



Original article

Hemangiomas of the uterine cervix: Association with abnormal bleeding and pain in young women and hormone receptor expression. Report of four cases and review of the literature

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ABSTRACT

Hemangiomas of the uterine cervix are rare with only about 55 cases reported in the literature. Increased awareness of this unusual cervical lesion can lead to early diagnosis and conservative therapeutic approaches. We present a series of four patients with cervical hemangioma with an extensive review of the existing literature on the subject. All four cervical hemangiomas were diagnosed incidentally in hysterectomy specimens performed for persistent menorrhagia or pain. The mean age at presentation was 34 years. The mean lesion size was 2.1 cm and the dominant location was posterior cervix (3 cases). Immunohistochemistry for estrogen and progesterone receptors showed expression of both markers in endothelial cells and stroma, the latter marker showing a stronger and more diffuse pattern. No other significant uterine abnormality was identified in two cases. The vast majority of cervical hemangiomas reported are in reproductive age women. In addition, these lesions express hormone receptors, indicating that their growth is at least in part due to sex hormone stimulation. Although most lesions are symptomatic (mostly bleeding), the diagnosis is frequently unsuspected. Cervical hemangiomas are benign with no recurrences or adverse outcomes reported to date. Conservative treatments are usually successful, and spontaneous remission has been observed. This entity should be included in the differential diagnosis of patients with abnormal vaginal bleeding, particularly in patients of reproductive age with no other clinical and radiologic findings that would explain the symptoms.

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1. Introduction

Hemangiomas of the uterine cervix are exceedingly rare with only a few dozen of cases reported in the literature [1–17]. Although commonly asymptomatic, cervical hemangiomas have been reported to manifest clinically with abnormal bleeding or as a mass on pelvic examination. In such cases therapy, either conservative (ablation, local excision) or definitive (hysterectomy), can be performed. In a significant proportion of cases, however, the diagnosis of cervical hemangioma is unsuspected and only discovered incidentally in hysterectomy performed for refractory abnormal uterine bleeding or pelvic pain. Importantly, many of these patients do not have a definitive diagnosis before definitive surgical treatment.

The pathogenesis of hemangiomas in the cervix and other gynecologic organs is still unclear. The role of hormonal stimulation has been postulated by some authors, since most patients with uterine hemangiomas described in the literature are of reproductive age, and tumors arising during pregnancy have been reported in Refs. [5,15]. In addition, focal estrogen receptor expression has been documented [13]. Further exploration of these observations is still warranted. In addition, a causative link between a hemangioma of the cervix and persisting symptoms requiring hysterectomy requires further documentation. Hence, we describe four cases of cervical cavernous hemangioma in four patients undergoing hysterectomy for symptomatic bleeding and pelvic pain and no preoperative clinical suspicion of this entity. We report the relevant pathologic findings and document their hormone receptor expression status. The current body of evidence in the English literature is discussed.

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Table 1
Clinical characteristics and imaging findings.

	Patient 1	Patient 2	Patient 3	Patient 4
Age (mean 34.5 years)	42 years	30 years	32 years	34 years
Symptoms	Heavy uterine bleeding Pelvic pain	Menorrhagia Dysmenorrhea	Menorrhagia Pelvic pain	Menorrhagia Dysmenorrhea
Past surgical history	Two C-sections Cholecystectomy Tonsillectomy	Tubal ligation Endometrial ablation (at 28)	Tubal ligation	Ectopic pregnancy with salpingostomy (at 25) Two endometrial ablations (at 21 and 31) Right salpingo-oophorectomy and lysis of adhesions (at 31)
Imaging	Magnetic resonance: Intramural and submucosal fibroids (largest 1.7 cm) Pelvic endometriosis	Pelvic and transvaginal ultrasound: No significant abnormality	Pelvic ultrasound: Retroverted uterus, no other abnormality	Pelvic ultrasound: Suggestive of adenomyosis, no other abnormality
Surgery	Hysterectomy, bilateral salpingectomy	Hysterectomy, bilateral salpingectomy	Hysterectomy, bilateral salpingectomy	Hysterectomy, left salpingectomy
Follow up	8 months Unremarkable	16 months Surgical site infection, resolved with antibiotics	One month Post-operative peritonitis with short admission	Not available

2. Materials and methods

Cases of cervical hemangioma diagnosed in the Department of Anatomic Pathology of The Ottawa Hospital in the last 5 years were reviewed. Medical records were also reviewed and patient information, including presentation, clinical and radiologic findings was recorded. Pathologic findings on hysterectomy and follow-up information were also obtained. The material was reviewed by one gynecologic pathologist (CPH) and a pathology resident (AB).

Formalin-fixed, paraffin embedded tissue was retrieved for immunohistochemical testing for estrogen receptor (Leica Biosystem, product code NCL-L-ER-6F11, clone ER6F11, dilution 1:150), progesterone receptor (Leica Biosystem, product code PA0312, clone 16), CD31 (Dako, product code M0823, dilution 1:25) and D2-40 (Dako, product code M3619, dilution 1:100).

3. Results

3.1. Clinical information

Patient 1 was a 42 years old woman with a history of menorrhagia and pelvic pain. Her past medical history included hypothyroidism, depression and obesity. Medical treatment was not successful in controlling the symptoms and hysterectomy was performed. Last follow-up, 8 months after surgery, was unremarkable.

Patient 2 was a 30 years old woman with a long standing history of menorrhagia and dysmenorrhea since the setting of menarche at 13 years old. These symptoms were severe and often required morphine administration. Symptoms persisted despite treatment with oral contraceptives and endometrial ablation. Her gynecological history included two spontaneous vaginal deliveries. An endometrial biopsy showed normal proliferative endometrium. Hysterectomy was performed. The patient developed infection at the surgical site 16 months after the procedure, which resolved with antibiotic treatment.

Patient 3 was a 32 years old woman with menorrhagia and pelvic pain which persisted despite hormonal treatment. Her past medical history was significant for migraine and recurrent ovarian cysts. Her gynecological history included two spontaneous vaginal deliveries. Patient underwent definitive surgical treatment. She developed infection of the surgical site one month after surgery which resolved with antibiotic treatment.

Patient 4 presented at 34 years of age with a long-standing history of menorrhagia and pelvic pain. She had a history of numerous procedures, including salpingostomy for ectopic pregnancy, two

endometrial ablations, right salpingo-oophorectomy and lysis of adhesions. Endometrial ablation specimens showed normal proliferative phase endometrium. The right ovary showed hemorrhagic corpus luteum cyst and follicular cysts. Her symptoms were poorly controlled and eventually worsened after the second ablation, suggesting the possibility of post-ablation syndrome. Imaging studies suggested the presence of adenomyosis. Total hysterectomy was performed. Follow-up after the procedure was not available. See [Table 1](#).

3.2. Histopathologic findings

All four patients underwent laparoscopic total hysterectomy and salpingectomy (bilateral in 3, unilateral in 1). The findings of pathologic examinations of the resection specimens are summarized in [Table 2](#). On initial gross examination the uterine cervix showed localized, ill-defined areas of congestion and hemorrhage ([Fig. 1A](#)). Three lesions were located in the posterior cervix; lesion in patient 1 involved both the anterior and posterior cervix.

Histopathologic examination showed a benign proliferation of dilated and congested blood vessels lined by flat unremarkable endothelium ([Figs. 1 and 2](#)). Lesions ranged from 1.5 to 2.7 cm in size. The vascular proliferation was transmural in all cases extending from submucosal to deep cervical stroma. In fact, the vascular proliferation was within 1.0 mm from the cauterized deep stromal margin in all cases. Hemangiomas were confined to the cervix; uterine corpus and fallopian tubes were uninvolved. An elastic stain was performed in all cases, showing absence of an elastic layer in keeping with the diagnosis of venous cavernous hemangioma ([Fig. 1D](#)).

Immunohistochemical results are depicted in [Fig. 3](#). In all cases, estrogen receptor expression was weak and patchy in the endothelial cells (less than 10%); accompanying stromal cells were diffusely positive ([Fig. 3A and C](#)). Progesterone receptor staining showed stronger and more diffuse staining in both endothelial cells and stroma in all four tumors ([Fig. 3B and D](#)). CD31 ([Fig. 3E](#)) and D2-40 ([Fig. 3F](#)) stains were performed in case 3, showing diffuse endothelial cells staining for the former only.

Additional pathologic findings in the hysterectomy specimens are included in [Table 2](#). Leiomyomata were present in 2 cases: Case 1 had focal adenomyosis involving superficial myometrium as well as numerous leiomyomas ranging from 0.5 to 2.3 cm; all leiomyomas present in histologic sections were intramural or subserosal except for a 1.8 cm submucosal leiomyoma. Case 3 contained a small (0.3 cm) single subserosal leiomyoma. Endometrium showed expected hormonal changes in 3 patients and evidence of

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