

Case Report

Benign Reactive Xanthogranuloma of the Cervix, a Rare Tumor of the Female Genital Tract in a Case Report

Georges Wehbe^{1,2}, Jessie Darido^{1,2}, Mohammad Khaled Ramadan^{1,2}, Rabih Chahine¹, Ahmad Estaitieh¹, Ahmad Lakis³, Omar Sheikh³, Khaled Habib⁴, Saad El Daou⁵, Nicolas Haddad²

¹Department of Obstetrics and Gynecology, Rafik Hariri University Hospital (RHUH), Beirut, Lebanon

²Department of Obstetrics and Gynecology, Lebanese University, Beirut, Lebanon

³Department of Radiology, Rafik Hariri University Hospital (RHUH), Beirut, Lebanon

⁴Department of Pathology, Rafik Hariri University Hospital (RHUH), Beirut, Lebanon

⁵Department of General Surgery, Lebanese University, Beirut, Lebanon

Email address:

jesydarido@hotmail.com (J. Darido)

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Abstract: Xanthogranuloma is a benign disorder that is considered to be a very rare event when it happens in the female reproductive organs. It can mimic adnexal tumors clinically and radiologically. The vulva, the vagina, the cervix or the endometrium can be involved. It is a process during which the tissues are infiltrated by lipid-laden histiocytes mixed with lymphocytes, plasma cells and polymorphonuclear leucocytes. In this article, we are reporting a rare case of an adult xanthogranulomatous inflammation of the uterine cervix with a detailed clinical, pathologic, radiologic and follow up data. A 46-year-old lady presented with vaginal bleeding, dysmenorrhea and pelvic heaviness. She underwent a diagnostic dilation and curetting at a peripheral hospital due to increased endometrial thickness on ultrasound. Then, she was misdiagnosed with a diffused large B cell lymphoma (Non-Hodgkin Lymphoma) and was treated with two cycles of chemotherapy. At our department, abdominopelvic ultrasound and CT scan showed a 10 x 9.4 cm round cervical mass associated with hydronephrosis. A cervical biopsy was also taken turned back with a pseudotumoral xanthogranulomatous cervicitis. Consequently, a hysterectomy with bilateral salpingo oophorectomy was decided and performed taking into consideration the possibility of a coexistent associated malignancy of the reproductive organs. In contrast with juvenile xanthogranuloma, we could not have expected from our case of adult xanthogranuloma to regress. Finally, more studies and reported cases are needed in order to assess the adequate management and utility of radiotherapy/chemotherapy in such cases of Xanthogranulomatous inflammation of the female genital tract.

Keywords: Cervical Adult Xanthogranuloma, Pseudotumoral Xanthogranulomatous Cervicitis, Associated Malignancy, Female Genital Tract

1. Introduction

Xanthogranuloma is defined as a benign tumor of two types: juvenile and adult form. Note that juvenile xanthogranulomas are reported to be most frequently localized on the skin [1]. It is considered destructive to the normal tissue of the affected organs.

Most of the time, it affects the kidney. However, there are other organs: the lungs, gallbladder, stomach, anorectal area, bone, urinary bladder, testis, epididymis, breast, vagina, vulva and endometrium [2-4].

During this uncommon process, foamy cells clusters are formed in the subepithelial connective tissue associated with inflammatory and giant cells. A reactive xanthogranuloma is defined by the presence of dense inflammatory infiltrates.

Cirstoiu M *et al* reported in 2015 a rare case of benign xanthogranuloma located on the uterine cervix in a 44-year-old lady who presented with vaginal bleeding and was treated with a total hysterectomy, bilateral oophorectomy when the histopathologic examination showed atypical hyperplasia of the endometrium and benign but non-reactive xanthogranuloma of the cervix with no evidence of giant cells but with only a slight chronic infiltrate. [5]

It is rare to have a Xanthogranulomatous inflammation affecting the female genital tract. Xanthogranulomatous inflammation mimics adnexal tumors clinically and radiologically. In 1976, Kunakemakorn was the first author to describe an inflammatory pseudotumor in the pelvis. [6]

We hereby report a rare case of adult xanthogranulomatous inflammation of the uterine cervix with detailed clinical, pathologic, radiologic and follow up data.

2. Case Presentation

A 46-year-old lady presented with abnormal uterine bleeding (menometrorrhagia), dysmenorrhea and pelvic heaviness. A transvaginal ultrasound was done showing an increased suspicious endometrial thickness. Diagnostic dilation and curetting was performed at a peripheral hospital and the patient was diagnosed with a diffused large B cell lymphoma (Non-Hodgkin Lymphoma). She received two cycles of chemotherapy but with no response.

At our department, the patient was complaining upon her admission of a bilateral flank pain. Abdominopelvic ultrasound showed a 10 x 9.4 cm round cervical mass associated with bilateral mild to moderate hydronephrosis. Abdominopelvic CT scan showed 10 x 9.4 cm well defined heterogeneously enhancing cervical mass displacing the bladder anteriorly and compressing the right distal ureter with a subsequent moderate hydronephrosis.

Cervical biopsy was also taken to rule out other possibly missed pathologies and to confirm the previous one. It was received in formalin three beige and brownish soft tissue segments ranging in size between 0.7 cm and 1.5 cm totally taken in one cassette. Histo-pathological findings on microscopic examination are as following: A cervical wall infiltrated by foam cells probably of histiocytic origin, mixed with few lymphocytes, associated with fragments of benign non-dysplastic squamous cervical epithelium. The pattern is consistent with cervical uterine xanthogranuloma. Immunohistochemical staining revealed that the foam cells noted in the cervical wall strongly express the Anti-CD68 but neither Anti-CD45 nor Anti-Cytokeratin Cocktail-AE1-AE3. This is compatible with both resorptive reaction and pseudotumoral xanthogranulomatous cervicitis.

Consequently, a hysterectomy with bilateral salpingo oophorectomy was decided and performed. On gross pathology, we had a 9 cm in height x 5 cm across the tubes x 4 cm in the anteroposterior dimension fresh uterus (180 grams). The cervix had a white yellowish mucosal deposit with smooth exocervical aspect measuring 5 cm in maximal diameter. On section the cervical wall was homogenous beige

yellowish. The lower third of the uterine corpus showed intramural myometrial yellowish deposits involving the anterior wall. As for the adnexa, 3 cm unremarkable ovaries and 7 x 0.7 cm fallopian tubes. On Microscopic examination, a section of the cervix and lower third of the myometrium showed a massive infiltration of the cervical wall and the lower third of the myometrial corpus by histiocytic foam cells having pale microvascular cytoplasm and eccentric small sized nucleus with inconspicuous nucleoli. They were arranged in large sheets and nests mixed with small clusters of lymphocytes. The cervico-vaginal surgical margin was infiltrated by the foam cells. The cervical squamous and glandular epithelium as well as the endometrium showed no pathological changes. Both parametria and adnexa were free and showed no pathological diagnostic change.

Immunohistochemical staining showed foam cells strongly expressing the anti-CD68 and mixed polyclonal B-cells CD20+ and T-cells CD3+ clusters expressing anti-Kappa and anti-lambda light chains. The anti-CD138 was expressed by few positive mucosal plasmocytes.

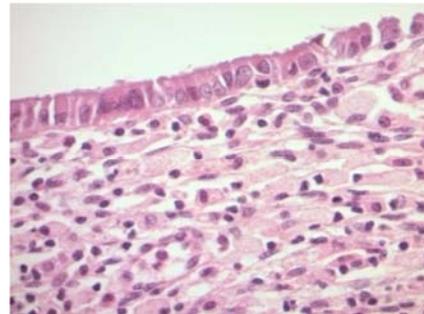


Figure 1. Microscopic examination: section of the endocervix: Massive infiltration of the cervical wall by histiocytic foam cells.

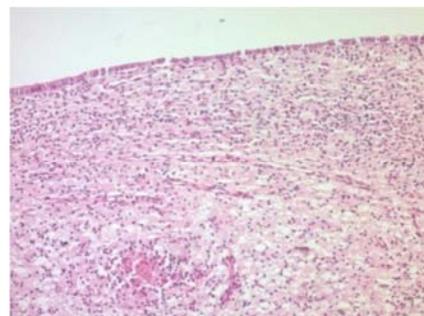


Figure 2. Endocervix.

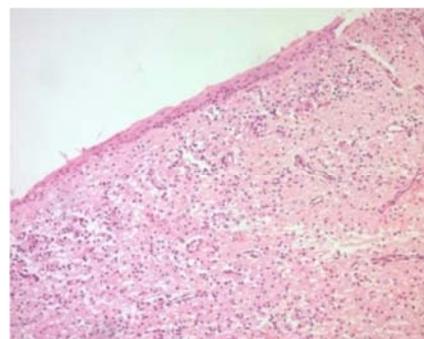


Figure 3. Exocervix.

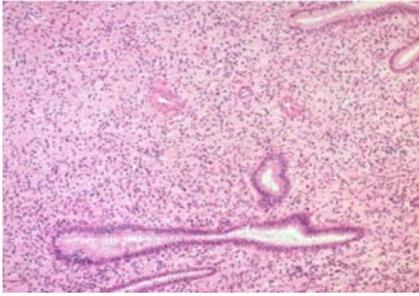


Figure 4. Endometrial infiltration.

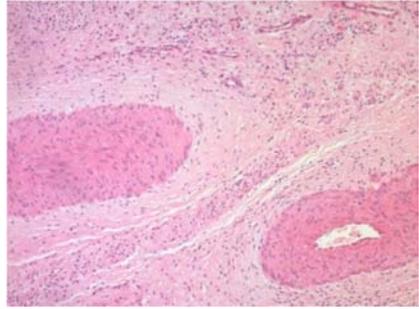


Figure 5. Wall infiltration.

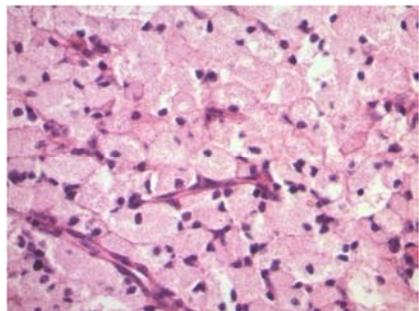


Figure 6. Foam histiocytes.

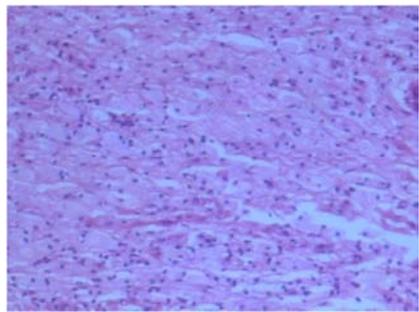


Figure 7. Foam histiocytes.

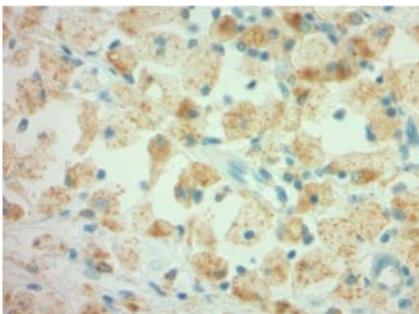


Figure 8. Anti CD 68 X40.



Figure 9. Gross section of round cervical mass.



Figure 10. Gross section of round cervical mass.



Figure 11. Gross section of round cervical mass.



Figure 12. Gross section of round cervical mass.



Figure 13. Abdominal ultrasound showing 10*9.4cm round cervical mass.

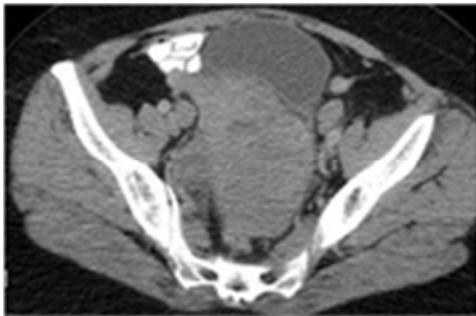


Figure 14. Abdominopelvic CT scan showing a well-defined heterogeneously enhancing cervical mass.

3. Discussion

Xanthogranuloma is a benign disorder characterized by foamy cells clusters that are formed in the subepithelial connective tissue associated with inflammatory and giant cells [7].

The presence of dense inflammatory infiltrates indicates a reactive xanthogranuloma [8, 9]. Taking into consideration the characteristic histopathological issues of our case, it is a reactive xanthogranuloma. Searching the literature, our case is one of the rare described cases of reactive cervical xanthogranulomas associated with dense inflammatory infiltrate.

The localization of the xanthogranuloma at the level of the female reproductive organs is by itself a rare event. the vulva, vagina and endometrium were mentioned as sites of Xanthogranulomatous involvement. Beside its rare location on the cervix, the particularity of our case is being first misdiagnosed for B cell lymphoma of the cervix.

Although the juvenile form often undergoes regression, the adult xanthogranuloma is persistent most of the time. [3] Juvenile form of xanthogranuloma was described in patients with hematologic malignancies such as B-cell acute lymphoblastic leukemia. [10]

It is important to consider the possibility of a coexistent malignancy of the reproductive organs. Russack and Lammers have reported six cases of xanthogranulomatous endometritis

associated with endometrial carcinoma. [11] The association with malignant neoplasias of the same site was also reported in some gastric xanthogranuloma cases that were associated with early gastric cancer. [2] Therefore, the surgical procedure performed in our case is considered as an important prophylactic measurement.

4. Conclusion

Benign reactive xanthogranuloma of the cervix is a rare tumor of the female genital tract. [12] The possibility of a coexistent associated malignancy made the surgical intervention to be an important prophylactic tool. More studies are needed in order to assess the adequate management and the utility of radiotherapy/chemotherapy in such cases of Xanthogranulomatous inflammation of the female genital tract.

Conflict of Interest Statement

All the authors do not have any possible conflicts of interest.

Consent

Written consent has been taken from the patient for publication of this report.

References

- [1] LeBoit PE, Burg G, Weedon D et al. Pathology and genetics of skin tumours. World health organization Classification of tumours Lyon IACR Press, 2006.
- [2] Kinoshita H, Yamaguchi S, Sakata Y, et al.-A rare case of xanthogranuloma of the stomach masquerading as an advanced stage tumor. World Journal of Surgical Oncology 2011; 9: 67.
- [3] Sung Ho Hwang, EunJu Son, KiKeun Oh, et al.-Bilateral Xanthogranuloma of the Breast Radiologic Findings and Pathologic Correlation. J Ultrasound Med 2007; 26: 535-537.
- [4] Castilla EA1, Ormsby A- adult xanthogranuloma of the vulva: case report and review Pathology 2002; 34: 86-7.
- [5] Cristoiu M, Mitache LE, Popa M, Mehotin NC, Sajin M, Cristoiu C-A rare case of benign xanthogranuloma located on the uterine cervix – a case report, Medica a journal of clinical medicine 2015; 10 (2): 127-130.
- [6] Kunakemakorn P, Ontai H. Pelvic inflammatory pseudotumor: A case report. Am J Obstet Gynecol. 1976; 26: 286-287. {PubMed}.
- [7] Kumar V, Abbas AK, Fausto N, et al. – Robbins and Cotran Pathologic Basis of Disease, Eight Edition. Saunders Elsevier Inc., 2010.
- [8] Kurman RJ, Ellenson LH, Ronette BM-Blaustein's Pathology of the Female Genital Tract, 6th edition. New York: Springer Science+Business Media, 2011.
- [9] Nayak S, Acharjya B, Devi B, et al.-Juvenile xanthogranuloma in an adult. Indian J Dermatol 2009; 54 (Suppl S) 1: 83-5.

- [10] Shoo BA1, Shinkai K, McCalmont TH, et al. Xanthogranulomas associated with hematologic malignancy in adulthood. *J Am Acad Dermatol*. 2008; 59: 488-93.
- [11] Narayan R, Mishra M, Mohanty R, et al.-Xanthogranulomatous cervicitis. *Indian J Pathol Microbiol* 2008; 51: 458-9.
- [12] A Rare Case of Benign Xanthogranuloma Located on the Uterine Cervix-a Case Report Monica CIRSTOIUa, b; Luminita Elena MITRACHE a, b; Manuela POPA a, b; Nicoleta Corina MEHOTIN b; Maria SAJINA, b; CatalinCIRSTOIUa, b a “Carol Davila” University of Medicine and Pharmacy, Bucharest, Romania b Emergency University Hospital, Bucharest, Romania.