Adenoma Malignum Detected on a Trauma CT

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ABSTRACT

Adenoma malignum is a rare subtype of cervical adenocarcinoma. Clinical presentation is variable with watery vaginal discharge being the most commonly associated finding. We report a case of adenoma malignum incidentally detected on pelvic computed tomography (CT) performed for a trauma patient. The cervical mass was further characterized by magnetic resonance (MR) imaging and remained compatible with adenoma malignum. Local cervical biopsy was suggestive of the diagnosis which was subsequently confirmed by gross surgical pathology. We briefly discuss adenoma malignum with particular attention to CT and MR imaging features.

CASE REPORT

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A 42 year-old woman was transferred to our Emergency Department after initially receiving primary trauma care at a peripheral hospital following a high speed motor vehicle accident.

Physical examination at the time of presentation revealed normal vital signs. The patient demonstrated tenderness in the lower chest, the left hand, and the right knee. The physical and laboratory assessments were otherwise negative. Past medical and surgical histories were non-contributory. A contrast enhanced CT of the abdomen and pelvis failed to reveal any traumatic abdominal or pelvic injuries, but it did demonstrate a complex, multiloculated cystic mass centered in the uterine cervix (Figure 1).

A pelvic MR confirmed the presence of a multiloculated cystic cervical mass. No enhancing septations or nodules were identified within the individual cysts. The cervical stroma was thinned but intact and there was no evidence of parametrial, vaginal or uterine corpus extension. Intervening

cyst septations had signal characteristics of normal cervical stroma (Figures 2, 3).

The right ovary contained a large complex mass with peripheral regions of increased T1 signal which demonstrated T2 shading; suspicious for a hemorrhagic cyst. More centrally, the right ovarian mass was more complex with heterogeneous T1 and T2 signal. The left ovary contained multiple small hemorrhagic cysts demonstrating increased T1 signal, T2 shading and fluid-fluid levels. Incidentally, these were found to represent bilateral endometriomas at pathology (Figure 4).

Loop electrosurgical excision procedure (LEEP) was performed. Biopsied tissue demonstrated mucinous metaplastic change of the endocervical epithelium with some areas of papillary architecture and focal cytologic atypia. The overall appearance was considered highly suspicious for adenoma malignum and a total abdominal hysterectomy with bilateral salpingo-oophorectomy was performed. Final surgical pathology confirmed the diagnosis of adenoma malignum (Figures 5 and 6). While the lesion expanded the cervix and abnormal tissue was present within 3mm of the

lateral cervical margins; it appeared confined to the cervix with no pathological evidence of parametrial or lymph node disease.

The patient was treated with total abdominal hysterectomy and bilateral inguinal lymph node dissection. Postoperative course was complicated by bilateral hydronephrosis requiring percutaneous nephrostomy tubes. To our knowledge the patient is currently doing well, approximately 4 years from her initial diagnosis.

DISCUSSION

Adenoma malignum is a rare subtype of adenocarcinoma of the cervix, constituting approximately 1% of cervical adenocarcinomas [1]. Owing to its well differentiated pathology the lesion is sometimes referred to as minimal deviation adenocarcinoma [2]. Clinically, patients may present with menometrorrhagia, mucoid or watery vaginal discharge, abdominal pain, postmenopausal bleeding, or may be asymptomatic [1, 3]. Average age of diagnosis is 42 years, with a range of 25 to 72 years [2]. Prognosis has been variably reported, but is generally accepted as poor [2]. A large series of 22 patients with a two year follow up found that only 3 patients remained disease free and 13 died of recurrent cervical malignancy [3]. There is an association between adenoma malignum and Peutz-Jeghers syndrome and mucinous ovarian tumors [2].

Findings on CT are somewhat nonspecific; often consisting of a poorly characterized cystic mass [4]. MR findings suggestive of adenoma malignum include a complex multicystic lesion, often with solid components located within the deep cervical stroma (4-7). While individual cysts may be small in size; the overall caliber of the cystic lesions is typically large, measuring greater than 20mm in axial spread and extending greater than 5mm into the cervical stroma (4). The presence of the typical multicystic lesion is not mandatory for the MR diagnosis of adenoma malignum. One series found that MR images of five of their ten patients demonstrated, "indistinct" "hazy" or "villous" regions of increased T2 signal intensity within the cervix in the absence of any cystic lesions (4). The T1 post-gadolinium enhancement pattern for these lesions is variable, as septal enhancement has been reported as both present (5) and absent (4, 7).

The differential diagnosis of a cystic mass located in the cervical stroma, with the imaging findings presented, includes adenoma malignum, deep nabothian cysts, and well differentiated adenocarcinoma (5-7). Other causes of cervical cysts include adenomatous glandular hyperplasia and mesonephric duct remnants (5, 8).

Deep nabothian cysts are benign cystic dilatations of endocervical glands found deep within the cervical stroma (9). Nabothian cysts are benign and are a common incidental finding in the superficial stroma (6). They are of primary concern when found deep in the stroma due to their close appearance to adenoma malignum on imaging studies. These benign cysts are typically small, lack complex portions, have

well circumscribed margins, exhibit T1 and T2 hyperintensity on MRI, and do not enhance with administration of gadolinium contrast (5, 7, 10, 11).

Adenomatous hyperplasia of the endocervix is differentiated from adenoma malignum based on location within the cervix. Adenomatous hyperplasia does not extend into the deep cervical stroma (5).

Well differentiated adenocarcinoma may exhibit similar imaging characteristics to adenoma malignum, as adenoma malignum is a well differentiated mucin producing adenocarcinoma (2, 6). In order for a cervical neoplasm to be considered adenoma malignum it should contain less than 10% of ordinary adenocarcinoma (12). Deciding where on the spectrum a lesion resides, is best determined by pathology.

Mesonephric remnants may also produce cystic lesions in the cervix, unlike adenoma malignum however, these lesions are typically solitary and found within the lateral wall midway through the cervix (8).

In our patient CT and MR imaging revealed a multilocular cystic mass involving the deep cervical stroma. The septae enhanced equally to the surrounding cervical stroma. In the absence of solid enhancing components, it may be difficult to differentiate multiple nabothian cysts from adenoma malignum (5). When the diagnosis is unclear, biopsy is warranted. The multiplicity of the cysts, extent of involvement of the deep stroma, and the overall large cyst size in our patient prompted the suggestion of adenoma malignum.

Given the high T1 signal within the ovarian lesions and the known association between adenoma malignum and mucinous adenocarcinoma of the ovary, the possibility of coincidental mucinous tumours of the ovaries was suggested. The ovarian lesions shown on imaging ultimately were found to represent endometriomas at final pathology.

TEACHING POINT

Adenoma malignum is a rare well differentiated adenocarcinoma of the cervix which can mimic benign deep Nabothian cysts. The typical imaging feature of this tumor type is of a large, multicystic mass in the deep cervical stroma which may or may not contain solid enhancing components.

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FIGURES



Figure 1: 42 year old female with adenoma malignum. Axial contrast enhanced CT of the pelvis demonstrates the multiloculated cystic mass in the uterine cervix (white arrow). (Images obtained on a 16 slice GE Light Speed Ultra scanner at 120kVp, 72 mAs, 5 mm slice thickness, 100 mL IV Optiray 320 and 8ml of Telebrix 38 Oral in 450mL of water.)

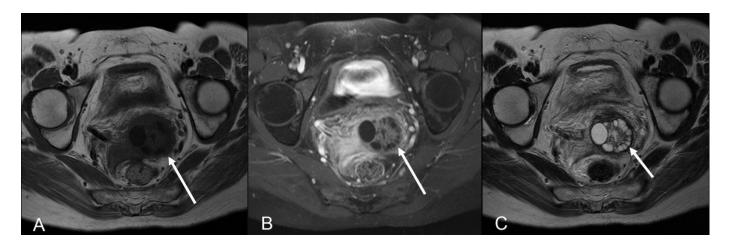


Figure 2: Axial MR images of a 42 year old female with adenoma malignum. All MR images acquired on a 1.5 T Siemens Symphony after 20mg IV buscopan and vaginal gel. Contrast enhancement with 15mL IV Optimark. (a) T1 (TE/TR; 12/604 msec): Expansion of the uterine cervix by a multiloculated cystic mass (white arrow). Cystic components are homogeneously low in signal intensity. (b) Post contrast T1 with fat saturation (12/590 msec): Enhancement of intervening septations is equal to that of the normal cervical stroma; no internal enhancing components to the individual cysts. (c) Turbo spin echo T2 (97/4000 msec): Multiple sharply defined round T2 hyper intense cysts expand the cervix. The mass is confined to the cervix with an intact ring of surrounding cervical stroma and no evidence of parametrial invasion.

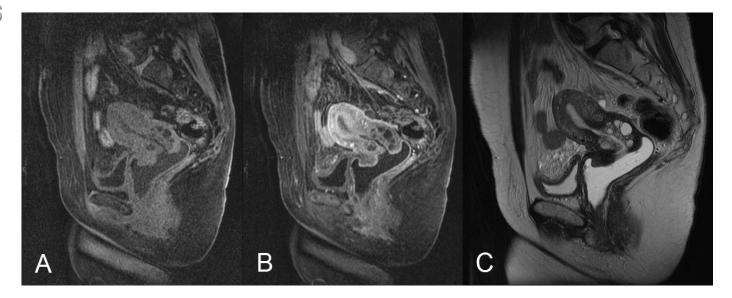


Figure 3: Sagittal MR images of a 42 year old female with adenoma malignum. (a) Unenhanced 3D spoiled gradient echo T1 weighted image (2.20/4.45 msec) with fat saturation demonstrates the low internal T1 signal within the multiple cysts in the deep cervical stroma. (b) After gadolinium injection there is no internal enhancement within the cysts; intervening septations follow similar enhancement to the normal cervical stroma. (c) T2 weighted image (97/4000 msec), demonstrates T2 hyperintensity within the individual cysts of the cervical mass.

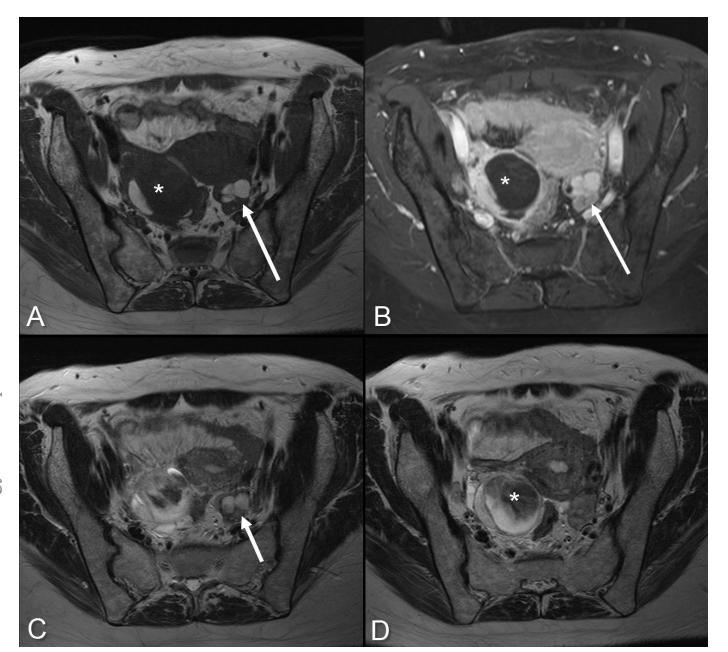


Figure 4: Axial MR images of a 42 year old female with adenoma malginum. Images are at the level of the ovaries. (a) Unenhanced T1 spin echo (12/604 msec) (b) Post contrast T1 with fat saturation (12/590 msec) (c,d) T2 spin echo (97/4000 msec). The right ovary (*) contains a large complex mass with peripheral regions of increased T1 signal which demonstrate T2 shading; suspicious for a hemorrhagic cyst. More centrally, the right ovarian mass is more complex with heterogeneous T1 and T2 signal. There is no internal enhancement within the right ovarian mass. The left ovary (white arrow) contains several small cysts demonstrating high T1 signal and T2 shading; compatible with small hemorrhagic cysts. Both ovaries were found to contain endometriomas at pathology.

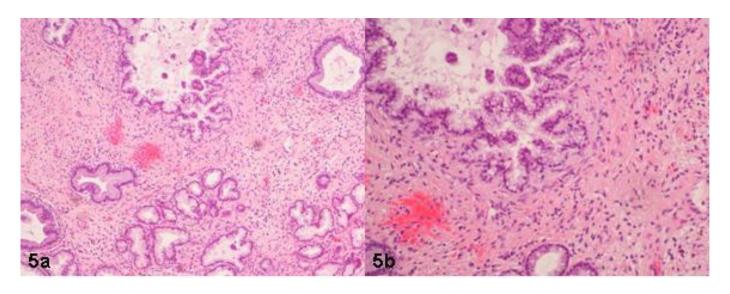


Figure 5: (a) Histological section shows endocervical gland proliferation. Epithelial cells are columnar and surrounded by a single layer of myoepithelial cells. No mitotic figures or squamous metaplasia present. There is some papillary protrusion present in the gland on the top half of picture. (20X) (b) High power image demonstrates better the papillary protrusion. (40X) No cellular atypia is noted. (Hematoxylin and eosin staining)

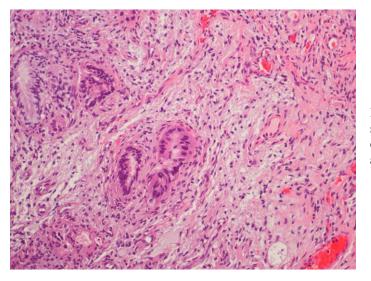


Figure 6: Histologic section of the cervix demonstrates a single focus of cellular atypia with hyperchromatic and elongated nuclei. Minimal desmoplastic reaction is found around this gland. (Hematoxylin and eosin staining, 40X)

Incidence	Rare; approximately 1% of cervical adenocarcinomas	
Etiology	Unknown	
Gender ratio	Female	
Age Predilection	25-72 years (average 42 years)	
Risk Factors	Peutz-Jeghers syndrome	
Treatment	Usually surgical	
Prognosis	Generally considered poor; better with earlier stage	
Findings on Imaging	Large multiloculated cystic mass expanding the cervix; may have enhancing solid components	

Table 1: Summary table for Adenoma Malignum

Pathology	CT	MRI
	expanding the uterine cervix.	Large multiloculated cystic mass extending into the deep cervical stroma. May lack typical cystic appearance; instead appearing as "hazy" or "villous" regions of increased T2 signal. Septations may or may not enhance post-gadolinium.
-	cervix.	Small cysts without complex portions. Well circumscribed margins, exhibit T1 and T2 hyperintensity on MRI, and do not enhance with administration of gadolinium contrast
		Similar imaging appearance to adenoma malignum. Differentiation is primarily pathological.

Table 2: Differential diagnosis table for Adenoma Malignum

ABBREVIATIONS

CT = computed tomography MR = magnetic resonance

MRI = magnetic resonance imaging

KEYWORDS

Adenoma malignum; cervical neoplasm; pelvic MR

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