	3 years	Previous myomectomy.	Laparoscopy	Hysterectomy offered to patient	
40	Iterative caesarean section, closure with absorbable sutures.	6 months	Fibromyoma, consultation for family planning, and dyspareunia.	Laparoscopy	Hysterectomy	Resolution of pain
41	Four iterative caesarean sections, iatrogenic cystostomy, closure with absorbable sutures.	9 years	Dyspareunia	Laparoscopy	Hysterectomy	Resolution of pain
44	Caesarean section for arrest of descent, no surgical protocol available.	17 years	Multiple medical consultations, dyspareunia.	Laparoscopy	Laparoscopic Adhesion lysis	Resolution of pain
52	Three elective caesarean sections, tubal ligation, no surgical protocol available.	24 years	Dyspareunia, conjugal separation, depression, and severe dysmenorrhea.		Hysterectomy	Resolution of pain

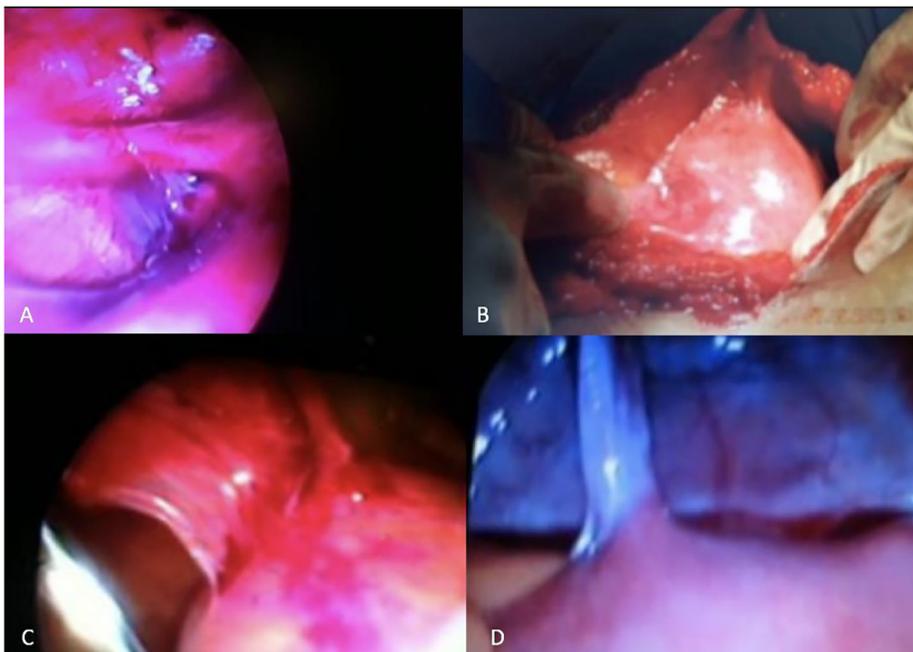


Fig. 1. Adhesions between the uterus and abdominal wall found upon surgical exploration. (A) Fibromuscular adhesion between the uterus (left) and the abdominal wall (right). (B) Adhesion between the uterus and the anterior abdominal wall going all the way to the left tube. (C) Adhesion between the uterus (right), the anterior abdominal wall (up) and the vesico-uterine fold. (D) Adhesion between the uterus (bottom) and the abdominal wall (up).

patients becomes clear.

However, in older women who consult at a later stage of the disease, the differential diagnosis includes a large fibromatous or adenomyomatous uterus, especially for women who already underwent a myomectomy. In cases where the clinical picture is not as clear, pelvic ultrasound may be a helpful complementary diagnostic tool (Fig. 2). In recent years, radiologists have begun studying the sonographic diagnosis of post-cesarean adhesions, as suggested by an elongation of the cervix, an immobility of the uterus and ovaries, an apposition of the uterus and abdominal wall, or the presence of uterine peaking [16,17]. Moro et al. were able to identify adhesions in 45% of patients who had undergone prior cesarean section and found a significant association between the presence of uterus-abdominal wall adhesions and chronic pelvic pain [16].

In clinical practice, the most essential diagnostic tool is laparoscopy, which has both diagnostic and therapeutic purposes, allowing the lysis of adhesions once they are identified. Laparoscopy techniques may be challenging when adhesions render abdominal entry difficult. This problem can be solved by using a sub-splenic entry or an open laparoscopy technique. Some cases do not allow for adhesion lysis through laparoscopy, in this situation the use of laparotomy or even abdominal hysterectomy is warranted [18].

This post cesarean pain syndrome is evocative of that experienced historically by women following uterine ventrofixation surgery as practiced since the late XIXth century (modified Gilliam uterine

suspension), and which has since been abandoned in favor of round ligament plication [19].

Adhesions are the result of micro or macro traumatism of the anterior wall of the uterus in direct apposition with the peritoneal region undergoing a reparation process. The quick regeneration of the peritoneum does not allow sufficient time for the uterus to reach the pelvic cavity during its involution. Thus, the uterus almost becomes an abdominal organ with an elongation of the uterus and cervix, but the bladder is generally left out of this ascension. The cervix becomes hard to visualize during speculum examination, especially with a full bladder. Finally, superior adhesions may form a musculo-connective cord that suspends the uterus.

The factors that increase this adhesive process are the following [20–22]:

- The precise nature of the surgical techniques employed: abdominal surgeries entering the abdominal cavity by peritoneal incision, and subsequent incision of the lower segment of the uterus create a refractory process allowing adhesion formation. During certain interventions, the lower segment is not properly developed, especially in cases of great prematurity and elective cesarean sections. In these circumstances, a classical or T uterine incision is performed which increases the risk of adhesion [23].
- Manipulation of tissues and suturing material can also be involved: the roughness of tissue manipulation increases the risk for micro



Fig. 2. Ultrasonographic appearance of adhesions. (A) Transverse view of the uterine body presenting an irregular outline and an anterior vaulting caused by the traction of an adhesion with the abdominal wall (arrow). (B) Sagittal view of the uterine body. The vaulting is seen again at the uterine fundus (arrow).

traumatism. Natural absorbable sutures are more inflammatory since they are made of animal proteins which induce a foreign body reaction. The white blood cells recruited in this reaction secrete proteolytic enzymes that allow the degradation of the suture material. This inflammatory reaction is thought to explain the increased risk of adhesions associated with natural absorbable sutures when compared with non-absorbable sutures [24–27].

- Additional surgeries at the time of the cesarean section are factors that increase operating time and increase manipulations, especially myomectomies, ovarian cyst removal, tubal ligation, and lysis of previous adhesions. Overall, these procedures increase the risk of adhesion formation post-operatively.
- The issue of the closure or non-closure of the peritoneum is primordial. Historically in the 1970s, with the advent of tubal microsurgery, the preservation of the peritoneum was generally accepted to be an important protective factor for the formation of adhesions. Indeed, in this period, the standard of care was to close all surgically opened layers to diminish the risk of adhesion formation, while manipulating the tissues as little as possible, and using non-absorbable sutures. Beginning the 1980s, surgical oncologists stopped closing the peritoneum during invasive oncological surgeries with lymphadenectomy and omentectomy and reported a decrease in adhesion formation. This question was addressed again by gynaecologists in the 1990s, first regarding hysterectomies, and later cesarean sections, and it was concluded that the non-closure of the peritoneum could be beneficial by decreasing operating time while not increasing the risk of adhesion. However, this topic resurfaced recently as new findings suggested that it was unclear whether the risk of adhesion was diminished by the non-closure of the peritoneum [28–34].

Diagnostic delay played an important role in the clinical presentation of our patients: the longer the delay, the more disastrous were the effects on their quality of life. The unrecognition of their condition led to social and psychological difficulties, secondary infertility, and significant morbidity in repeat cesarean sections. A pelvic ultrasound was rarely performed, apart from cases with a previous myomectomy or in very late diagnoses. The causal factors listed above also played an important role in the genesis of adhesions, such as iterative cesarean sections, myomectomies, concomitant lysis of adhesions, and tubal ligation. The use of non-absorbable sutures is also to be noted. Confirming the absence of tubal or digestive pathologies in these patients is also of critical importance in making the diagnosis. In the cases where hysterectomy was ultimately performed, it was practiced in the presence of very severe adhesions in women who no longer wished to become pregnant (some of them having undergone prior tubal ligation).

The disappearance of the pain is a topic that may stir up controversy, however in this group of patients, the clear concordance between the post-cesarean clinical course and the onset of the pain can only be explained by the physiological knowledge of the phenomenon of the fixation of the uterus to the abdominal wall. After lysis of the adhesions, a complete and sustained (over the course of at least 5 years of follow-up for all patients) resolution of the pain was noted.

It is also possible that patients might present this same type of adhesions without chronic pelvic pain, but we have not encountered such patients in our practice.

Several limitations are associated with an observational study such as the case series hereby presented, which limits the statistical conclusions that can be drawn, but given that patients were referred from various centers with heterogeneous surgical methods, we deemed the realization of a well-constructed case-control study unfeasible. Moreover, since a similar clinical syndrome could not be retrieved in the literature, we deemed the publication of our results necessary.

Further analytical studies concerning this emerging condition will be required to validate its definition and to investigate specific aetiologies for this syndrome.

Conclusion

The warning signs of chronic pelvic pain related to severe post-cesarean adhesions are characteristic and should arouse suspicion of physicians to the presence of captive uterus syndrome. The rate of occurrence of this problem is difficult to establish, especially in a small case-series such as the one presented, but the increase in the rate of cesarean sections can allow us to assume that its incidence will also increase. We hope that a better recognition of this condition will allow earlier treatment of the affected patients, thus increasing drastically their quality of life. Further epidemiological studies will be required to confirm the veracity of this observational study.

Conflict of interest

None.

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