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CASE REPORT

"Tunnel Clusters" an Unexplored World for Gynaecologists: A Case Report

Pesona Grace Lucksom[1] · Mingma Sherpa[2] · Barun Kumar Sharma[3] · Vatika Tiwari[1]

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Introduction

Tunnel clusters (TCs) are the presence of multiple cysts in the wall of the cervix due to accumulation of benign endocervical glands in the endocervical wall leading to dilatation of the cervix. Many times they have been reported as large or complex Nabothian cysts of the cervix. A Nabothian cyst is a retention cyst on the surface of the cervix, at the transformation zone, which are caused when the stratified squamous epithelium on the ectocervix grows over the columnar epithelium and blocks the crypt opening. Tunnel clusters are usually incidental finding on hysterectomy and cone biopsy specimens or on radiology. Two types of tunnel clusters have been identified where "Type A" comprises of small noncystic glands, while "Type B" is composed of dilated cystic glands [1]. Women with TCs are usually asymptomatic except for some cases of Type B TCs where the cysts are very large in size. The exact pathogenesis of TCs is not known; however, they are often thought to be due to sub-involution of endocervical glandular hyperplasia following incidences such as previous pregnancies [2]. We report a case of Type B TCs in a multipara which caused a dilemma in diagnosis.

Case Report

A 47-year-old female with previous two vaginal deliveries attended the Out Patient Department of Obstetrics and Gynaecology of Central Referral Hospital, Sikkim, with complain of menorrhagia and pain lower abdomen for past 15 days. She complained of similar episodes for the past one year for which she was given progesterone and tranexamic acid tablets intermittently which did not relieve her symptoms. Other than hypothyroidism she had no comorbidities and no significant surgical or family history. Urine pregnancy test was negative. Her haemoglobin was 12.6 g/dl with adequate platelet count and normal liver function tests and coagulation profile. Ultrasonography pictures showed large multiple cysts in the cervix which was very unusual. On speculum examination there was no growth on the cervix; however, the whole of cervix was difficult to be visualized. Bimanual examination revealed that the uterus was bulky and her cervix was enlarged, but there was no growth. Endometrial biopsy could not be taken as the cervix was ballooned out, making it difficult to take the endometrial aspiration. MRI revealed a multiloculated cystic "grape-like" space occupying lesion measuring approximately

Dr. Pesona Grace Lucksom is a Associate Professor, Department of OBG, Sikkim Manipal Institute of Medical Sciences. Dr. Mingma Sherpa is a Professor, Department of Pathology, Sikkim Manipal Institute of Medical Sciences. Dr. Barun Kumar Sharma is a Professor, Department of Radiology, Sikkim Manipal Institute of Medical Sciences. Dr. Vatika Tiwari is a Post-graduate trainee, Department of OBG, Sikkim Manipal Institute of Medical Sciences.

✉ Pesona Grace Lucksom
pesonadoc@gmail.com

Mingma Sherpa
mingma_sherpa@yahoo.com

Barun Kumar Sharma
drbarun2003@yahoo.com

Vatika Tiwari
vatika.tiwari22@gmail.com

[1] Department of OBG, Sikkim Manipal Institute of Medical Sciences, 5th Mile Tadong, Gangtok, Sikkim 737102, India

[2] Department of Pathology, Sikkim Manipal Institute of Medical Sciences, 5th Mile Tadong, Gangtok, Sikkim 737102, India

[3] Department of Radiology, Sikkim Manipal Institute of Medical Sciences, 5th Mile Tadong, Gangtok, Sikkim 737102, India

4.7 cm × 4.2 cm × 5.8 cm noted in posterior wall of cervix extending to both lateral wall showing T1 heterogeneity with predominant hypointense signal and T2 hyperintense signal of internal content without any abnormal contrast enhancement (Fig. 1). Cervical biopsy was not attempted due to the fear of rupturing the cyst; hence, Pap smear was taken which was negative for dysplasia or malignancy.

She underwent total abdominal hysterectomy. There was approximately 100 ml free fluid in the abdominal cavity of the patient which was mucinous in appearance. Grossly the

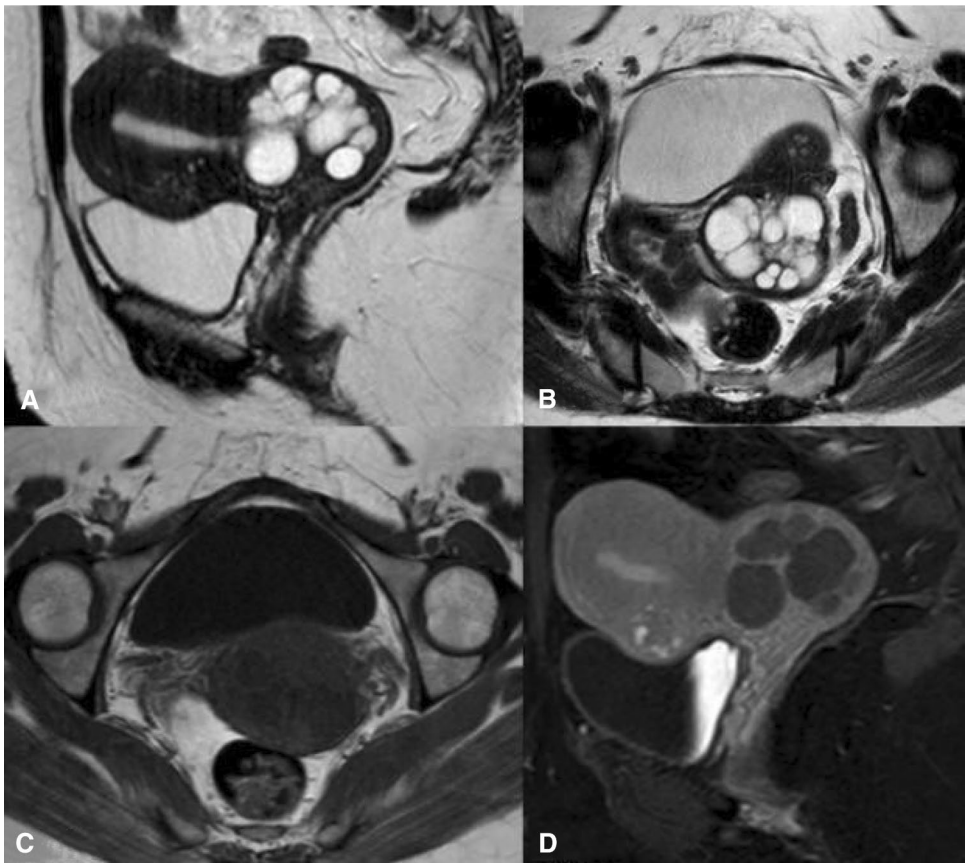

Fig. 1 T2 sagittal (**a**), T2 axial (**b**), T1 axial without contrast (**c**) and T1 sagittal with contrast (**d**) showing multiloculated cystic "grapelike" space occupying lesion in cervix without any contrast enhancement representing tunnel cluster with associated diffuse adenomyosis of uterus and small adenomyoma of anterior wall of body of uterus

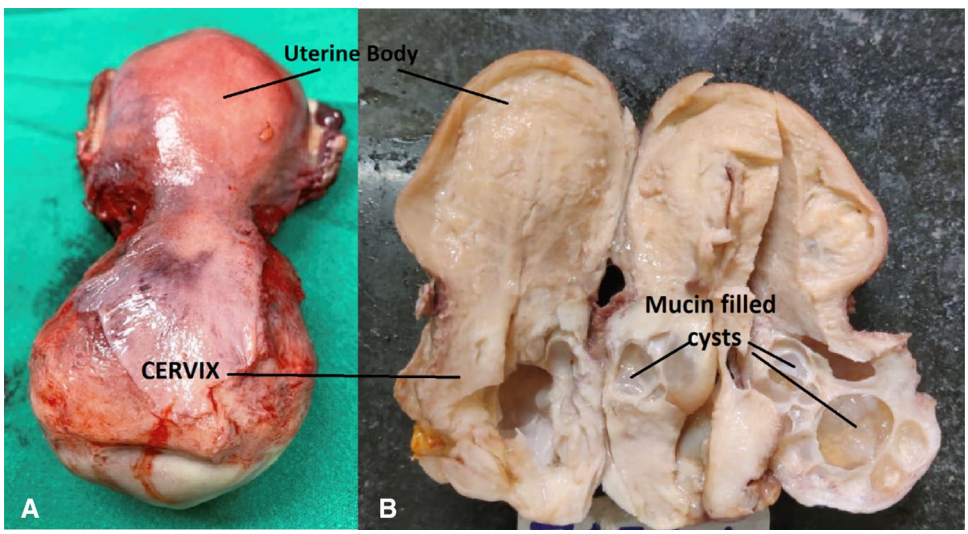

Fig. 2 Gross appearance of the TAH specimen showing cervix to be larger than uterine body (**a**). Cut section of the uterus and cervix showing large mucin-filled cystic spaces in the cervix (**b**)

uterus was bulky (10-week pregnant uterine size), and cervix was found to be larger than the uterine body (Fig. 2a). The cut section of the cervix showed multiple mucin-filled large cystic spaces in the cervix (Fig. 2b).

Her post-operative period was uneventful and was discharged on the 3rd post-operative day. She was followed up once after 2 weeks with her histopathology report. Microscopic examination revealed features of chronic cervicitis with the presence of multiple large multiloculated cysts filled with mucinous material. Cysts are lined by benign columnar epithelium without any atypia. Fallopian tubes showed normal histology. Diagnosis of tunnel clusters Type B was made.

She was not kept under regular follow-up as these cysts are benign.

Discussion

Fluhmann in 1958 first described that the blindly ending small tubules of the columnar gland in the cervix may at times proliferate to form tunnel clusters. TCs are formed as a sequelae to the obstruction at the cleft origin leading to dilation of the cysts due to collection of the mucin secreted by the lining epithelium which is identical to the development of Nabothian cysts. Chief diagnostic concern is its distinction from adenoma malignum also known as minimal deviation adenocarcinoma (MDA) of the cervix. The uniform cystic dilation, absence of mitotic activity or nuclear atypia, and maintenance of lobular architectural arrangement, help to confirm tunnel clusters. There have been reports of deep Nabothian cysts where the Nabothian cyst may occasionally penetrate deep into the cervical stroma and reach the paracervical soft tissue or serosa. Deep Nabothian cysts and TCs have similar appearance under the microscope; hence, whether these deep Nabothian cysts are actually TCs Type B or not is not yet known.

Tunnel clusters have been reported in the literature since a very long time; however, due to its rarity the cysts in the cervical wall are often still misdiagnosed. In the year 1990, 29 cases of tunnel clusters were reported from hysterectomy and conisation specimens out of which except one (96.6%) were multigravida. The age and gravidity of the patients with TCs were found to be greater than those without them; hence, they may represent subinvolution of endocervical glandular hyperplasia [2]. The patient in our case report was also in the perimenopausal age group with two previous vaginal deliveries. Type A TCs are often grossly unremarkable and are usually asymptomatic or maybe associated with mucoid discharge, but Type B TCs show a gross lobular mass in 40% of cases and multiple lesions in 80% of cases [1]. They are rarely visible on clinical and colposcopic examination. In our patient, cervix appeared bulky, while no cyst was visible to the naked eye. Tunnel clusters are benign and have no risk of recurrence or malignant transformation. Type A TCs are of more concern to the pathologists as the cervix appears grossly normal, but the microscopic appearance needs to be differentiated from adenoma malignum. Type A TCs have small elongated noncystic glands lined by columnar to low cuboidal cells. The presence of mild cytologic atypia with pseudostratification, nuclear enlargement, hyperchromasia, vesicular chromatin or prominent nucleoli sometimes makes it difficult to differentiate it from MDA. Jones et al. [3] reported 14 cases of Type A TCs where the mean age of diagnosis was 44.8 years with a mean gravidity of 2.5 and parity of 2.1, where all but one patient were multigravida. All of these lesions were incidental findings, and none was associated with a gross abnormality of the cervix. Gastric mucin expression has been demonstrated in up to 15% of Flumann's Type A TCs [4]. Type B TCs are of more concern to the radiologists and the gynaecologist as these appear as large dilated cysts in the endocervix causing enlargement of the cervix. Histopathologically these TCs show dilated glands containing mucin and lined by columnar or cuboidal or flattened epithelium with no atypia. TCs Type A and B may also occur together. They are also commonly associated with multiple Nabothian cysts [2]. We found 100 ml of free fluid in the abdominal cavity of our patient which was mucinous, and it may have been due to mucin-secreting capacity of the hugely dilated cysts in the cervix.

Type B TCs are very rare and they have often been reported as deep Nabothian cysts [5, 6]. Tunnel clusters Type B should be thought of when there are multiple cysts in the cervix on USG, and an MRI should be done to identify its nature. Gynaecologists should also have a knowledge about TCs because even though clinical and radiological appearances may mimic malignancy they are benign. As TCs Type B are rare it is not yet known whether it is a variant of deep Nabothian cyst or vice versa or they are separate entities.

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Declarations

Conflict of interest The authors declare that they have no conflict of interest.

Human and Animals Rights This report does not involve any research involving Human Participants and/or Animals.

Informed Consent Informed consent from the patient was taken.

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About the Author

Dr. Pesona Grace Lucksom got her undergraduate and Master's degree from West Bengal University of Health Sciences, India. She has worked as a consultant Gynaecologist under the NRHM and under the Government of Sikkim. She has completed training course in Sexual and Reproductive Health Research awarded by Geneva Foundation of Medical and Educational Research. She completed fellowship in gynaecology oncology from Tata Medical Center, Kolkata, India. She has been awarded many prestigious international awards and fellowships in oncology. Dr. Lucksom is currently working as an Associate Professor in the Department of Obstetrics and Gynaecology at Sikkim Manipal Institute of Medical Sciences, Sikkim, India. She has great concern for the health of the people living in rural areas where medical facilities are very difficult to reach.