# Large cervical leiomyoma: An experience from Sudan

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## Abstract

Cervical leiomyomas - even small ones - are extremely uncommon. Two consecutive cases of very large cervical leiomyomas in young women came to our attention during the summer 2018 in Sudan. Both women presented with symptoms of heaviness/discomfort in the lower abdominal region and signs of anemia. Their management and surgical therapy are outlined.

## Introduction

Matthew Baillie is credited with the first description of uterine myomas back in 1793.[1,2] Since leiomyoma of the uterus has been identified as the most common benign neoplasm of the female reproductive organs. The incidence of symptomatic leiomyomas is 20-25% in women of reproductive age group, rising to 40% at the onset of the menopause.[3,4] This can increase to a detection rate of more than 75% for leiomyomas in general once meticulous histology studies are undertaken on hysterectomy specimen.[5]

Leiomyomas in the cervical region are considered extremely uncommon.[6]

We present two cases of very large cervical leiomyoma identified during the summer 2018 at the Lwere Cap Anamur Hospital in the Nuba Mountains of Sudan.

## Materials and Methods

The surgery is done through a lower midline incision from the umbilicus superiorly to the pubis inferiorly. Invariably the small uterus is sitting on top of the much larger polypoid cervical leiomyoma embedded in the pelvis. The appearance has been dubbed 'Lantern on top of St. Paul'.[7] Initially the subverted anatomy is analyzed to identify the pressure effects of the pelvine leiomyoma on the surrounding structures. After separating and protecting the bladder and other structures first by anatomical dissection the smallish uterus is mobilized according to a standard practice.[8]

Following the ligation of the uterine vessels and the upper part of the cardinal ligament a retrograde hysterectomy is performed, because the large size of the cervical leiomyoma prevents identification of the border between the cervix and the vagina.[9] A longitudinal incision is made from the anterior wall of the cervix/polypoid mass to the vagina to open the vaginal cavity thereby avoiding a vaginal shortening. The procedure progresses through the opening of the anterior vaginal wall, the ligation and cutting of the bilateral vesicouterine and sacrouterine ligaments respectively and finally the incision of the posterior vaginal wall. At this stage the polypoid mass attached to the small uterus can be retrieved from the pelvic cavity while rocking it back and forth and side to side. The bilateral ends of the vagina are knotted, the longitudinal incision is sutured and the vaginal cuff is closed with interrupted mattress sutures. A pelvic drain is kept for 48-72 hours postoperatively to allow for the drainage of serous fluids as the vagina vault was closed. The abdomen is closed through a continuous mass suture followed by the separate closure of the subcutaneous layer and the skin. An indwelling urinary catheter remains for 7-10 days to aid the recovery of the immobile patient with difficult access to the wash room.

## Results

### Case #1

In June 2018 a 29-year old mother of one boy (KK) came to our attention with a significant anemia (Hb 4.3 g%) and a lengthy history of abdominal discomfort and menorrhagia. A vague pelvic mass was palpable associated with a cervical mass replacing the cervical opening on speculum examination. The ultrasound examination showed a tumor of mixed echogenicity associated with a smallish uterus without adherence to the pelvic wall. Whilst consenting for surgery the patient and her partner were made aware of the possibility of the need for a potentially curative hysterectomy. At laparotomy a small uterus atop a pelvic mass was identified. In conjunction with a blood transfusion after ligation of the main vessels a retrograde hysterectomy and mobilization of the bulk of the leiomyoma was done. The postoperative recovery was uneventful, the urinary catheter was removed after ten days. Post discharge she was asked to continue her iron supplement to aid her recovery from the anemia. The histological work up of the tumor revealed findings consistent with an intra-cervical leiomyoma replacing the cervix. She is doing well 3.5 years after her surgery.

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Key words: Myoma; large cervical leiomyoma; hysterectomy.

Acknowledgements: The authors would like to thank Professor Dr R. Knuechel-Clarke Department of Pathology, University Clinics Aachen, Germany, who kindly provided the histological analysis of the specimen in case #1.

Contributions: CHH operated on the patients, conceived the report and drafted the article. JYE assisted in the operations and contributed to the pre- and post-operative management.

Conflict of interests: the authors declare no potential conflict of interests

Availability of data and materials: All data underlying the findings are fully available.

Ethics approval and consent to participate: No ethical committee approval was required for this case report by the Department, because this article does not contain any studies with human participants or animals. Informed consent was obtained from the patient included in this study.

Consent for publication: The patients gave their written consent to use their personal data for the publication of this case reports and any accompanying images.

Received for publication: 9 September 2021.
Revision received: 28 March 2021.
Accepted for publication: 8 April 2021.


*Healthcare in Low-resource Settings 2022; 10:10094*
*doi:10.4081/hls.2022.10094*



### Case #2

A 28-year old nulliparous woman (FU) complained of a longstanding history of abdominal discomfort and irregular vaginal bleeding attended the outpatient department in July 2018. She was found to have a mild

anemia. During the gynecological examination a smooth large mass was identified within the vagina having replaced the cervix but not infiltrated the sidewalls of the pelvis. The ultrasound confirmed the mixed echogenicity of this polypoid mass connected to the uterus. Her operation was planned with her and her caretaker on the understanding she might lose her reproductive ability. So far she had not conceived.

At the laparotomy the small uterus was situated between a pedunculated subserosal myoma originating from the fundus of the uterus and the large central cervical myoma occupying the pelvis (Figure 1). Following identification of the structures of the urinary tract a retrograde hysterectomy and removal of the pelvic leiomyoma was done. The postoperative recovery was smooth, the urinary catheter was kept in place for seven days. She continued her iron supplementation after her discharge form the hospital. The patient is doing fine 3.5 years after her hysterectomy.

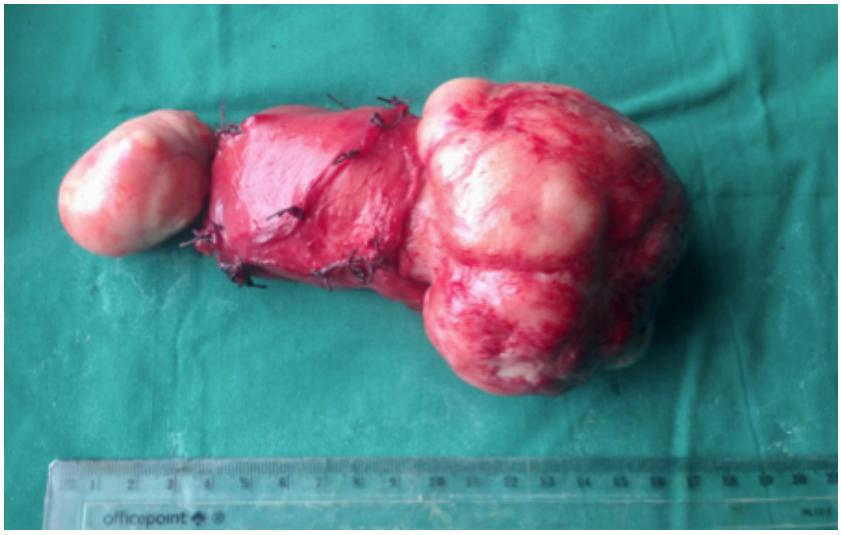

**Figure 1. Hysterectomy specimen of a 28-years old woman with a small myoma attached to the fundus of the uterus (left) and a much larger leiomyoma distal to the small uterine body replacing the cervix (right)**

## Discussion

A large - if not the only - study on cervical myomas undertaken by Tiltman on the mainly black population of South African women identified only 4 cervical myomas in 661 hysterectomy specimen (0.6%); of which two cases (0.3%) were clinically significant.[6] One was a 6 cm submucosal leiomyoma within the endocervical canal the other replaced the cervix by an interstitial leiomyoma protruding into the upper vagina.[6] In our cohort both leiomyomas originated in the central portion of the cervix replacing the cervical opening and occupying the pelvis through a circumferential expansion whereby distorting the pelvic anatomy in the process (see Figure 1). It is accepted that black women are more at risk of developing uterine myomas than caucasian women; epidemiological studies conclude the risk at 2-3 times higher in black women.[3,10,11] We have no explanation for the development of such large cervical leiomyomas in young African women.

Numerous case reports highlight the difficulties tackling a large - in some publications even called a 'giant' - cervical myoma and present the various surgical techniques adopted to achieve its removal.[4,7,12-14] Invariably these reports present postmenopausal women. [4,7,12-14] In our cohort both patients were only in their late twenties when they required surgical intervention.

Attempts are being made to preserve the reproductive function for women in the premenopausal age group through resection of a myoma arising from the wall of the cervical lip.[15,16] Others use the option of trachelectomy.[17]

Both of our patients had a complete replacement of their cervix by the leiomyoma rendering a trachelectomy not suitable.

Case reports on 'giant' cervical leiomyomas implying a uterus weight of more than 500g secondary to the tumor highlight modifications or new techniques for the hysterectomy ranging from laparoscopic hysterectomy to hybrid procedures.[12,14] Experience with uterine artery embolisation is limited to smaller and mostly extracervical leiomyomas.[18]

Whilst affluent economies have the means to initiate and experiment with new treatments like the aforementioned variations, a hysterectomy through an open procedure as first described by Keith in 1887 is the most valuable option for the low resource setting of hospitals in northeastern Africa.[19] It appears to be by far the safest option.

Having the choice between a Pfannenstiel-Kerr incision and a lower midline approach to the pelvis, we opted for the incision through the linea alba in order to minimize blood loss.[18,20] It is acknowledged that the difficulties arising through the distortion of the pelvic anatomy by the large myoma leads to a prolonged operating time.[18,20]

Some practitioner leave the vaginal vault 'open'; whilst we recognize the rationale we opted to close the vaginal opening and placed a temporary drain within the pelvic cavity in the hope to avoid an ascending pelvic infection.[20]

## Conclusions

Two women in this cohort were of reproductive age, of which one was nulliparous and the other had one child. It is disappointing for these women to lose their fertility at such an early age within a society that places a significant emphasis on having large families. There was no option - even with techniques available in affluent societies - to preserve/restore their fertility in view of the extent of the disease process. The cervical opening was occluded through the interstitial growth of the leiomyoma effectively replacing the cervix (see Figure 1).

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