A giant cervical nabothian cyst compressing the rectum, differential diagnosis and literature review

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Summary

Nabothian cysts generally being small-sized and multiple are common gynecopathological conditions of women in reproductive age. We report a case of a giant nabothian cyst compressing the rectum apparently without gynecologic symptoms. Initial symptoms of pain and difficulty in defecation due to compression on the rectal wall because of the huge size of the cyst might be the first case in the literature. We made the differential diagnosis from adenoma malignum (minimal-deviation adenocarsinoma) by using immunhistochemical and histopathological techniques.

Key words: Nabothian cyst; Minimal deviation adenocarcinoma; Adenoma malignum; Cervical adenocarcinoma.

Introduction

Nabothian cysts generally being small-sized and multiple are common gynecopathological conditions of women in reproductive age. In some cases, like ours, they rarely advance to large sizes and penetrate the cervical stroma deeply. In cervical smear and biopsy samples they are generally considered as benign structures. It is difficult to distinguish them from a minimal-deviation adenocarcinoma (MDA), which is classified as a special type of cervical adenocarcinoma.

In the differential diagnosis of nabothian cysts, there are only few reports in the literature using immunohistochemical, clinicopathological and magnetic resonance imaging (MRI) techniques. We present a case of a giant nabothian cyst compressing the rectum. We made the differential diagnosis from adenoma malignum (minimal-deviation adenocarsinoma, MDA) by using immunhistochemical and histopathological techniques.

Case Report

A 47-year-old woman with a history of five pregnancies and five deliveries had come to the General Surgery Department of Kağızman State Hospital. She had had significant pain and tingling for the previous six to eight months. Both her personal and family medical histories were uneventful. She had been experiencing regular menstrual periods and did not have any gynecologic complaints.

A mass compressing the rectal cavity at a level of 15-16 cm away from the inlet was the finding on digital rectal examination. Biochemical and hematological analyses consisting of liver function tests, renal functions tests, tumor markers, and urine analysis had been done and the results were found to be at normal levels.

Computed tomography (CT) examination showed an oil-fluid dense cystic mass of approximately 2 cm (20 mm) in diameter with marked partial septal borders on the left lateral side of the rectum in the pelvis and a slightly bulging cecum. The ovaries were not clear in the CT image. Although, the size and structure of the uterine corpus were evaluated as normal, the uterine cervix had not been evaluated by using CT. Afterwards, a colonoscopic examination was done, and the results showed normal images of the descending colon, transverse colon and ileocecal valve. Compression on the external rectal wall was observed at the level of 15-16 cm away from the rectal inlet and a biopsy was taken from that area. Histopathological examination of the rectal biopsy concluded nonspecific rectal inflamation (Figure 1).

The patient was then referred to the General Surgery Department of Kafkas University Medical Faculty for further investigation.

After admission to the General Surgery Department of our hospital, diagnostic and therapeutic laparotomy was planned with the indication of a pelvic mass. Before the procedure a gynecological consultation was requested. In the vaginal and rectal bimanual digital examination the uterus was found mobile and normal in size and consistency. The adnexal and parametrial regions were free from any pathological finding. Compression on the rectal wall was determined by rectal digital examination.

On vaginal examination by the aid of a speculum, an approximately 4-6 cm in size, giant, cystic mass originating from the lower lip of the uterine cervix was detected (Figure 2). Compression of the mass onto the anterior rectal wall was determined by rectal digital examination.

By transvaginal sonographic (TVS) examination, the size and appearance of the uterus and ovaries were determined as normal and the double layer thickness of the endometrium was 16 mm. A cystic mass size of 56 x 49 mm with septations and irregular contours originating from the uterine cervix and protruding into the pouch of Douglas was determined at TVS examination.

After admission of the patient to the hospital gynecology department, as first-line therapy under general anesthesia the giant nabothian cyst was removed and cervical and endometrial biopsies were performed. Atypia and hyperplasia were not detected.

Revised manuscript accepted for publication February 25, 2010

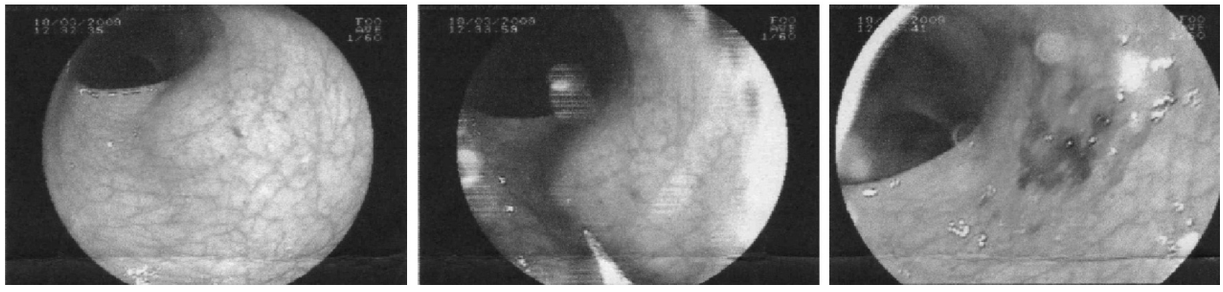


Figure 1. — External compression on the rectum and rectal biopsy view.

Figure 2. — Giant nabothian cyst at the lower lip of the uterine cervix.

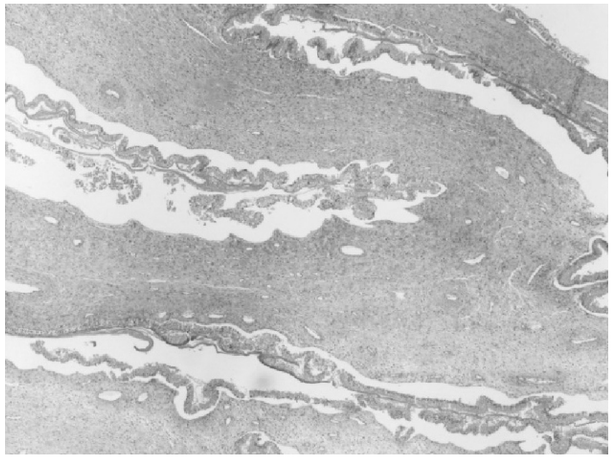
As second-line therapy, total abdominal hysterectomy with bilateral salpingo-oophorectomy was carried out. All pathological samples were evaluated in the Pathology Department of Gaziantep University. Pathological examination of the hysterectomy specimen demonstrated normal uterine, endometrial, and ovarian tissue structures and simple extended glands with flat glandular cells and mucous fluid. The glandular cells had no atypia. Histochemical studies done for hormone receptor determinatation revealed estrogen receptor negative (–), progesterone receptor negative (–) in mucosal lesions, and estrogen receptor negative/positive (±), progesterone receptor negative/positive (±) in normal endocervical mucosa (Figures 3 and 4).

Discussion

Nabothian cysts represent dilated endocervical glands and are common incidental findings on clinical and pathological examination of the uterine cervix [1, 2]. If there are multiple, large and/or deep cysts, it is important and difficult to differentiate them from a MDA which is an uncommon neoplastic lesion of the cervix [1, 3]. Unlike MDA, a nabothian cyst is a common, non neoplastic gynecological disorder. However, it is difficult to distiguish large and/or multiple nabothian cysts, especially deep ones, from MDA [3]. Up to date most nabothian cysts have measured less than 5 mm in diameter but occasional examples have been as large as 15 mm. On microscopic examination, the cysts are found to have the characteristics of nabothian cysts, lined by columnar to flattened endocervical-type cells devoid of atypical features or mitotic activity. Most the cysts are more or less round, but some have slightly irregular contours [1]. They are usually located superficially in the uterine cervix at the location of normal endocervical glands. However, they are rarely located deep or extend into cervical stroma [1, 2]. With their small sizes and well-defined margins nabothian cysts are easily differentiated from most cervical neoplasms [2]. The differential diagnosis of cervical glandular pathologies, neoplasms, nabothian cysts (mucin-filled cervical glands), tunnel clusters (focal hyperplasia) and endocervical glandulary hyperplasia from minimally deviating adenocarcinoma and other premalignant lesions of the cervix can be made histologically. In cases of cervical masses of unexpected large size and appearance, MDA should also be presumably in the differential diagnosis.

MDA of the cervix, often termed “adenoma malignum”, is a rare finding which represents about 3% of the 389 adenocarcinomas of the uterine cervix which are well documented by the Armed Forces Institute of Pathology [2, 4].

Although diagnostic imaging, histopathological and immunohistochemical methods have recorded remarkable advancement in recent years, today a number of abnormalities may not be imaged even if they are the most developed diagnostic techniques. Cystic cervical pathologies are among these abnormalities. Nabothian cysts which present as cervical glands filled with mucous fluid are common gynecologic findings. Tunnel clusters and endocervical glandular hyperplasia are distinguished by means of histological diagnosis [3]. If an unusual cyst is present in the cervix, it must be distinguished from MDA, a rare disease that is extremely difficult to be diagnosed by either cytology or histology from a small biopsy [3, 5, 6].



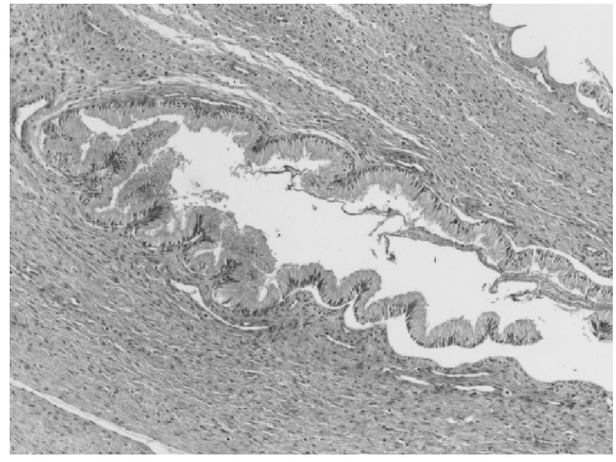


Figure 3. — Glandular structures were furnished with single row columnar endocervical-like glandular epithelium in fibrous stroma (H&E x 100).
Figure 4. — The gland was furnished with single row columnar endocervical-like glandular epithelium (H&E x 200).

Table 1. — *Oguri et al.'s six cases of endocervical pathology*.

Cases	Complaint and signs	USG	Cervical and endocervical biopsy	MRI	Operation
Case 1 (MDA)	Watery discharged and cervical soft tumor	Cervical tumor	Negative (needle biopsy from atypical tumor glandular cells)	Large and multiple cervical cysts	Modified radical TAH and BSO
Case 2 (MDA)	Profuse vaginal discharged	Cervical multiple cysts	Negative	Uterine myoma and large one cervical cyst	Modified radical TAH and BSO pelvic Lenfaden
Case 3 (MDA)	Enlarged cervix Profuse watery discharged	Cervical irregular multiple cysts	Negative	Large and irregular cysts	TAH and right BSO
Case 4 (nabothian cyst)	Vaginal bleeding	Multicystic masses	Negative	Multiple small cysts (5-10 mm)	Deep conization
Case 5 (nabothian cyst)	Enlarged cervix	Multicystic masses	Negative	Several large and small cervical cysts (5-25 mm)	Deep conization
Case 6 (nabothian cyst)	Enlarged cervix	Multicystic cervical lesions	Negative	Multiple small cysts (5-15 mm)	Deep conization

TAH: total abdominal hysterectomy. BSO: bilateral salpingo-oophorectomy. MDA: minimal-deviation adenocarcinoma.

The designation of "adenoma malignum" has been used for a cervical adenocarcinoma characterized by glands with a deceptively benign histological appearance.The first reported case was credited to Gusserow's 1870 report [7]. It was not until 1963, however, that Mckelvey and Goodlin described the first few cases in the English literature [8]. Their report of five cases with a generally poor prognosis was followed a decade later by a report of a series of five additional cases from Silverberg and Hurt [9]. In contrast the tumors of these series had a good prognosis, thus the term MDA was preferred. Only 39 additional examples of adenoma malignum have been reported in the last 25 years. The prognosis in some of these reports has varied markedly [7-11].

Oguri *et al*. published six cases with endocervical disorders. Three of these cases were diagnosed as MDA and the other three cases were diagnosed as deep nabothian cysts by using immunohistochemical and histolopathological techniques and MRI imaging [3] (Table 1).

Li *et al*. reported [12] that MDA had uniform intact membrane tissue and did not have an irregular margin; however, for Oguri *et al*., three cases had irregular walls in deep nabothian cysts and they were diagnosed histologically [3]. In regard to clinical signs and symptoms, MDA presents with a profuse watery discharge; however, deep nabothian cysts do not provoke any complaint of discharge. This suggests that a profuse watery discharge is one of the symptoms of MDA [3]. Gilks et al. reported that most patients with MDA had a watery discharge [3, 11].

Clement *et al*. reported four cases of nabothian cysts which extended deeply into the cervical wall. Well differentiated adenocarcinoma of the minimal-deviation type (adenoma malignum) was the initial diagnostic consideration in three cases [1]. The neoplastic cells of MDA and

most cervical adenocarcinomas were typically immunoreactive for carcinoembryonic antigen (CEA) [10, 11, 13-15]; whereas, normal endocervical cells typically were not. As demonstrated by the findings in case 3, however, there were exceptions to these CEA-staining patterns. Deep nabothian cysts can be distinguished from adenocarcinoma on the basis of examination of HE-stained sections alone [1]. The designation "deep nabothian cysts" has been used to refer to the presence of nabothian cysts deep in the cervical wall, sometimes extending to the serosa [1].These cysts are lined by mucinous epithelium that is typically flattened and lacks an associated stromal response; it is also exceptional for the glands in adenoma malignum to be prominently and extensively dilated in those conditions [1, 10, 11].

Yamashita *et al*. reported two cases of adenoma malignum that mimicked nabothian cysts on MRI. In their two cases, the correct diagnosis was not considered; rather, the preoperative diagnosis of the cystic lesions was nabothian cysts [2]. About 1% of adenocarcinomas of the cervix are so well differentiated structurally and cytologically that they can be diagnosed as malignant only because of the presence of distorted glands with irregular outlines positioned deep in the cervix [2, 5, 6, 12]. Considering that the present case as well as the cases encountered previously had typical histologic features of adenoma malignum, differentiating between adenoma malignum and a nabothian cyst may be difficult without knowing the biopsy or cytologic findings [2].

Conclusion

The case we have defined apparently lacked gynecologic symptoms. Initial symptoms of pain and difficulty in defecation due to compression on the rectal wall because of the huge size of the cyst might be the first case in the literature. We did not find any atypical signs of any supporting adenoma malignum in the cyst and the endocervical samples of this case. However, in the literature, in these types of nabothian cyst cases, though rare, adenoma malignum has been identified.

In light of the literature, evaluation of such cases, whether they are small or large, superficial or deep – each nabothian cyst should be thought to be an adenoma malignum and a more detailed and a skeptical approach should be taken into consideration in the differential diagnosis.

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