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Case Report

Pathology Section

Cervical Adenosarcoma with Sarcomatous Overgrowth Presenting as a Gangrenous Polyp: A Rare Case Report

RESHMI ARUMUGAM[1], MANJIRI DHILIP PHANSALKAR[2], SAMPATH KARL[3], MOSES AMBROISE[4]

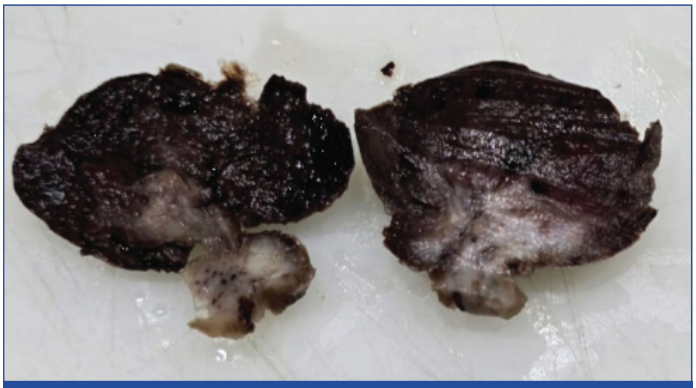
ABSTRACT

Cervical Müllerian adenosarcoma is a rare biphasic neoplasm, representing 2-10% of all Müllerian adenosarcomas and only 0.16% of cervical malignancies. It is characterised by benign epithelial glands embedded within a malignant stromal component with Sarcomatous Overgrowth (SO) signifying an aggressive variant associated with poorer outcomes. Owing to its frequent clinical resemblance to benign cervical polyps, timely diagnosis depends on careful histopathological and immunohistochemical evaluation. The authors hereby report a case involving a 39-year-old unmarried nulliparous woman who presented with one-week history of a mass per vaginum. Perspeculum examination showed a gangrenous polyp arising from the cervix. Grossly, the excised specimen consisted of a greybrown polypoidal tissue (4.0×3.5×1.8 cm) with a smooth, ulcerated and congested surface and a cut surface demonstrating a central greywhite zone surrounded by a peripheral greybrown area. Microscopy revealed a highly cellular malignant spindlecell neoplasm with marked pleomorphism, brisk mitotic activity {50/10 High-Power Fields (HPF)}, haemorrhage, necrosis and periglandular cuffing of entrapped benign endocervical or endometrialtype glands- features diagnostic of adenosarcoma with SO. Immunohistochemistry (IHC) demonstrated stromal CD10 and Cyclin D1 positivity, diffuse p16 expression, a p53 null pattern and negativity for Oestrogen Receptor (ER), Progesterone Receptor (PR), Smooth Muscle Actin (SMA), desmin and myogenin, effectively excluding other mesenchymal tumours. No residual tumour was identified on completion hysterectomy. The novelty of the present report lies in its unusual presentation as a gangrenous cervical polyp and the diagnostic challenge posed by its close mimicry of a benign lesion. The present case highlights the essential role of comprehensive histopathology and immunohistochemistry in distinguishing cervical adenosarcoma from more common benign entities.

Keywords: Cyclin D1, High grade stromal neoplasm, Müllerian tumour, Periglandular cuffing

CASE REPORT

A 39-year-old unmarried nulliparous woman who attained menarche at the age of 12 years presented with a one-week history of a mass per vaginum with no other associated symptoms such as pain, bleeding, or discharge. Per speculum examination revealed a gangrenous polyp arising from the cervix. Cervix and vagina otherwise appeared healthy. No preoperative imaging {ultrasound, Magnetic Resonance Imaging (MRI), or Computed Tomography (CT)} was performed. An excisional polypectomy of the cervical lesion was performed. Grossly, the excised specimen was a grey brown polypoidal tissue measuring 4.0×3.5×1.8 cm with attached stalk measuring 0.5 cm in diameter. Outer surface appeared smooth, ulcerated and congested. Cut surface was solid, displaying a central grey-white zone surrounded by a peripheral grey-brown area [Table/Fig-1].



[Table/Fig-1]: Gross image of excised cervical polypoidal mass.

Microscopy revealed a highly cellular neoplasm composed predominantly of malignant spindle cells arranged in both long and short intersecting fascicles. These cells exhibited pale eosinophilic cytoplasm and vesicular nuclei with notable nuclear pleomorphism and chromatin clumping, indicative of high-grade cytologic atypia. Mitotic figures were abundant with a brisk mitotic rate quantified at 50 per 10 High-Power Fields (HPF), underscoring the aggressive proliferative nature of the tumour.

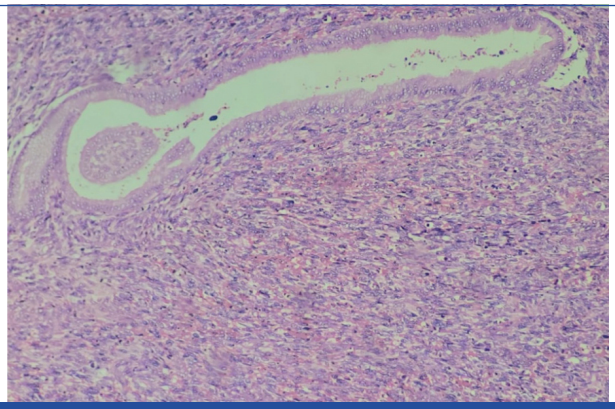
Scattered foci of tumour necrosis and areas of hemorrhage were observed, further supporting the high-grade morphology. Within the malignant stromal background, a few irregular benign endocervical glands and occasional glands resembling endometrial-type epithelium were identified. These glandular elements were surrounded by a dense cuff of malignant stromal cells- a characteristic feature known as periglandular cuffing, often seen in Müllerian adenosarcomas. However, the stalk was free of tumour.

Histology suggested a high-grade malignant spindle-cell tumour. The differential diagnosis included adenosarcoma with SO, high-grade leiomyosarcoma and high-grade endometrial stromal sarcoma. Immunohistochemical analysis was carried out to confirm Müllerian stromal differentiation and to exclude histologic mimics, using a panel that included CD10, cyclin D1, ER/PR, SMA, desmin, myogenin and p16/p53.

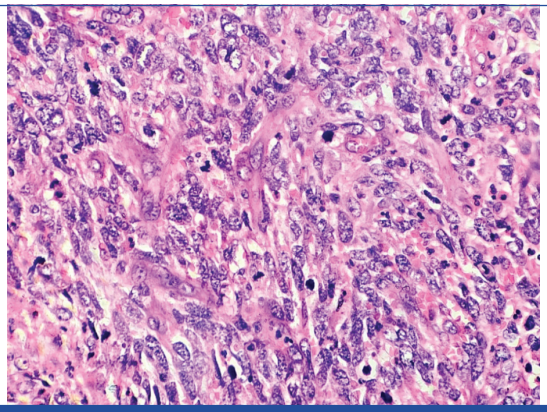
The combination of high-grade spindle cell proliferation, glandular entrapment and periglandular stromal condensation was consistent with a diagnosis of adenosarcoma with SO, a rare and aggressive variant of cervical mesenchymal tumours [Table/Fig-2a,b].

Immunohistochemical staining revealed strong positivity for CD10 and Cyclin D1 within the stromal component of the tumour, indicating active proliferation and supporting a diagnosis of high-grade stromal neoplasm [Table/Fig-3a,b]. Immunostaining for p53 revealed a complete absence of nuclear staining in tumour cells, consistent with a null mutation pattern [Table/Fig-3c]. In contrast, p16 immunostaining demonstrated strong and diffuse nuclear as

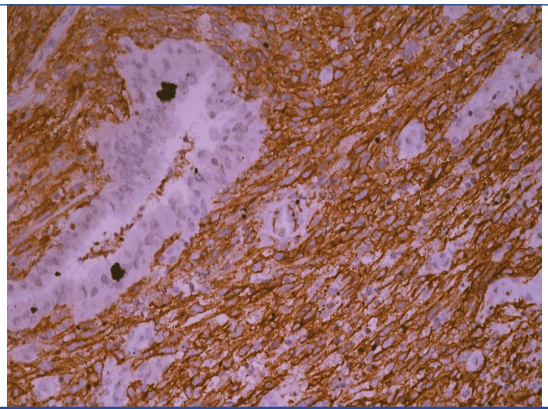
well as cytoplasmic positivity across the tumour cells [Table/Fig-3d]. Markers typically associated with smooth muscle and epithelial differentiation- including Oestrogen Receptor (ER), Progesterone Receptor (PR), Smooth Muscle Actin (SMA), desmin and myogenin- were uniformly negative. This immunoprofile effectively ruled out leiomyosarcoma, rhabdomyosarcoma and other mesenchymal or epithelial neoplasms.



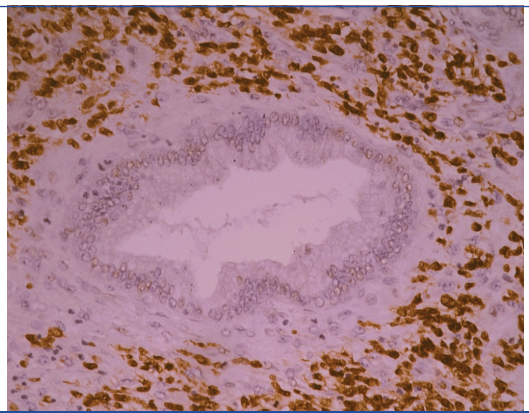
[Table/Fig-2a]: Histological section at low power illustrating benign endocervical glands, (H&E,100x).



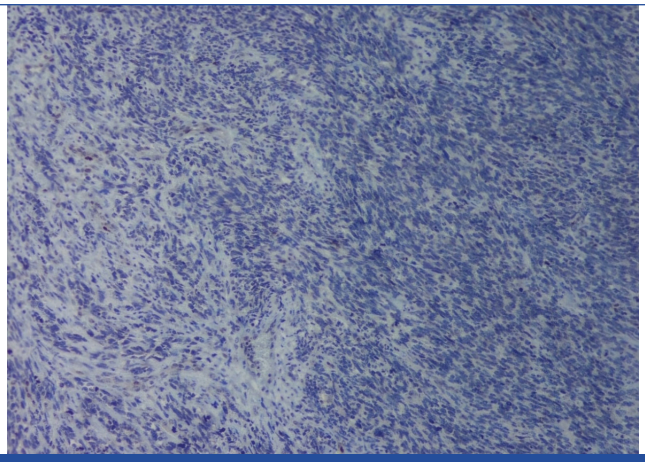
[Table/Fig-2b]: High-power view- Pleomorphic spindle cells with vesicular nuclei and brisk mitoses, (H&E, 400x).



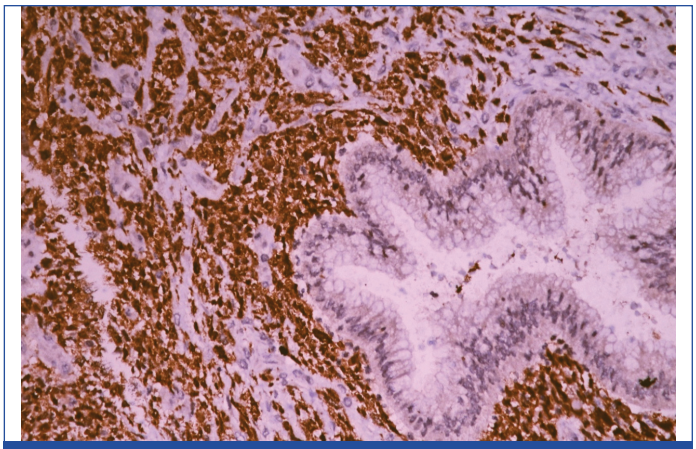
[Table/Fig-3a]: CD10 showing diffuse cytoplasmic positivity, (IHC, 400x).



[Table/Fig-3b]: Cyclin D1 showing strong nuclear positivity in stromal tumour cells, (IHC, 400x).



[Table/Fig-3c]: p53 showing null type (Mutant), (IHC, 400x).



[Table/Fig-3d]: p16 showing strong nuclear and cytoplasmic positivity, (IHC, 400x).

The histomorphology demonstrated a biphasic pattern with benign glandular elements embedded within a malignant stromal background, along with areas of SO characterised by increased cellularity, nuclear atypia and mitotic activity. Taken together, the morphological features and immunohistochemical findings were consistent with a diagnosis of cervical adenosarcoma with SO, a rare and aggressive variant of Müllerian mixed tumours. HPV testing was not performed due to logistical constraints; however, p16 and p53 immunohistochemistry were carried out on the tissue samples. Completion hysterectomy was performed later no tumour found. Adjuvant therapy was not given, as the patient was subsequently lost to follow-up.

DISCUSSION

Adenosarcoma of cervix accounts for about 2-10% of all Müllerian adenosarcoma and constitutes about 0.16% of all cervical malignancies. It is an uncommon biphasic Müllerian tumour composed of benign epithelial elements and malignant stromal components. The SO, defined as pure sarcomatous areas comprising >25% of the tumour, is a high-risk feature associated with aggressive clinical behaviour and poor prognosis [1-3].

Its clinical presentation often mimics benign cervical polyps, particularly in younger women, leading to diagnostic delays. Additional adverse features include high mitotic index, heterologous differentiation (such as rhabdomyoblastic or cartilaginous elements) and lymphovascular invasion. Immunohistochemistry plays a vital role in diagnosis with CD10 and Cyclin D1 supporting endometrial stromal differentiation, while ER, PR, SMA, desmin and myogenin help exclude other mesenchymal tumours. Rarely, cervical adenosarcoma may arise in the background of endometriosis, further complicating diagnosis due to overlapping symptoms and histologic features [4]. However, no evidence of endometriosis was

identified in the present case upon extensive histopathological examination.

Treatment typically involves surgical excision with radical hysterectomy preferred for complete resection [5]. As the patient was lost to follow-up, adjuvant therapy could not be administered. Fertility-sparing approaches may be considered in select low-grade, early-stage cases, but require careful counselling and rigorous follow-up due to recurrence risk. Imaging modalities such as MRI and Positron Emission Tomography (PET)-CT are useful for assessing myometrial invasion and metastatic spread, although findings may overlap with benign entities. Early recognition and comprehensive histopathological evaluation are essential for guiding management and improving outcomes in this diagnostically challenging entity [6,7].

Cervical Müllerian adenosarcoma must be differentiated from several benign and malignant biphasic cervical lesions. Adenofibroma lacks stromal condensation, significant atypia and elevated mitotic activity seen in adenosarcoma [6]. Embryonal rhabdomyosarcoma (sarcoma botryoides) shows a cambium layer with primitive rhabdomyoblasts and lacks benign glandular epithelium, particularly in younger patients [2,3]. Carcinosarcoma contains malignant epithelial and stromal components, unlike adenosarcoma where the epithelium remains benign [4]. Low-grade endometrial stromal sarcoma lacks phyllodes-like architecture and periglandular stromal cuffing [5]. Atypical or pseudosarcomatous cervical polyps show minimal atypia and low mitotic activity without SO [6].

Togami S et al., reported that SO and deep cervical stromal invasion is key poor prognostic factors in cervical Müllerian adenosarcoma [5]. Among the six cases reviewed one patient with SO developed distant metastases and died of disease, underscoring the aggressive nature of tumours with these features [5].

Mohapatra D et al., (2019) reported a cervical Müllerian Adenosarcoma with Sarcomatous Overgrowth (MASO) case with heterologous differentiation in a 55-year-old postmenopausal woman. The tumour showed high mitotic activity (34/10 HPF), extensive necrosis and mature and immature cartilage. IHC demonstrated diffuse vimentin and CD10 positivity and S-100 expression in cartilaginous areas, with ER and PR negativity. She underwent total hysterectomy with bilateral salpingo-oophorectomy followed by chemotherapy and radiotherapy. This case underscores the aggressive behaviour of MASO with heterologous elements and the need for multimodal treatment in high-risk patients [6].

Chen JW et al., (2024) reported a case of Müllerian adenosarcoma with sarcomatous overgrowth presenting with cervical involvement in a young woman [8]. Histology showed SO without heterologous elements and IHC demonstrated stromal vimentin and CD10 positivity with ER/PR negativity. HPV testing was negative, supporting that cervical MASO is not HPV-related [2,3]. Despite high-risk features, the patient chose fertility preservation and declined adjuvant therapy, remaining disease-free on short-term follow-up, emphasising individualised management and close surveillance in young patients [8,9].

Cervical Müllerian adenosarcoma generally carries a better prognosis than uterine corpus adenosarcoma, particularly when confined to the cervix and lacking high-risk histologic features [4,5]. However, SO, heterologous elements, deep stromal invasion and a high mitotic index are well-established poor prognostic factors and are associated with aggressive clinical behaviour [5,7]. Several reports describe rapid progression in tumours with SO, even in young patients, indicating that age does not guarantee a favourable outcome [3,9].

The recurrence rate for cervical adenosarcoma is approximately 15-30%, largely influenced by histopathological features [2,5]. Recurrence is most often pelvic or vaginal, though distant metastases can occur, particularly in tumours with SO or heterologous differentiation [6]. Incomplete excision and extra-cervical spread further increase recurrence risk [2,5]. Long-term surveillance is essential, as late recurrences have been reported several years after initial treatment [5,10].

As summarised in [Table/Fig-4], cervical MASO is a rare but clinically significant tumour occurring across a broad age range (15-72 years) and often presenting as a polypoid cervical mass that mimics a benign lesion [1-4,6-10]. Prognosis is strongly influenced by SO and heterologous differentiation. Radical surgery remains the cornerstone of management, while fertility-sparing approaches may be cautiously considered in select cases. Outcomes are heterogeneous- ranging from long-term remission to early recurrence or death- underscoring the importance of meticulous histopathological assessment and vigilant follow-up, particularly in patients with high-risk features or

Author/Year of study	Age (in years)	Invasion (myometrial)	Heterologous element	Immunohistochemistry	LN/LVI	Surgery	Adjuvant	Outcome
Duggal R et al., 2010 [1]	15	No	Yes: Chondrosarcoma, Myxoid liposarcoma, Leiomyosarcoma, Rhabdomyosarcoma	Vimentin +/++; CD10 + (low-grade); S-100 + in liposarcomatous areas In high grade areas:Desmin+,Vimentin +	NA	TAH + BSO + omentectomy	Chemo + RT	Recurrence at 1 year; DOD
Seagle BLL et al., 2014 (Case 1: SO) [2]	54	No	No	SMA,MSA,ER, PR:+ Desmin,CKAE1/3,CAM 5.2: Focally + Myogenin, EMA,CD10,Caldesmon, HMB- 45, CD31, CD34, Inhibin, S100: -	LN dissection: Negative for malignancy	Radical Hysterectomy + BSO and B/L Pelvic and para-aortic lymph node dissection	Vaginal cuff brachytherapy and chemotherapy	NED 66 months after diagnosis
Seagle BLL et al., 2014 (Case 2: heterologous, no SO) [2]	47	NA	Yes (rhabdomyoblastic)	NA	NA	Cervical biopsy	Preoperative whole pelvis EBR and chemo	DOD at 12 months after diagnosis
Morales DA et al., 2016 [3]	39	No	Yes Rhabdomyoblastic Chondrosarcoma	CD 10: Focal + S100:+ (Cartilaginous) Myogenin:+ (Rhabdomyoblastic) Ki 67 – 60%	LVI - Present	Radical hysterectomy +PNSR	RT + chemo	AWD 21 months
Atanda AT et al., 2017 [4]	18	NA	Yes Chondrosarcoma	Desmin:+(Spindlecells) ER:+ (Glandular)	NA	Local excision (Cervix)	Nil	Lost to follow-up
Mohapatra D et al., 2019 [6]	55	No	Yes: mature/ immature cartilage with few osseous areas	Vimentin +; CD10 +; S-100 + in cartilage; ER/ PR negative	NA	TAH + BSO	Chemotherapy + RT	NED at 3 months
Park HM et al., 2023 [7]	38	Focal invasion	No	Not done	Nil	TAH+BSO+PLD	Chemo	NED at 15 months

Qing X et al., 2024 [8]	56	Yes	No	ER (+), PR (+), CD10 (+/–), SMA (–), Desmin (–), P53 (–) and Ki-67 (+; hot spot area of approximately 5%)	Nil	Lap Hysterectomy +Bilateral adnexectomy	Nil	NED at 1 month
Chen JW et al., 2024 [9]	20	NA	No	Vimentin +; CD10 -; ER/PR –; HPV –	NA	Fertility-sparing excision	Nil	NED at 6 months
Bruguier H et al., 2024 (on endometriosis) [10]	41	Nil	No	ER,PR:+	Nil	Hysteroscopy + removal of endocervical polyp + lap excision of endometriosis Followed by Lap Hysterectomy +BSO	Nil	Referred to oncology specialist
Present case	39	Nil	No	CD10 +; Cyclin D1 +; p16 strong; p53 null; ER/PR/SMA/Desmin –	NA	Excision of cervical polyp Followed by TAH+BSO	Nil	Lost to follow-up

[Table/Fig-4]: Summary of MASO cases reported in English literature [1-4,6-10].
TAH: Total abdominal hysterectomy; BSO: Bilateral salpingo-oophorectomy; PNSR: Pelvic lymph node removal; PLD: Pelvic lymph node dissection; LN: Lymph node; LVI: Lymphovascular invasion; SMA: Smooth muscle actin; ER: Oestrogen; PR: Progesterone; EMA: Epithelial membrane antigen; HMB 45: Human melanoma black; RT: Radiotherapy; AWD: Alive with disease; NED: No evidence of disease; DOD: Dead of disease; NA: Not applicable

those declining adjuvant therapy. These cases provide valuable insights into the evolving understanding and clinical approach to this uncommon entity.

CONCLUSION(S)

Cervical adenosarcoma with SO is a rare but aggressive neoplasm that can clinically mimic benign cervical polyps. The present case underscores the importance of thorough histopathological assessment, where features such as periglandular cuffing, high mitotic activity and necrosis raise suspicion for malignancy. Immunohistochemistry- particularly CD10 and Cyclin D1 positivity- was instrumental in confirming endometrial stromal differentiation and excluding other mimics. Early recognition and accurate diagnosis are critical for guiding appropriate treatment and improving patient outcomes.

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PARTICULARS OF CONTRIBUTORS:
1. Postgraduate Student, Department of Pathology, Pondicherry Institute of Medical Sciences, Puducherry, India.
2. Professor, Department of Pathology, Pondicherry Institute of Medical Sciences, Puducherry, India.
3. Surgeon, Department of Surgery, St. Thomas Hospital, Polur, Chettupattu, Tamil Nadu, India.
4. Professor and Head, Department of Pathology, Pondicherry Institute of Medical Sciences, Puducherry, India.

NAME, ADDRESS, E-MAIL ID OF THE CORRESPONDING AUTHOR:
Dr. Reshmi Arumugam,
Room No. 510, 'S' Block, OPD Building, 4th Floor Pondicherry Institute of Medical Sciences, Ganapathichettikulam-605014, Puducherry, India.
E-mail: areshmi1998@gmail.com

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