

# Surgical excision of benign spindle cell neoplasm of the cervix predating miscarriage due to cervical insufficiency: A case report

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## ABSTRACT

This case report is on cervical spindle cell neoplasm and complications of its excision. A 34-year-old multiparous woman presented with a one-year history of mild to moderate non-radiating lower abdominal pain and a sensation of a mass in the vagina when urinating. These symptoms were associated with a recurrent foul-smelling yellow vaginal discharge which was unresponsive to antibiotic therapy prescribed at a primary healthcare clinic. Vaginal examination at a gynaecological clinic revealed a firm circular cervical mass. Excision biopsy of the mass (attached between the 1 and 3 o'clock positions on the cervix) showed a benign spindle cell neoplasm. The patient became pregnant six months after the surgical excision and had a miscarriage at 18 weeks of gestation due to cervical insufficiency. The report highlights the importance of antenatal surveillance for women following surgical excision of a cervical lesion such as spindle cell neoplasm as they may be predisposed to cervical insufficiency.

## 1. Introduction

Second-trimester miscarriages occur in 2–3% of clinically recognized pregnancies [1]. The causes are varied and include factors such as cervical insufficiency/incompetence. Of note, cervical insufficiency may be due to idiopathic or iatrogenic causes, with cervical surgery being a notable factor in the latter. The surgery may be performed to treat a lesion such as spindle cell neoplasm of the cervix, which is a rare tumour. Spindle cell neoplasia may be benign or malignant (details about the tumour are presented in the Discussion below). This report concerns a case of benign spindle cell neoplasm of the cervix presenting as a cervical mass, and highlights the importance of antenatal surveillance for such patients following surgical excision of the lesion as they may be predisposed to cervical insufficiency.

## 2. Case Presentation

### 2.1. Initial Encounter

A 34-year-old woman (para 2) presented at a gynaecological emergency unit with a one-year history of mild to moderate non-radiating lower abdominal pain and a sensation of a mass in the vagina when

urinating. These symptoms were associated with a recurrent foul-smelling yellow vaginal discharge which was unresponsive to antibiotic therapy prescribed as part of syndromic management of vaginal discharge syndrome (for sexually active women) [2] at a primary healthcare clinic. The symptoms did not interfere with other functions or daily activities. She had regular menstrual periods with a normal flow of three days. The patient was not on any contraceptives and had had a normal routine Papanicolaou testing (pap smear) at the primary healthcare clinic two months prior to presentation. She had had a normal vaginal delivery for the first pregnancy and an uncomplicated cesarean delivery for the second pregnancy, both at term. The indication for the cesarean delivery was unclear to the patient. She had no comorbidities and was of sober habits.

Physical examination revealed a healthy looking, stable patient with normal findings on general examination. The abdomen was soft with no tenderness nor palpable mass. Vaginal examination revealed a firm reddish circular mass which obscured the cervical os. Other system evaluations were unremarkable. Ultrasonography of the pelvis showed a normal uterus with a simple ovarian cyst 3.4 cm in diameter (follicular cyst). Following a provisional diagnosis of cervical polyp, a punch biopsy was performed. Histology showed tissue fragments covered with small collections of squamous epithelium, consisting of stroma with no

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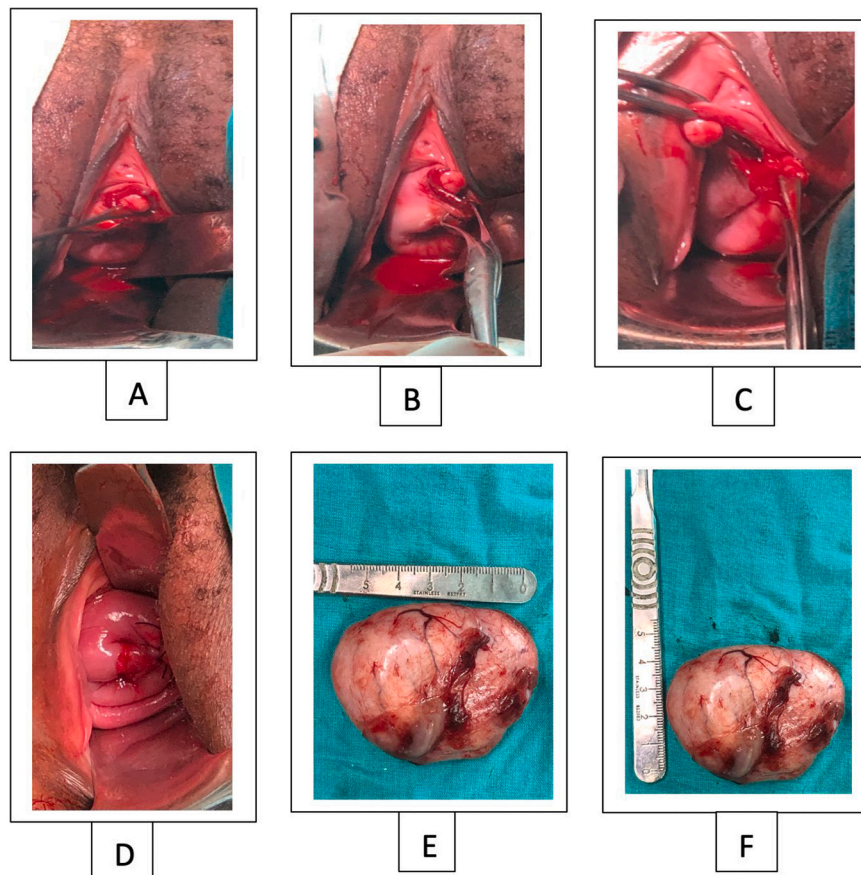
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**Fig. 1.** Excision of benign spindle cell neoplasm of the cervix (A – C), the cervix after suturing the site of excision (D), and the excised neoplasm measuring  $5.5 \times 4.5 \times 2.0$  cm in size (E and F).

endocervical glands. No morphological abnormalities were noted. Due to the inconclusiveness of the findings to aid diagnosis, the patient was counselled, underwent examination under anesthesia and excision of the cervical mass four months after initial presentation. The four-months wait (delay) was due to COVID-19 pandemic restrictions, a phenomenon which has been previously reported [3]. Intra-operatively, a circular firm mass of  $5.5 \times 4.5 \times 2.0$  cm attached between the 1 and 3 o'clock positions on the cervix was excised, and the defect sutured (Fig. 1). The cervical os was closed and no other abnormality was found. Histology of the specimen showed benign encapsulated spindle neoplasm of the cervix. There were no epithelial cell, mitotic figures nor any evidence of nuclei pleomorphism in the spindle cell lesion. Subsequent immunohistochemistry did not identify additional abnormality. There was no immediate post-operative complication, and the patient was discharged home on post-operative day 1, to be followed up in the gynecology outpatient clinic in 6 weeks' time. The patient, though, was lost to follow-up, with failed telephone attempts to contact her for post-operative examination of the cervix and counselling about the histology result.

## 2.2. Subsequent Encounter

Six months after the excision of the cervical mass, the patient presented at the gynaecological emergency unit with a history of amenorrhoea and spontaneous rupture of membranes. She had a normal body mass index, did not book for antenatal care and was gravida 3 para 2 at 18 weeks of gestation. There was no specific reason for her defaulting post-operative follow-up appointment and for not commencing antenatal care. History revealed that she had no symptoms after the excision of the cervical mass. The spontaneous drainage of amniotic fluid was associated with discomfort in the lower abdomen, but the patient had no

pain. Speculum examination of the vagina confirmed drainage of liquor and a cervix that was dilated with membranes bulging. There was no lesion on the cervix. An assessment of an inevitable miscarriage due to incompetent cervix was made, and the patient subsequently expelled a normal-looking abortus and placenta spontaneously. She had grief counselling and a normal six-week puerperal period. She agreed to booking for antenatal care and having cervical cerclage insertion in the next pregnancy.

## 3. Discussion

Spindle cell neoplasms consist mainly of spindle-shaped cells (on histopathological examination) which are longer in length than width and have tapering ends [4]. However, spindle cells can be found in normal, healthy tissues [5]. The most common normal type of spindle cell is called a fibroblast, usually found in the connective tissue stroma [4]. Spindle cell tumours may be sarcomas or carcinomas, and can also be benign [6], as in the index case. Few cases of spindle cell neoplasm of the cervix are reported in the literature [7–11], limiting the knowledge about its characteristics, and making this case report relevant. The occurrence of only spindle cells in the tumour without the presence of other cells makes this case rare. Understandably, neoplasia may be composed almost entirely of spindle cells and are called spindle cell tumours [5].

Surgical operation on the cervix, such as the excision biopsy that the patient had, may increase the risk of cervical insufficiency by altering the mechanical properties of the cervix and reducing the strength of the cervix to retain a pregnancy [12]. Other complications, which the patient did not have, that may result from cervical surgery include infection in the short term and stenosis of the cervix in the long term. If the

patient becomes pregnant again, a history-indicated cervical cerclage is recommended, given her risk factors, including the index cervical surgery and second-trimester miscarriage. This recommendation is reassuring because in the context of non-emergency cerclage insertion, a cervical suture does not increase the risk of second-trimester pregnancy loss or preterm birth [13]. Nonetheless, we are aware of a study that showed that with the use of propensity score matching, physical examination indicated cerclage is the only indication associated with a statistically significant reduction in recurrent preterm births in singleton pregnancies [14]. Certainly, using findings on physical examination (i.e. cervical dilatation) as the indication for cerclage will result in many emergency cervical cerclage insertions and expose patients to high surgical risks, unlike elective procedures. Although the authors of a meta-analysis of emergency cerclage in singleton pregnancies [15] have shown that emergency cerclage is effective and safe, they conclude that the lack of randomized controlled trials makes the quality of their evidence low to very low and that their findings should be viewed with caution [15].

There was a delay in making the diagnosis in the index case due to the cervical lesion being missed when a pap smear was performed at the primary healthcare clinic. This calls for meticulous examination of the cervix by health care professionals during such procedures. Punch biopsy performed at the gynaecological outpatient department was unhelpful due to inadequate tissue sampling.

This case also highlights the challenges associated with patient follow-up, which are often encountered in many clinical settings [16]. Despite counselling prior to hospital discharge about the possibility of the lesion being malignant, the patient failed to return for follow-up post-operative assessment and review of the histology result.

#### 4. Conclusion

Spindle cell neoplasm is a differential diagnosis of a mass in the cervix, and its excision may result in cervical insufficiency and miscarriage.

#### Contributors

Ntasako Nkanyane managed the patient during the second encounter.

Nnabuike Chibuoke Ngene managed the patient during the first and second encounter, and supervised the processes involved in completing the case report.

The authors made equal contributions to the writing of the case report.

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#### Patient consent

Obtained.

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#### Conflict of interest statement

The authors have no conflicts of interest relevant to this article.

#### References

- [1] K.M. McNamee, F. Dawood, R.G. Farquharson, Mid-trimester pregnancy loss, *Obstet. Gynecol. Clin. N. Am.* 41 (1) (2014) 87–102.
- [2] Department of Health: Republic of South Africa, Sexually Transmitted Infections: Management Guidelines. <https://health.gov.za/wp-content/uploads/2020/11/sti-guidelines-27-08-19.pdf>, 2018 (Accessed 20 May 2022).
- [3] N.C. Ngene, C.O. Onyia, C.O. Chigbu, L. Chauke, Delay in managing benign gynaecological conditions in women of reproductive age during the COVID-19 pandemic in low-and middle-income countries, *Case Rep. Womens Health* 31 (2021), e00314.
- [4] A. Marino-Enrquez, J.L. Hornick, Spindle cell tumors of adults, in: J.L. Hornick (Ed.), *Practical Soft Tissue Pathology: A Diagnostic Approach*, 2nd ed., Elsevier, Philadelphia, 2019, pp. 15–105.
- [5] MyPathologyReport.ca, Spindle Cells. <https://www.mypathologyreport.ca/spindle-cell/>, 2021 (Accessed 12 May 2022).
- [6] National Cancer Institute, NCI Dictionary of Cancer Terms. <https://www.cancer.gov/publications/dictionaries/cancer-terms/search/spindle%20cell/?searchMode=Begins>, 2022 (Accessed 12 May 2022).
- [7] S.A. Choi, M.R. Lee, M.J. Kim, H.J. Jeon, Y.S. Kim, D.H. Bae, C.J. Kim, A case of spindle cell carcinoma of the uterine cervix, Taehan Sanbuinkwa Hakhoe Chapchi (Korean Society of Obstetrics and Gynecology) 55 (4) (2012) 265–268.
- [8] S.S. Chandel, A.K. Nigam, "Spindle cell sarcoma of uterine cervix" - a rare case report, *Int. J. Sci. Res. Publ.* 4 (5) (2014) (ISSN: 2250-3153).
- [9] C.M. Zahn, B.S. Kendall, C.Y. Liang, Spindle cell lipoma of the female genital tract. A report of two cases, *J. Reprod. Med.* 46 (8) (2001) 769–772.
- [10] K. Rahimi, P.A. Shaw, R. Chetty, Solitary fibrous tumor of the uterine cervix, *Int. J. Gynecol. Pathol.* 29 (2) (2010) 189–192.
- [11] V.K. Jayaram, J. Parikshith, G.S. Narayanan, R. Tiwari, R. Veena, S. Prathima, M. S. Ganesh, C.S. Snehith, E. Prais, Multimodality management of leiomyosarcoma of the cervix, *Ecancermedalscience* 12 (2018) 830.
- [12] F.K. Lotgering, Clinical aspects of cervical insufficiency, *BMC Pregnancy Childbirth* 7 (Suppl. 1) (2007) S17.
- [13] Royal College of Obstetricians & Gynaecologists, Cervical Cerclage - Green-top Guideline No. 75. <https://www.rcog.org.uk/guidance/browse-all-guidance/green-top-guidelines/cervical-cerclage-green-top-guideline-no-75/>, February 2022 (Accessed 10 June 2022 2022).
- [14] R. Seyama, S. Makino, J. Takeda, S. Takeda, A. Itakura, The retrospective study for effectiveness of cervical cerclage in preventing recurrent preterm birth, *Taiwan J. Obstet. Gynecol.* 61 (1) (2022) 63–69.
- [15] C. Chatzakis, A. Efthymiou, A. Sotiriadis, G. Makrydimas, Emergency cerclage in singleton pregnancies with painless cervical dilatation: a meta-analysis, *Acta Obstet. Gynecol. Scand.* 99 (1) (2020) 1444–1457.
- [16] N.C. Ngene, J. Moodley, T. Naicker, The performance of pre-delivery serum concentrations of angiogenic factors in predicting postpartum antihypertensive drug therapy following abdominal delivery in severe preeclampsia and normotensive pregnancy, *PLoS One* 14 (4) (2019), e0215807.