Case report

Polypoid adenomyoma endocervical type- report of two cases

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Abstract:
Cervical adenomyoma of endocervical type is an under recognized and under reported entity. They usually present as polypoid masses in the endocervical canal and cause non specific clinical complaints like spotting or bleeding per vaginum. We report two cases of this rare entity and a brief literature review in order to create awareness amongst pathologists about this benign but interesting entity.

Key-words: Adenomyoma, endocervical type, polypoid

Introduction:
Most pathologists are aware of endometrial type adenomyomas and report them frequently in general practice. Similar adenomyomas with endocervical type lining have been reported in endocervical as well as endometrial canal and are designated as adenomyomas of endocervical type.[1] The patient age is usually between 21 and 55 years. Most lesions are grossly polypoid though few may be intramural or exophytic. The differential diagnosis includes lobular endocervical glandular hyperplasia, adenofibroma, endocervicosis, tunnel clusters and adenoma malignum. [2] The key histological feature is the presence of bland mucinous glands of endocervical type surrounded by stroma rich in smooth muscle.

Case reports:
Case 1. A 28 year old lady presented with complaints of abnormal uterine bleeding for 6 months in the Gynecology OPD. The bleeding was not cyclical and varied in severity from spotting to few large blood clots. The previous menstrual history was unremarkable. She was P1L1 and the child was born by normal vaginal delivery 4 years back. A cervicoscopy and hysteroscopy was performed which showed a small endocervical polyp measuring 2.0 cm in largest dimension. A polypectomy with endometrial and endocervical curettage was performed and the specimens sent for histopathological examination. Histopathological examination of the polyp showed several glands lined by endocervical type lining epithelium surrounded by stroma with smooth muscle bundles.[Fig.1A] The glandular mucin was alcianophillic on combined alcian blue periodic acid Schiff stain [Fig.1B] and the glands showed nuclear positivity for estrogen[Fig 1C] and progesterone receptors. There was no nuclear atypia in the glands or the stroma. The presence of smooth muscle bundles around the glands and immunopositivity for estrogen and progesterone receptors confirmed the diagnosis of polypoid adenomyoma endocervical type- since these features are absent in adenoma malignum and lobular endocervical glandular hyperplasia (LEGH) which come in the differential diagnosis. The symptoms regressed after surgery.
Case 2. A 35 year old lady presented with complaints of increased vaginal discharge and occasional spotting for last six months to the gynaecological OPD . The previous menstrual history was unremarkable. On cervicoscopy an endocervical polyp measuring 3.5 cm was identified and the patient underwent polypectomy with endometrial curettage . The polyp was sent for histopathological examination. On histopathology, it showed endocervical type glands surrounded by stroma with several smooth muscle bundles.[Fig.2A]. In addition it also showed tubal metaplasia [Fig.2B] and fat metaplasia [Fig.2C] in the stroma. The glands were positive for estrogen receptor and progesterone receptors. A diagnosis of adenomyomatous polyp ,endocervical type was made. The patients symptoms regressed after surgery.

Discussion:
The importance of this entity lies in its differential diagnoses-namely adenoma malignum and lobular endocervical glandular hyperplasia (LEGH). Both these lesions are a part of an emerging spectrum of benign and malignant endocervical glandular lesions with gastric differentiation. Gastric differentiation can be demonstrated by immunopositivity for Muc6 ,M-GGMC-1 and HIK-1083. However these markers are not widely available and an easier approach to confirm the diagnosis of endocervical type adenomyoma is to show estrogen and progesterone receptor positivity since they are uniformly negative in adenoma malignum and LEGH.[1]

Pyloric type metaplasia has also been reported in a rare case of polypoid endocervical adenomyoma.[3] Hence estrogen and progesterone receptor immunohistochemistry is better than markers for gastric differentiation for distinguishing adenomyoma of endocervical type from adenoma malignum. Ectopic gastric mucosa or gastric metaplasia occurs in various organs more widely than previously believed and may be an important cause of carcinogenesis [4] . The other morphologically bland lesions which come in the differential diagnoses include adenomyoma of usual type ,endocervicosis and adenofibroma. Both endocervicosis and adenofibromadonot have a prominent smooth muscle component. The adenomyoma of usual type have endometrioid type glands and stroma . A newly described entity called cervical mesonephric adenomyoma is also a differential[1]. The mesonephric type glands are lined by non mucinous type epithelium with luminal eosinophilic colloid like material. The characteristic immunophenotype of mesonephric glands is- estrogen and progesterone receptor negative,luminal positivity for CD10 and diffuse positivity for vimentin. The unusual histological features described in endocervical type adenomyoma include- focal gland rupture and mucin extravasation, adenofibroma like areas, prominent intraglandular papillary projections with stromal cores, focal atypia of smooth muscle component with giant cell formation and fat metaplasia [1,2]. We found focal tubal metaplasia and fat metaplasia in case two .

Till date there have been two case series each of ten cases of adenomyomas of endocervical type and two case reports in literature [1,2,3,5]. The tumor is usually polypoid but can be intramural or a mass projecting into pelvis from outer aspect of cervix. The presenting complaints are spotting, haemorrhage or copious discharge. Copious mucoid discharge is also the presenting complaint of patients with adenoma malignum which is also a histological differential. The biological behaviour of polypoid endocervical adenomyoma is benign, though one case report has suggested that intraepithelial mucinous adenocarcinoma may arise
from a pre-existing endocervical type adenomyoma of the uterine corpus. The diagnosis was made on endometrial aspirate and the tumor was fragmented so some doubt may exist about the veracity of the claim.[6]

In conclusion the authors conclude that there is a need to create awareness about this interesting entity so that more cases are identified and their biological behaviour is better understood.

Fig.1(A)- Hematoxylin and Eosin (10x)- Endocervical type glands surrounded by stroma rich in smooth muscle with no atypia in the glands and stroma. (B)-Alcian Blue and Periodic Acid Schiff stain (10x)- The glandular mucin is alcianophillic. (C)-Immunohistochemistry for estrogen receptor (10x)- Nuclear positivity in the glands and also in the stroma.

Fig.2(A)-Hematoxylin and eosin (10x)- Endocervical type glands surrounded by stroma rich in smooth muscle with no atypia in the glands and stroma. (B)-Hematoxylin and eosin (10x)- Tubal metaplasia in the glands. (C)-Hematoxylin and eosin (10x)- Fat metaplasia in the stroma.

References