Universidad Pontificia Bolivariana
Vigilada Mineducación
Global Congress on Hysteroscopy
April 26th, 27th and 28th
endogine
GCH2023

Atypical polypoid adenomyoma

Carlos Arturo Buitrago Duque, Daniel Peña Tobón, Miguel Angel Bigozzi

Introduction

Atypical polypoid adenomyoma (APA) is a rare benign tumor, classified by the WHO as a mixed epithelial and mesenchymal tumor of the uterine body. There have been few cases reported, and despite its benign nature, it can coexist with glandular atypia. It occurs in premenopausal women and presents with abnormal uterine bleeding, vaginal discharge, pelvic pain, and postcoital bleeding. Its location in the uterine segment, upper endocervix, and uterine fundus can easily be confused with endometrial polyps or submucosal fibroids. (1,2)

First described by Mazur in 1981, around 500 cases of this entity have been published and its importance lies in the fact that despite its benign nature, these lesions can be confused with invasive adenocarcinoma or malignant mixed Mullerian tumors (2,3)

Objective

To describe two cases of young women with APA diagnosed in two hysteroscopy reference centers in Medellin, Colombia, and Buenos Aires, Argentina.

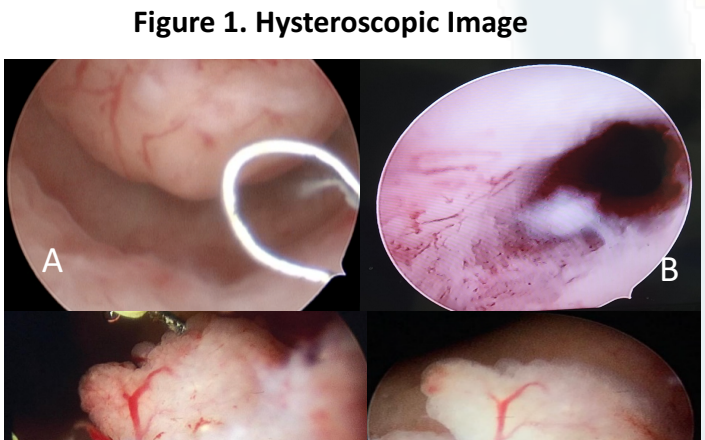
Cases description

Case 1

A 24-year-old patient presented with abnormal uterine bleeding and underwent hysteroscopy based on a transvaginal ultrasound report of an endometrial thickness of 1.7 mm with an image suggestive of a polyp. During hysteroscopy, a glandular endocervical polyp measuring 12 mm in diameter was found in the high endocervical canal, and a parcial laser resection was performed.

The pathological anatomy report showed an atypical glandular lesion with diffuse Pax2 staining, diffuse estrogen receptor staining, patchy P16 positive, negative CK7 staining, CD10 positive in the epithelium and negative in the stroma, and positive h-caldesmon staining in the stroma and negative in the glands, compatible with atypical polypoid adenoma. (Figure 2). The patient underwent resection in the operating room under anesthesia with an invidia 13 Fr miniresectoscope, until the base was completely resected with a deep resection margin. Office Hysteroscopy follow-up with biopsies was initiated every 4 months and after 24 months since polyp resection, no recurrence has been documented.

Figure 1. Hysteroscopic Image



Case 1. A. Hysteroscopic Image Before surgery B. After Hysteroscopy resection
Case 2. C and D Hysteroscopic Image Before surgery

HISTEROSCOPIC FINDINGS

The most usual is to find a single polyp, with increased vasculature on its surface indistinguishable from a hyperplastic polyp (Figure A, C and D).

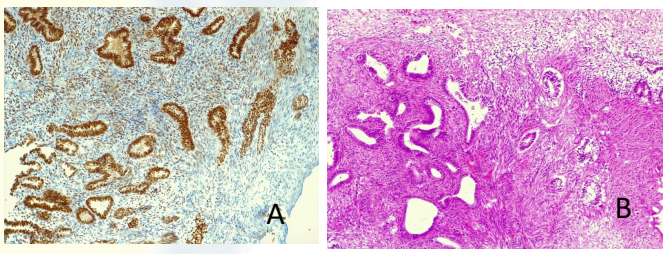
Case 2

A 26-year-old patient presented with abnormal uterine bleeding and desired fertility. A transvaginal ultrasound showed a submucosal fibroid measuring 33 x 17 x 23 mm. An office hysteroscopy was performed in which a polypoid formation was observed, with an irregular, soft surface, approximately 3 cm on the posterior side, close to the OCI with a great increase in vascularity. Complete resection was performed using conventional resectoscopy in the operating room. (Figure 1, C and D)

The pathology report described an APA with branched glands lined by basophilic columnar epithelium and numerous foci of squamous metaplasia forming morulae, separated by a stroma with numerous smooth muscle bundles. Extensive dissociated deciduoid transformation was observed in the superficial zones, with a focus of necrosis on the surface.

The patient achieved pregnancy quickly and returned one year after delivery again with SMA and ultrasound reporting type 0 submucosal myomatous nucleus, measuring 25 x 19 x 19 mm with central and peripheral vascularization, predominantly central. A new operating room hysteroscopy is performed, where they again report an APA lesion, Therefore, it was decided to carry out a total laparoscopic hysterectomy.

Figure 2. Inmunohistoquimic



A. Immunohistochemistry for H caldesmon (brown color) demonstrating muscle fibers surrounding the glands. B. H&E staining with glands surrounded by smooth muscle fibers.

Discussion

Although APA has a benign behavior, it can recur in 44% of postoperative cases, and in meta-analysis the time-to event analysis showed a 10 years cumulative recurrence of 59.54%. However, recurrence is less when management is done by hysteroscopy. The association with endometrial cancer is 16% and concurrent with APA diagnosis in 12%.

Although most of the scientific evidence on this topic is of low quality, with limited sample sizes due to the epidemiology, conservative management is a safe option that requires close follow-up to detect recurrences and avoid malignant transformation of the polyp. (3-5)

The diagnosis of APA is essentially histological. There are no characteristic hysteroscopic findings, but 80% are recognized at the cervical/isthmus level.

Conclusion

APA is a rare benign condition of young women that has a risk of progression to malignancy, conservative treatment is only possible with hysteroscopy. Office hysteroscopy is the best tool for proper follow-up.

Conservative treatment in patients with fertility desires seems to be safe despite the low evidence, but strict follow-up is mandatory due to its high recurrence.

None of the authors do not report disclosure

References

1. Jiang QY, Wang L, Wu RJ. A multiple perspectives on atypical polypoid adenomyoma of uterus. Gynecological Endocrinology. 1 de julio de 2013;29(7):623-5.
2. Domeniconi L, Amadori A, Maniglio P, Saragoni L. Atypical polypoid adenomyoma of the endometrium: diagnosis and treatment. A case report. Pathologica. 1 de diciembre de 2020;112(4):214-8
3. Mikos T, Tsolakidis D, Grimbizis GF. Clinical presentation and management of atypical polypoid adenomyomas: Systematic review of the literature. European Journal of Obstetrics & Gynecology and Reproductive Biology. 1 de mayo de 2019;236:14-21
4. Raffone A, Travaglino A, Saccone G, Alviggi C, Mascolo M, De Placido G, et al. Management of women with atypical polypoid adenomyoma of the uterus: A quantitative systematic review. Acta Obstet Gynecol Scand. julio de 2019;98(7):842-55
5. Biasioli A, Londero AP, Orsaria M, Scrimin F, Mangino FP, Bertozzi S, et al. Atypical polypoid adenomyoma follow-up and management. Medicine (Baltimore). 26 de junio de 2020;99(26):e20491