



Paper Accepted*

ISSN Online 2406-0895

Case Report / Приказ случаја

Radmila Sparić^{1,2,†}, Nebojša Radunović^{1,2,3},
Andrea Tinelli^{4,5}, Olivera Radević², Saša Kadija^{1,2}

Large myomas as a complicating factor necessitating cesarean myomectomy followed by cesarean hysterectomy

Хистеректомија као компликација великог миома и неопходне миомектомије током царског реза

¹ Clinic for Gynecology and Obstetrics, Clinical Centre of Serbia, Belgrade, Serbia;

² School of Medicine, University of Belgrade, Belgrade, Serbia;

³ Serbian Academy of Sciences and Arts, Belgrade, Serbia;

⁴ The International Translational Medicine and Biomodelling Research Group, Department of Applied Mathematics, Moscow Institute of Physics and Technology (State University), Moscow, Russia;

⁵ Department of Obstetrics and Gynecology, Division of Experimental Endoscopic Surgery, Imaging, Technology and Minimally Invasive Therapy, Vito Fazzi Hospital, Lecce, Italy

Received: April 4, 2017

Revised: July 17, 2017

Accepted: July 21, 2017

Online First: August 1, 2017

DOI: <https://doi.org/10.2298/SARH170404150S>

* **Accepted papers** are articles in press that have gone through due peer review process and have been accepted for publication by the Editorial Board of the *Serbian Archives of Medicine*. They have not yet been copy edited and/or formatted in the publication house style, and the text may be changed before the final publication.

Although accepted papers do not yet have all the accompanying bibliographic details available, they can already be cited using the year of online publication and the DOI, as follows: the author's last name and initial of the first name, article title, journal title, online first publication month and year, and the DOI; e.g.: Petrović P, Jovanović J. The title of the article. *Srp Arh Celok Lek*. Online First, February 2017.

When the final article is assigned to volumes/issues of the journal, the Article in Press version will be removed and the final version will appear in the associated published volumes/issues of the journal. The date the article was made available online first will be carried over.

† **Correspondence to:**

Radmila SPARIĆ

Clinic for Gynecology and Obstetrics, Višegradaska 26, 11000 Belgrade, Serbia

E-mail: radmila@rcub.bg.ac.rs

Large myomas as a complicating factor necessitating cesarean myomectomy followed by cesarean hysterectomy

Хистеректомија као компликација великог миома и неопходне миомектомије током царског реза

SUMMARY

Introduction Although uterine myomas are becoming more common in pregnancy due to advanced maternal age, literature lacks reports on complications, such as hysterectomy following a cesarean myomectomy (CM).

The aim of this work was describe when a CM is inevitable, complicated by severe intrapartum hemorrhage and requiring a hysterectomy.

Case report A pregnant, with a term-pregnancy and large multiple myomas, was referred for elective cesarean section (CS). During the CS, after forced enucleation of a 100mm anterior and left myoma previa (pre-fetal extraction) a 2800g neonate was delivered through the lower uterine segment (LUS) incision. After the delivery and another CM, it was necessary to stop a massive hemorrhage from the myometrial myoma bed. Following provisory suturing of hysterotomies, urgent hysterectomy was performed with left salpingoophorectomy, due to a large hematoma in the left retroperitoneal space. The patient's further recovery was uneventful and she was discharged with her baby on the 6th postoperative day. The histopathology report revealed a 135×190×150mm uterus, weighing together with the enucleated myomas and left adnexa 5000g in total. The weight of the enucleated myomas was 1670g. The histopathological examination also showed 12 intramural and subserous myomas in the myometrium, ranging from 30 to 190mm.

Conclusion Large myomas, especially previa, may present a serious problem for fetal extraction during a CS. Therefore the authors suggest an informed consent for CM, in patients who should undergo a CS. Additionally, such patients should be counseled about the possibility of an intrapartum hysterectomy.

Keywords: myoma; pregnancy; complication; delivery.

САЖЕТАК

Увод Миоми утеруса се све чешће срећу у трудноћи због година старости мајке. Мало је публикација о хистеректомији као компликацији миомектомије током царског реза.

Циљ овог рада је био да прикаже неизбежну миомектомију током царског реза, компликовану тешким интрапарталним крварењем које је захтевало хистеректомију.

Приказ болесника Трудница у терминској трудноћи, са великим мултиплим миомима је упућена за прођај елективним царским резом. После неопходне енуклеације миома превија од 100 mm лоцираног напред и лево (пре екстракције фетуса) новорођенче телесне масе 2800g је порођено кроз инцизију у пределу истмичног сегмента. После порођаја и додатне миомектомије, опсежно крварење из ложе миома у миометријуму је заустављено, али због развоја великог хематома у левом ретроперитонеалном простору урађена је хитна хистеректомија са левом салпингоофоректомијом. Даљи опоравак је протекао без компликација и жена је отпуштена са дететом шестог постоперативног дана. Хистопатолошки преглед је показао димензије материце од 135 × 190 × 150 mm, тежине 5000 g са левим аднексама и миомима. Тежина енуклеисаних миома била је 1670 g. Хистопатолошки преглед је показао и 12 интрамуралних и субсерозних миома у миометријуму, димензија од 30 до 190 mm.

Закључак Велики миоми, нарочито миоми превија, могу представљати озбиљну потешкоћу при феталној екстракцији током царског реза. Због тога је неопходан преоперативни пристанак жене за миомектомију током царског реза, али и обавештене о могућности хистеректомије.

Кључне речи: миом; трудноћа; компликација; порођај

INTRODUCTION

During the past two decades numerous studies on cesarean myomectomy (CM) have been published, mainly promoting its safety and feasibility [1, 2]. Despite myomas being the most frequent benign tumors of the female reproductive organs and the cesarean section (CS) the most common surgery in women, literature lacks reports on cesarean hysterectomy following a CM [3, 4].

In the era of favoring CM, one of the possible reasons for the lack of such reports might be the obstetricians' reluctance to publish unfavorable outcomes, even if intrapartum hysterectomy is absolutely necessary and justifiable. This could possibly further lead to the greater promotion of CM,

as an uncomplicated obstetric surgery, and its implementation even in cases when interval myomectomy would be a safer option.

The aim of the presented report is to underline the possibility of intractable hemorrhage following an inevitable CM, and the necessity of cesarean hysterectomy due to multiple large myomas.

CASE REPORT

A 36-year-old primipara was referred to the university-affiliated hospital in the 38th week of gestation for an elective CS due to myoma previa and oblique lie. Prior to her referral, the patient had been diagnosed during pregnancy with large multiple myomas (Figure 1), which were sonographically confirmed after her admission (Figure 2). The pregnancy course was uneventful, with appropriate fetal growth, development and oxygenation. Prior to a CS, the patient was informed about the possibility of an inevitable myomectomy during CS and the likelihood of massive hemorrhage after fetal extraction. Moreover, prior to the surgery, an informed consent was obtained for a CM and possible cesarean hysterectomy.



Figure 1. Multiple myomas in the uterus at seven weeks of pregnancy.



Figure 2. Large myoma during a scan at term pregnancy.

As intraoperative hemorrhage was expected, intraoperative cell salvage was implemented from skin incision and further on. During the CS, a 100 mm anterior and left subserosal myoma previa was enucleated, and a 2800g neonate was delivered through the lower uterine segment (LUS) incision used for CM. Corporal incision without myomectomy was not possible without an additional myomectomy of another myoma on the anterior wall and to the right, which was 120 mm in diameter (Figure 3). Following delivery, massive hemorrhage from the myoma previa site was encountered. Neither LUS suturing nor further hysterectomy was possible without the additional myomectomy of the anterior and right subserosal myoma, which was performed through another hysterotomy. Following provisory suturing of both incisions, a necessary hysterectomy was performed. Due to hematoma formation starting from the myomectomy site to the left into the retroperitoneal space, a contemporary left salpingoophorectomy was performed. The surgery lasted for 185 minutes, and it

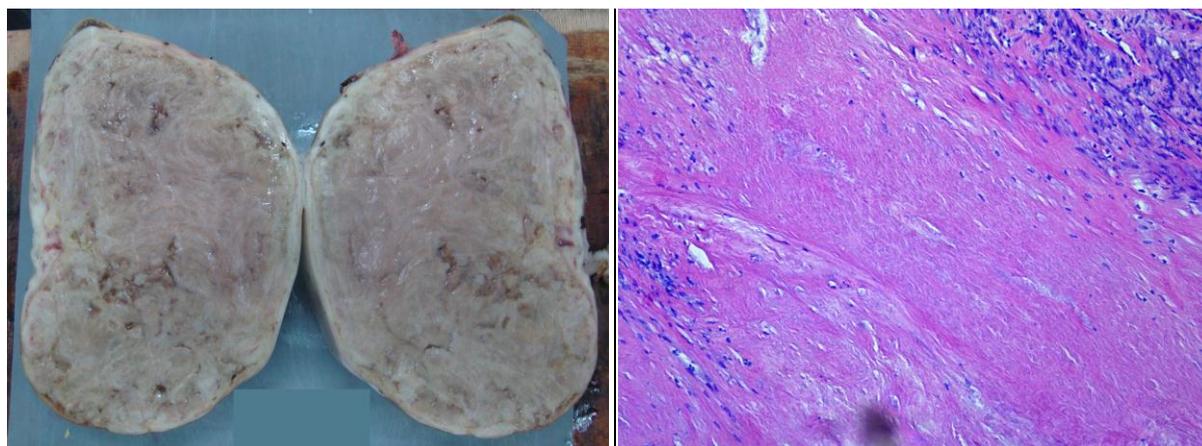


Figure 3. Macroscopic section of a myoma enucleated during a cesarean section.

Figure 4. Microscopic image of a histologic section of a fibroid with hyalinisation (H&E)

was performed by two senior surgeons. During the operation, the patient received a total of 1700 ml of autologous blood collected by cell salvage, 1325ml of heterologous blood, 1250ml of fresh frozen plasma, and 20 units of cryoprecipitate together with colloid and crystalloid solutions. After the hysterectomy, she spent two days in the obstetric intensive care unit without further blood transfusion. The patient's further recovery was uneventful, and she was discharged with her baby on the 6th postoperative day.

The histopathology report revealed a 135×190×150mm uterus, weighing together with the enucleated myomas and left adnexa 5000g in total. The weight of myomas that were enucleated was 1670g. There were also 12 intramural and subserous myomas left in the uterus with a diameter of 30 to 190 mm in range, and some of these with hyaline degeneration (Figure 4).

DISCUSSION

Although the obstetric textbooks generally consider CM to be a risky procedure, there is a consensus in modern obstetric literature about it being relatively safe, if performed by experienced obstetricians and in selected cases [1]. Most researchers agree on its benefits and advise it in cases of pedunculated and subserous myomas, anterior wall myomas, and particularly, if additional incision is not required, for myoma enucleation [5]. CM is also recommended for a safe fetal extraction when myomas compromise the hysterotomy site or hysterorrhaphy, or to reduce probable uterine torsion [5, 6]. Literature also showed that CM is inevitable in some instances, despite the risk of perioperative complications [6]. Our case supports these observations, as the delivery of the fetus was not possible either through LUS, or through corporeal uterine incision without a prior myomectomy. In such cases, with limited access to a normal myometrium, the decision to enucleate myoma previa and deliver the baby through LUS, or even corporal incision is justifiable. Nevertheless, in the presented case, another hysterotomy and a myomectomy were necessary to create a surgical space for urgent hysterectomy.

As already described in literature, myomas in the proximity of uterine vessels represent a significant risk factor for massive hemorrhage, which was confirmed in our case by the hematoma spreading into the left retroperitoneal space [5]. This also forced left adnexectomy with a hysterectomy.

Looking at literature of more than a century ago, Spencer [7] was most probably the first who indicated that total abdominal hysterectomy after a CS is the best available method to ensure the uneventful recovery of the patients affected by large myomas in pregnancy. In his report published in 1909, he underlined that "it will usually be wise to remove the uterus, especially if it is the seat of multiple tumors".

Although modern medicine nowadays provides obstetricians with a significant number of options to avoid cesarean hysterectomy in cases of a myomatous uterus, it still remains necessary in some instances to achieve definite hemostasis and to ensure the mother's uneventful recovery.

Despite the growing number of reports on CM safety and its feasibility, data on cesarean hysterectomy as a complication are quite infrequent. A recent meta-analysis on CM safety included 1082 patients with myomas, who were delivered by a CS; out of these, 443 had a CM without a hysterectomy [2]. Moreover, hysterectomy reports are present more commonly as isolated case reports, rather than in CM studies [8, 9].

This, our extensive literature search revealed a few reports on cesarean hysterectomy following CM. Li et al. [10] evaluated a total of 1438 women with myomas, who were delivered by a CS. Out of these, 51(5.3%) had a hysterectomy after a CS mainly due to multiple myomas, which were present in 84.3% of the cases. The mean diameter of the myomas was 81 ± 39 mm. The authors did not report any postoperative complications in these 59 patients. Exacoustos and Rosati [8] evaluated ultrasonographic documented myomas in relation to complications during pregnancy, at delivery and in the puerperium. Out of 492 pregnant women with myomas, nine underwent a myomectomy at delivery, of which three had had a hysterectomy due to severe hemorrhage. Another study from Ghana investigated the indications and outcomes of relaparotomy after a CS [9]. Out of a total of 44 relaparotomies, three (7%) were performed for hemorrhage following a CM. There were 4 maternal deaths in the study. Out of these, one woman had a CS for transverse lie and myomectomy. She was referred to within one hour, transfused with three units of blood and "when she appeared stable she was reopened and hysterectomy was successfully performed". The patient died 12 hours after reoperation due to disseminated intravascular coagulopathy.

Pattanaik et al. [11] analyzed the clinical outcomes of CM in 23 patients with an average age of 31.2 years, mostly primigravidas with term pregnancies. Intraoperative blood loss was over one liter in 19 (82.6%) patients, of whom one required subtotal hysterectomy. Tian et al. [12] presented their experience with a series of cervical myomas in 17 women, of whom nine had a CM. One of these women had had a myomectomy due to the inability to suture uterine incision. Because of massive intraoperative hemorrhage (about 4200ml) occurring after the myomectomy, the patient underwent a

cesarean hysterectomy. The true incidence of cesarean hysterectomy as a procedure associated with CM remains unknown. The most probable reason for this is insufficient reporting, since literature on CM is abundant and there are only a few reports on cesarean hysterectomy.

Myomas may present an obstacle for fetus delivery during a CS. Therefore, it would be appropriate to obtain an informed consent for CM from all the women undergoing a CS, in case this procedure is inevitable. Furthermore, such patients should be counseled about the necessity of an immediate hysterectomy in cases of large multiple myomas and/or uncontrollable massive intraoperative hemorrhage.

REFERENCES

1. Malvasi A, Stark M, Tinelli A. Cesarean myomectomy. In: Tinelli A, Malvasi A. Uterine myoma, myomectomy and minimally invasive treatments. Berlin: Springer; 2015. pp. 237–52.
2. Song D, Zhang W, Chames MC, Guo J. Myomectomy during cesarean delivery. *Int J Gynaecol Obstet* 2013; 121(3): 208–13.
3. Sparic R, Malvasi A, Kadija S, Babovic I, Nejkovic L, Tinelli A. Cesarean myomectomy trends and controversies: an appraisal. *J Matern Fetal Neonatal Med*. 2017; 30(9): 1114–23.
4. Tinelli A, Malvasi A, Mynbaev OA, Barbera A, Perrone E, Guido M, et al. The surgical outcome of intracapsular cesarean myomectomy. A match control study. *J Matern Fetal Neonatal Med* 2014; 27(1): 66–71.
5. Sparic R, Malvasi A, Tinelli A. Analysis of clinical, biological and obstetric factors influencing the decision to perform cesarean myomectomy. *Ginekol Pol* 2015; 86(1): 40–5.
6. Sparić R, Lazović B. Inevitable cesarean myomectomy following delivery through posterior hysterotomy in a case of uterine torsion. *Med Arch* 2013; 67(1): 75–6.
7. Spencer HR. Caesarean section and total abdominal hysterectomy for retroflexion of the uterus at term by fibromyoma adherent in Douglas's pouch. *Proc R Soc Med* 1909; 2(Obstet Gynaecol Sect): 74–80.
8. Exacoustos C, Rosati P. Ultrasound diagnosis of uterine myomas and complications in pregnancy. *Obstet Gynecol* 1993; 82(1): 97–101.
9. Seffah JD. Re-laparotomy after cesarean section. *Int H Gyn Obstet* 2005; 88: 253–7.
10. Li H, Du J, Jin L, Shi Z, Liu M. Myomectomy during cesarean section. *Acta Obstet Gynecol Scand* 2009; 88(2): 183–6.
11. Pattanaik T, Pati BK, Samal S. Cesarean myomectomy: a descriptive study of clinical outcome. *Int J Reprod Contracept Obstet Gynecol* 2014; 3(1): 172–4.
12. Tian J, Hu W. Cervical leiomyomas in pregnancy: report of 17 cases. *Aust N Z J Obstet Gynaecol* 2012; 52(3): 258–61.