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Incidence, Clinical Characteristics and Outcome of Unexpected Uterine Sarcoma after Hysterectomy for Uterine Mass: A Retrospective Study of 774 Cases

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Abstract

Background: Women often require a hysterectomy via laparotomy or laparoscopy. Morcellation is often necessary to perform a laparoscopic surgery. The objective of this study is to determine the incidence of unexpected uterine sarcomas (UUSs) after hysterectomy and to reduce the occurrence and avoid the morcellation of UUSs by analyzing their characteristics.Methods: Women who had a hysterectomy for various reasons in Chandulal Chandrakar Memorial Medical College, Durg, India between January 2014 and September 2016 were selected for the study, and their clinical characteristics were analyzed.Results: During the period, 5 UUSs were found in 774 cases, and the overall incidence was 0.65

Index terms— laparoscopy, morcellation, uterine sarcoma, uterine fibroid, hysterectomy.

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Methods: Women who had a hysterectomy for various reasons in Chandulal Chandrakar Memorial Medical College, Durg, India between January 2014 and September 2016 were selected for the study, and their clinical characteristics were analyzed.

1 I. Introduction

terine fibroids are one of the most common types of pelvic tumors in women. Various treatments are available for the management of uterine fibroids, such as having a hysterectomy or a myomectomy. The surgical route is shifting from an abdominal to a laparoscopic approach, which confers a more rapid recovery and fewer perioperative complications.

1 Morcellation of the specimen is often necessary to perform a laparoscopy.

Uterine sarcoma is a rare, malignant tumor affecting the female genital system that accounts for only 1%-3% of uterine malignancies. The 5-year survival rate is approximately 30%.

2 2,3

The degree of malignancy is high and the prognosis is poor. Inadvertent morcellation of unexpected uterine sarcomas (UUSs) is a surgical risk of laparoscopic hysterectomies and myomectomies. Evidence suggests that morcellation of UUSs potentially upstages the disease, which portends a poor prognosis.

3 4,5

The lack of specific symptoms, signs, or diagnostic techniques for preoperative differentiation from uterine fibroids results in most patients being diagnosed after surgery. If a uterine sarcoma is mistakenly diagnosed as a uterine fibroid and is morcellated via laparoscopy, serious consequences may arise.

The objective of this study is to determine the incidence of UUSs after hysterectomy for uterine fibroids. Until now, few studies have described the characteristics of UUSs in detail. This study is also designed to help reduce the occurrence and avoid the morcellation of UUSs in future by analyzing their characteristics.

4 II. Materials and Methods

5 a) Case selection

This study was approved by the Institutional Review Board at CCM Medical College, Durg, India and all subjects provided informed consent. Women who had a hysterectomy in CCM Medical College, Durg, India between January 2014 and September 2016 were selected for the study. The average patient age was 48.20±7.64 years. Among them, patients who were diagnosed with uterine sarcomas based on postoperative pathology were also selected. Their median age was 41 years. Data analysis was performed to determine the incidence of UUSs. Finally, we reviewed the patient's clinical manifestations, laboratory tests, imaging studies, intraoperative findings, surgical pathologic stage, postoperative pathology, and prognosis.

6 b) Statistical analysis

Statistical analysis was performed with SPSS 19.0 software (IBM Corporation, Armonk, NY, USA). All results are expressed as the mean ± standard deviation. The Pearson's chi-square test was used for comparison of percentages between the groups. Statistical significance was assumed at a P?0.05.

7 III. Results

8 a) Incidence of unexpected uterine sarcomas

During the period, 5 UUSs were found in 774 cases, and the overall incidence was 0.65%. Among the 5 patients, open surgery was performed on 4 patients (80%), accounting for approximately 0.01% of 625 laparotomies. There was one laparoscopy (20%), accounting for approximately 0.01% of 149 laparoscopies. There was no statistical difference (P=1.05) regarding the incidence of UUSs between laparotomies and laparoscopies.

The details of the UUSs are shown in Table **??** The results showed that 4 patients were between 40 and 49 years, comprising the highest proportion (80%) of the 5 UUSs. There was only one patient aged 23 years, accounting for 20%. Thus, it can be seen that UUSs occurred more commonly in women aged 40-49, but patients aged 21-39 and aged 50-59 cannot be ignored.

9 b) Clinical manifestations

The clinical manifestations of uterine sarcomas are nonspecific. Classically, uterine sarcomas are always present as rapidly growing pelvic masses, which may be accompanied by abnormal uterine bleeding and abdominal or pelvic pain.

10 6,7

In our study, 3 UUSs (60%) presented with abnormal uterine bleeding (menorrhagia, menostaxis, and irregular uterine bleeding), 1 UUSs (20%) presented with acute abdomen, and one UUS (20%) presented with rapidly growing pelvic masses. Abnormal uterine bleeding was the main clinical manifestation. A rapidly growing pelvic mass may be an indication of uterine sarcoma.

11 c) Laboratory tests and imaging studies involving unexpected uterine sarcomas

Of the 5 UUSs, laboratory tests for tumor markers, especially CA-125, were performed on 2 (40%). Only one case had high CA-125 values i.e. 51.2 mIU/mL, and in other it was found to be normal i.e. 23.0 mIU/mL. The sensitivity of CA-125 for uterine sarcomas is only 50% and its specificity is poor. We concluded, therefore, that CA-125 contributes minimally to the early diagnosis of uterine sarcoma.

The 5 UUS cases all underwent ultrasonic examinations, and the preoperative ultrasound diagnosis was a uterine fibroid in 4(80%) and ovarian mass in 1 (20%). The diameters of the pelvic mass ranged from 6 to 15 cm and the median value was 7 cm. The diameter of the mass in 3 cases (60%) was less than 8 cm, in one case (20%) from 8 to 10 cm, and only one case (20%) was above 10 cm. Among the 5 UUSs, only one case (20%) had irregular margins. The margins of typical uterine sarcomas are mostly nodular, irregular, or ill defined, and the uterus is significantly enlarged, but the size and margins of most UUSs prompted by ultrasonography in this study were atypical, which may cause UUSs to be misdiagnosed as uterine fibroids. Color Doppler flow imaging

of one of the 5 (20%) UUSs demonstrated rich blood flow signals, and degeneration of uterine fibroids was found in one of 5 (20%) cases. A rich blood flow signal around a pelvic mass and the degeneration of a uterine fibroid may suggest that more methods are needed to rule out the possibility of a uterine sarcoma.

In our study, 2 out of 5 UUSs (40%) were examined by computed tomography (CT). The preoperative CT diagnoses was uterine fibroid in one and ovarian mass in one. CT cannot be used to help distinguish uterine sarcomas from uterine fibroids. This study showed that it is more difficult to distinguish between benign degenerating uterine fibroids and malignant uterine sarcomas. When the pathology reports were available, the CT images were reviewed again by the pathologist, but a preoperative CT diagnosis of a uterine sarcoma still could not be made so easily. In such patients, alternative treatment options should be carefully considered. According to the postoperative pathology report after the first operation, there were three leiomyosarcomas (60%), two low-grade endometrial stromal sarcomas (ESSs, 40%). According to the FIGO (International Federation of Gynecology and Obstetrics) 2009 staging for uterine sarcomas, the number of uterine sarcomas for stages I, II, and III after the first operation was four, zero, and one, respectively, thus accounting for approximately 80%, 0%, and 20% of the five UUSs. When postoperative pathology was confirmed, one case (20%) underwent a second operation. Stage I was the first operative stage. This case underwent laparoscopy that was converted to laparotomy during the first surgery. The time interval between the two operations was 15 days.

The follow-up percentage was 60%. The followup time was 4-11 months, and the median follow-up time was 7 months. All these patients are of Stage 1 sarcoma. One case died after 9 months of diagnosis and was found after a telephonic call as she was lost to follow-up. She took 2 cycles of chemotherapy. One more case lost to follow-up and is not traceable.

12 IV. Discussion

A number of studies 10,14,15 have reported a poor prognosis for uterine sarcomas closely related to the clinical stage and the method used in the primary operation. In this study, the staging of most UUSs (80%) is stage I, which may indicate a good prognosis, but how to make an accurate diagnosis and choose an appropriate treatment is critical. To reduce the occurrence of UUSs and the risk of spreading UUSs by laparoscopic morcellation, the method used for the preoperative diagnosis of a uterine sarcoma is very important. Brohl et al found the risk of UUSs varied significantly across age groups, and the risk of uterine sarcoma ranged from a peak of 10.1 cases per 1,000 for patients aged 75-79 years to, 1 case per 500 for patients aged, 30 years. However, our study found that UUSs occurred more commonly in women aged 40-49. Abnormal uterine bleeding was the main clinical manifestation, and cases with no symptoms cannot be ignored. A rapidly growing pelvic mass may be indicative of a uterine sarcoma. The size and margins of most UUSs as suggested by ultrasonography were atypical, and UUSs can be easily misdiagnosed as uterine fibroids. Attention should be paid to a rich blood flow signal around a pelvic mass and the degeneration of uterine fibroids, and more methods are needed to rule out the possibility of uterine sarcomas. CT may have a role in the evaluation of pelvic masses. In our study, CT was of no use as it was unable to differentiate between fibroids and sarcomas.

When the pathology reports were available, the pathologist reviewed the CT images again, but it was still difficult to make a preoperative CT diagnosis of a uterine sarcoma. This told us that it is more difficult to distinguish between benign degenerating uterine fibroids and malignant uterine sarcomas. When the malignant potential of a uterine mass is uncertain, alternative treatment options should be carefully considered.

A number of studies [20][21][22][23][24][25] have shown that the incidence of UUSs ranges from 0.09% to 0.49% among women undergoing benign hysterectomy or myomectomy. In this, the incidence of UUSs during hysterectomy performed for various reasons was 0.65%, and there was no statistical difference (P=1.04) in the incidence between laparotomy and laparoscopy (0.01% vs 0.01%).

V.

13 Conclusion

In conclusion, the incidence of UUSs after hysterectomy and myomectomy was low and their clinical characteristics are atypical. It is critical to make a complete and cautious preoperative evaluation to reduce the occurrence and avoid the morcellation of UUSs. Due to the limitations of sample size, further research should be carried out to assess the impact of morcellation on the prognosis of UUSs and summarize the clinical characteristics of UUSs to avoid the risk in future.

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Figure 1:

S.no	Age (years)	Clinical Presentation	Associated Co-morbidities	Type and Stage	Treatment Received
1	49	Lump (24 weeks) Abdomen	Hypertension, Sickle Cell disease	Leiomyosarcoma 3	Abdominal Hysterectomy with With regional Lymphadenector
2	41	AUB	Nil	Leiomyosarcoma 1	
3	23	AUB Dysmenorrhoea	Anemia	Endometrial Stromal Sarcoma 1	First Surgery -Myomectomy Hysterectomy Second Surgery -W
4	40	Acute Abdomen	Nil	Carcinosarcoma 1	Radical Bilateral retroperitoneal pelvic Hysterectomy with lyn
5	45	AUB	Nil	Leiomyosarcoma 1	Total Abdominal Hysterectomy with Bilateral Salpingio-oophorectomy

Figure 2:

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