

Case Report: Adenocarcinoma Of The Appendix Presenting As A Cervical Polyp.

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1 Title page

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24

Abstract

25

Background: The differential diagnosis of a vaginal mass after hysterectomy is broad. Malignancies of the genital tract or metastasis from another primary tumor should be excluded. Appendiceal carcinoma is a rare cause, case reports are limited.

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Case presentation: A 77-year-old woman with a history of vaginal hysterectomy presented with a 12-month history of vaginal pressure. The vaginal mass, penetrating through the cervix, is resected (vaginal approach). A Microscopic examination of the removed tissue revealed an adenocarcinoma of the appendix. Right hemicolectomy, resection of the vaginal vault and resection of the regional lymph nodes showed no residual tumor.

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Conclusions: Full diagnostic work-up of a vaginal mass should be performed to avoid unexpected malignancy. Transvaginal resection was in this case feasible and safe.

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Keywords

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Adenocarcinoma, mucinous; Intestinal type; Vaginal neoplasms; Appendiceal cancer, Transvaginal appendectomy.

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Background

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A 77-year-old Caucasian woman is referred to our hospital with a 12-month history of vaginal pressure. In 1990, she underwent a supracervical hysterectomy because of excessive bleeding due to uterine myoma's. She has a sensation of vaginal pressure, progressively increasing over time, combined with vaginal discharge and blood loss. She has also urge incontinence and urinary urgency.

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On physical examination, there was no abdominal mass palpable. On vaginal examination, friable mass, coming out of the cervix is seen. A modest cystocele (first or second degree) is also diagnosed.

46

47 On rectal examination, no mass is felt. Transvaginal ultrasound shows the transcervical mass of 15 x
48 10 mm, no ascites is seen. Normal aspect of the bladder.

49

50 Recent cytology of the cervix showed abnormal cells, suggestive for an endometrial adenocarcinoma.

51 Blood examination shows a subtle decrease of her kidney function (estimated glomerular filtration
52 rate of 61 ml/min/1.73m², normal >90 ml/min/1.73m²). Carcinoembryonic antigen (CEA) is raised at

53 8.2 µg/L (normal 2-4 µg/L). The value is raised since 2009, excessive examinations (gastroscopy,
54 colonoscopy, ileoscopy, chest X-ray, computed tomography (CT) scan, thyroid ultrasound and
55 mammogram) couldn't withhold a source. No biopsy of the mass is taken. A cervical polyp is
56 suggested as the reason of her complains and she is scheduled for polypectomy.

57

58 Perioperative dissection shows epiploic appendages of the sigmoid, so a cervical polyp is unlikely.

59 The mass is removed in total and the rectal mucosa, muscularis and serosa is closed with separate
60 stitches. Microscopic examination of the tissue shows an intestinal type mucinous adenocarcinoma
61 of colorectal origin.

62

63 Postoperative laboratory findings show a normal cancer antigen 125 and cancer antigen 19.9,
64 suggesting a gastrointestinal origin rather than an urogenital origin. No metastasis is seen on
65 postoperative exams (magnetic resonance imaging of the pelvis and rectum (MRI), CT scan of the
66 thorax and abdomen). The CT scan shows a process in between the caecum and vaginal vault,
67 suggestive for an appendicular process with vaginal breakthrough. Colonoscopy shows no
68 abnormality beside diverticulosis. A pT4bN0M0 stage IIc appendiceal carcinoma is diagnosed. She
69 undergoes a laparoscopic right hemicolectomy with resection of the vaginal vault. Conversion to
70 laparotomy due to adhesions of the small intestines occurred.

71

72 Microscopic examination of the tissue showed no residual tumor (R0 resection). Zero out of 14
73 dissected regional lymph glands show metastasis.

74

75 At the time of writing this report, 2 years after diagnoses, CT thorax/abdomen and PET scan didn't
76 show recurrence.

77

78 Methods

79 This case report gives an overview of the differential diagnosis of vaginal bleeding of total
80 hysterectomy and it's anatomopathological findings. It's the first case report of a pT4bN0M0
81 appendiceal tumor with complete transvaginal resection. The patient is followed in a secondary
82 health care center, diagnosis and treatment November 2018.

83

84 Results

85 Similar case reports of appendiceal cancer penetrating the vaginal vault, are rare [1,2]. The
86 differential diagnosis of a mucinous adenocarcinoma in the vagina are endometrial cancer,
87 carcinomas of a woman's genital tract (vagina, cervix, fallopian tubes or ovaries) or metastases from
88 other tumors or intestinal tumor with infiltration in the vagina [3]. Vaginal vault recurrences of
89 genital tract malignancy are not uncommon and may be the first sign or recurrent disease [4].

90

91 Primary mucinous vaginal adenocarcinoma of intestinal-type is an extremely rare malignancy of
92 unknown histogenesis with a diagnostic dilemma for the clinician and histopathologist. Extensive
93 investigation should be performed to exclude a primary adenocarcinoma of another location such as
94 the rectum, colon, breast, ovary, uterus or cervix [5]. CT scan, gastroscopy, colonoscopy,
95 proctosigmoidoscopy, positron emission tomography (PET), MRI, cystoscopy and serum CEA level

96 should be executed [6]. Subsequent immunohistochemical stains of the biopsied lesion, if available,
97 is essential in this regard [7].

98

99 Primary vaginal cancer is rare, comprising approximately 1 to 2% of all gynecologic malignancies
100 [8,9]. Extensions from cancer of the cervix or vulva or metastatic cancer are seen more frequently
101 than primary vaginal cancers [8]. The majority of primary vaginal cancer is squamous cell in origin,
102 followed by adenocarcinoma, sarcoma, melanoma, and various other histological types [10].

103

104 Subtypes of vaginal adenocarcinomas are clear cell, endometrioid, serous and mucinous carcinomas.
105 The most common variant is the clear-cell adenocarcinoma, which can occur spontaneously and in
106 women with in utero exposure to diethylstilbestrol (DES) [6]. It was prescribed from 1938 to 1971. It
107 can also cause clear cell adenocarcinoma cervix, structural malformities of uterus and decreased
108 fertility [11].

109

110 Mucinous adenocarcinomas of the vagina are extremely rare and can be further sub-classified as
111 endocervical and intestinal types [6]. The histogenesis of vaginal adenocarcinoma of the mucinous
112 type remains unclear [12]. Hypothesized origins of the lesions included derivation from congenital
113 rests of the intestinal epithelium, neoplastic transformation from another cell type directly to into
114 intestinal type and or even dysplastic enteric epithelium secondary to surgical manipulations [13].
115 During repair of the rectovaginal septum after childbirth, rectal mucosa can be incorporated into the
116 repair and involute toward the vaginal mucosa. Appropriate repair is critical and may play a role in
117 cancer development. [14]

118

119 Primary adenocarcinoma of the rectovaginal septum is another extremely rare clinical entity that
120 arises in most of the cases from endometriosis. Only 9 case reports are published. Diagnosis was
121 always made during surgery by pathologic exam based on immunohistology findings. Positive

122 staining for OC 125, an antibody that recognizes epithelium of coelomic origin such as Müllerian
123 epithelium [13].

124

125 Appendiceal carcinoma is also very rare. It accounts for less than 0.5% of all intestinal tumors. Mostly
126 appendiceal carcinomas present with acute right lower abdominal pain suggestive of appendicitis
127 [15]. Appendiceal carcinoma can also present as a palpable abdominal mass, acute intestinal
128 obstruction or ascites [16].

129 Discussion

130 A full workup is required when vaginal mass is diagnosed. Raised CEA was suspicious, biopsies should
131 be taken to differentiate a gastrointestinal origin for the tumor. When adenocarcinoma of the vagina
132 is diagnosed, a primary tumor of the colon should be excluded [17]. Primary intestinal-type glandular
133 lesions of the vagina are rare. They are a challenge for the histopathologists as the tumor
134 morphology together with the immunocytochemical stains of the tumor resembles closely that of a
135 gastrointestinal adenocarcinoma [5]. Our patient's previous hysterectomy probably allowed the tip
136 of the appendix to move near to the vaginal vault thus causing infiltration.

137

138 It was a challenge to remove the tumor in total trough the vagina. Robotic or laparoscopic resection
139 of sigmoid or gastro-intestinal cancer with transvaginal extraction is described [18,19]. It is feasible
140 and safe in a selected group of patients [18]. Natural orifice transluminal endoscopic surgery (NOTES)
141 is a new approach that allows for minimal invasive surgery through the natural orifices such as the
142 vagina. No special equipment such as long trocars or flexible endoscopes are needed for a
143 transvaginal appendectomy. A trend towards shorter hospital stays, quicker recovery and less
144 analgesic requirement is seen in comparison to conventional laparoscopic appendectomies [20].

145

146 Conclusions

147 Appendiceal cancer can presents itself in various ways. This case describes an atypical presentation
148 of appendiceal cancer through the cervix. Diagnostic work-up is needed to differentiate with other
149 pathologies (biopsy and immunohistology, CT scan and MRIA, CEA and CA 125). Vaginal resection can
150 be safe and feasible. Literature concerning vaginal approach for appendectomy is growing.

151 List of abbreviations

152	CA125	Cancer antigen 125
153	CA19.9	Cancer antigen 19.9
154	CEA	Carcinoembryonic antigen
155	CT	Computed tomography
156	DES	Diethylstilbestrol
157	NOTES	Natural orifice transluminal endoscopic surgery
158	MRI	Magnetic resonance imaging
159	PET	Positron emission tomography

160

161 Declarations

- 162 • Ethics approval

163 Not applicable

- 164 • Consent for publication

165 Written informed consent was obtained from the patient for publication of this case report. A copy
166 of the written consent is available for review by the Editor-in-Chief of this journal.

- 167 • Availability of data and materials

168 Data sharing is not applicable to this article as no datasets were generated or analyzed during the
169 current study.

170

- Competing interests

171 The authors declare that they have no competing interests" in this section.

172

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174

- Authors' contributions

175 BP performed the total vaginal resection of appendiceal carcinoma and NS assisted during the
176 operation. KV performed right hemicolectomy with resection of the vaginal vault. EDS analyzed and
177 interpreted the patient data and wrote a literature review. EDS was a major contributor in writing
178 the manuscript. All authors read and approved the final manuscript.

179

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181

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