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Case Report

Case Report: Cervical Cellular Angiofibroma

Mohammad Syarif Hidayatullah[1], Sharvianty Arifuddin[1], Nur Rakhmah[1]

[1]Department of Obstetrics and Gynecology, Faculty of Medicine, Universitas Hasanuddin, Indonesia

Corresponding Author:
Name: Mohammad Syarif Hidayatullah
Email: mohsyarifday@gmail.com

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ABSTRACT

Introduction and importance: *Cellular angiofibroma is a rare benign tumor in the distal genitalia that can occur in women and men aged 46 – 54 years. These tumors are often diagnosed as Bartholin cysts, vulvar cysts, myomas, and unspecified complex tumors.* ***Presentation of case:*** *A 51-year-old female patient with complaints of lumps since 4 months ago but enlarged within 1 week that then diagnosed as a cervical tumor. She was already menopause 1 year ago. On inspection, a reddish solid mass came out of the vaginal introitus with some blackish necrotic appearance. Total abdominal hysterectomy and bilateral salpingo-oophorectomy were performed, and the histological examination confirmed it as a non-specific chronic inflammation and atypical complex endometrial gland hyperplasia.* ***Discussion:*** *Cellular angiofibroma can be caused by the influence of estrogen and progesterone hormone receptors and the sudden transformation of the surrounding tissue. The treatment of cellular angiofibroma is a complete excision of the tumor tissue. However, based on the patient preference and operator consideration regarding the tumor size.* ***Conclusions:*** *Cellular angiofibroma is a rare tumor that usually occurs in the vulva and vagina, usually misdiagnosed as Bartholin's cyst, vulvar cyst, myoma, and unspecified complex tumor. Total excision of tumor tissue and histological examination is essential for diagnosis.*

1. INTRODUCTION

Cellular angiofibroma is a rare benign tumor in the distal genitalia introduced by Nucci et al. in 1997. In women, this disease usually occurs in the vulvovaginal region, especially in the labia majora and vulva, while in men, it can be associated with testicular hydrocele, inguinal hernia, and the inguinoscrotal region. [1,2,3]

Cellular angiofibroma is common in middle-aged women with an average age of 46 to 54 years. These tumors are reported to have initial characteristics of a small, asymptomatic, painless subcutaneous mass that develops slowly and usually takes 1 – 2 years to show symptoms. This condition leads to late diagnosis and worse prognosis.[3]

On Pathological examination, the mass is usually small, around 2.7 cm in size, with clear boundaries and a rubbery gray-white solid surface. While histologically, it has well-defined uncapsulated characteristics consisting of fascicles cells surrounded by collagenous and spindle-shaped cells with medium-sized thick-walled blood vessels. [3,4.]

2. CASE PRESENTATION

A female patient (P5A0) aged 51 years with a chief complaint of mobile lumps in the birth canal along *canalis cervicalis* in the last 4 months that enlarged rapidly within 1 week before admission to the hospital. The patient was previously treated for a cervical tumor in March 2021. There was no history of menstrual disorders before and she has been in menopause since 1 year ago.

The patient's general condition is within a good condition with stable hemodynamics. On physical examination, there was no mass or tenderness in the abdomen. On inspection, a reddish mass with necrotic spots appeared on the introitus vagina. On vaginal examination, a mass with a spongy consistency was found on the cervix with a tumor stalk that could not be identified. On ultrasound examination, a mixed echoic mass was found on the cervix with a size of 6 cm x 7.61 cm. No metastatic process was found in the lungs on chest X-ray examination, and bones were within normal limits. Laboratory examination showed hemoglobin 10 g/dl, leukocytes 8,510 /UL, hematocrit 31.6%, and platelets 316,000 /UL. The next laboratory result found hemoglobin 9.23 g/dl, leukocytes 10,900 /UL, hematocrits 28.1 %, platelets 295.000/UL, prothrombin time 15.5 seconds, thromboplastin time 30.8 seconds, SGPT 21 u/L, urea 11 mg/dl, creatinine 0.6 mg/dl and blood glucose 139 mg/dl.

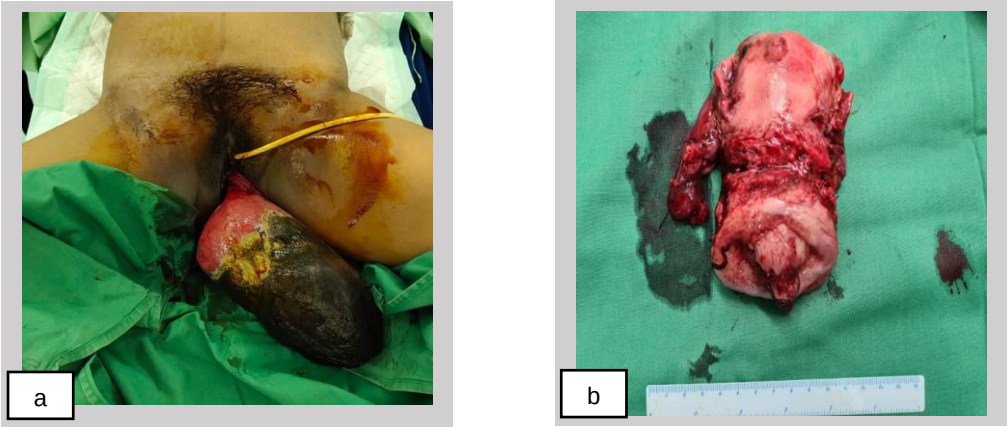

Fig. 1.a) Preoperative. (b) Postoperative.

A total abdominal hysterectomy and bilateral salpingo-oophorectomy were performed, and the tissue was sent to the Pathology Anatomy laboratory. Postoperative management includes observing general condition, vital signs, and vaginal bleeding. The histological examination result was cervical angiofibroma with non-specific chronic inflammation and atypical complex endometrial gland hyperplasia.

3. DISCUSSION

In the case of cellular angiofibroma, tumor progression takes 1-2 years to give symptomatic symptoms. The average size is 2.7 cm, while the largest ever reported is 25 cm. In this case, the tumor size is 20 x 14 cm. The rapid growth of the tumor should be considered the possibility of malignancy or possible aggressive angiomyxoma, a rare mesenchymal tumor that is invasive to the surrounding tissue and has a high recurrence rate. The pathogenesis is still not clear, but the current belief is that it is related to the number of progesterone and estrogen receptors. The clinical manifestations usually appear within 1 – 2 years after the tumor becomes large enough and occur in the population aged 46 – 54 years.

The morphology of the two tumors cannot be specifically distinguished and requires histopathological examination where cellular angiofibroma gives a well-defined, uncapsulated mass that consists of fascicles cells surrounded by collagenous tissue and spindle-shaped cells accompanied by thick-walled blood vessel network measuring medium, while aggressive angiomyxoma provides an overview as well as the extent of invasive into the surrounding tissue with an overview myxoid and hypocellular.[9,10,11] The diagnosis of cellular angiofibroma is challenging, because it has a shape and consistency that resembles other more common tumors, in this case, uterine myoma. This patient was diagnosed early with cervical myoma, which is also rare with a prevalence of 2%, but gives a histopathological appearance of smooth muscle tissue with fibrous tissue. In this case, it was found that the distal part of the tumor gave a necrotic appearance, while in the case of uterine myoma, degeneration due to reduced vascularity would give a necrotic appearance in the center of the tumor due to more vascularization at the peripheral, while cellular angiofibroma consisted of more vascular tissue. [3,5,6]

In addition to histopathological examination, the histochemical examination can also be performed to confirm the diagnosis of cellular angiofibroma by examining the expression of CD34 protein because research by *Mulders et al.* found that 55% of cellular angiofibroma cases secrete CD34 and 50% have receptors for progesterone and estrogen. So, it is believed that the trigger for the occurrence of cellular angiofibroma can be caused by the influence of estrogen and progesterone hormone receptors, and the sudden transformation of the surrounding tissue is also believed to be the trigger for this tumor. Still, the hypothesis has not been fully explained. In this patient, a postoperative histopathological examination showed an appearance of cellular angiofibroma with a uniform, atypical, eosinophilic stromal spindle fibroblast tissue surrounding proliferating blood vessel cells. No CD34 histochemical examination was performed because the initial diagnosis was cervical myoma, and the CD34 count was found to be elevated in myoma cases.

The treatment of cellular angiofibroma is a complete excision of the tumor tissue. However, based on the patient preference and operator consideration regarding the

tumor size, we performed a total hysterectomy abdominal accompanied salpingo-oophorectomy bilateral for this case. All the borders of this tumor cannot be identified and the recurrence risk is higher if we fail to evacuate all the mass. During the operation, intraoperatively, after cutting a portion of the tumor stalk, which was then removed from the vaginal introitus, due to the large size of the mass, we could not evacuate it through the operating field. Therefore, we perform a hysterectomy. A total abdominal hysterectomy in cases of benign tumors can be performed if the mass size is large, accompanied by difficulty in excision of tumor tissue.[5,6] Bilateral salpingo-oophorectomy was conducted with the consideration that the patient had been menopausal in the past 1 year before hospital admission and as a prevention against the possibility of ovarian cancer with a prevalence of 80% after the age of 50 years.[7,8]

Complaints that can be experienced by women who underwent a total hysterectomy are a slight decrease in the frequency of sexual intercourse, libido, and depression. Depression and anxiety are associated with a feeling of pessimism and worthlessness by the illness. Furthermore, it was reported that total hysterectomy also improved complaints related to pelvic pain, previous tumor masses, and patient psychology and anxiety. Complaints related to total hysterectomy were reported to improve 12-24 months after the procedure. This could not be separated from family and social support. Total abdominal hysterectomy with bilateral salpingo-oophorectomy in cases of benign tumors is carried out by considering the benefits to the patient and the basis of bioethical rules in the form of respect for autonomy where the patient has previously been given informed consent regarding the procedure offered and has the right to agree or not to the procedure and also the principle of beneficence where the doctor choose a procedure that scientifically believes provides the best benefit to the patient.[9,10,11]

4. CONCLUSION

Cellular angiofibroma is a rare benign mesenchymal tumor usually found in the vagina and vulva, and other parts of genital organs. Diagnosis of cellular angiofibroma is difficult because it resembles other diseases such as Bartholin cysts, vulvar cysts, complex tumors that are not specific, and myomas. A definite diagnosis can be made by histopathological examination and can also be added by histochemical examination. Treatment can be done by total excision of the tumor tissue with a low recurrence rate. In this case, total abdominal hysterectomy was performed with bilateral salpingo-oophorectomy, considering the large tumor mass and the non-palpable border of the proximal part of the tumor.

CONSENT FOR PUBLICATION

At the point of acceptance, all contributing authors are asked to provide consent to publication, to confirm that they have approved the final version of this manuscript and have made all required statements.

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Conflict of Interest Statement:

The author declares that the case report was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.