

Sudden rapid growth of an aggressive angiomyxoma after taking pomegranate seeds oil

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Aggressive angiomyxoma (AA) is a rare soft tissue tumor of undetermined etiology that occurs mainly in the female vulva and perineum. It has been demonstrated to increase in size under the estrogenic stimulation and shown to respond to treatment with a GnRH agonist. A 47-year-old woman was presented with a large vulvar mass. The mass had grown slowly before she took pomegranate seeds oil on a regular basis for a year, which is known to contain natural estrogen for a year. The almond-sized mass enlarged into a fetal head-sized one in a year. Our case is implying that external administration of estrogenic substance may be a contributory factor to the sudden accelerated growth of AA. Although the past medical history in this case relies on an unverified observation by the patient alone, the unique clinical course and impressive gross appearance of AA may help clinicians with diagnosis in practice.

Key Words : Angiomyxoma, Pomegranate seeds oil, Vulva

INTRODUCTION

Aggressive angiomyxoma (AA) is a rare neoplasm of the female pelvis and perineum. In 1983, Steeper and Rosai presented nine cases of distinctive soft tissue tumors in the female genitalia and pelvic soft tissues, and termed it 'aggressive angiomyxoma'.¹ The vulva, pelvic floor, vagina, and perineum are usual sites of origin in women. AA tends to occur most commonly during the reproductive years with a median incidence in the fourth decade.² AA is a slowly growing and locally infiltrating tumor. In occasional cases, metastasis has been described. Local, destructive recurrence occurs in approximately 30-40% of cases, and often appears many years or sometimes decades after the initial excision. The etiology of AA is undetermined.³ It has been demonstrated to increase in size under the estrogenic stimulation and shown

to respond to treatment with a GnRH agonist.⁴ In this report, we describe a case of sudden rapid growing AA after regularly taking pomegranate seed oil, which is known to contain natural estrogen.

CASE REPORT

A 47-year-old woman, gravida 2, para 2, was found to have a huge mass of the right vulva when she presented to the department of obstetrics & gynecology clinic at Ewha Womans University Hospital on October 5, 2005. The patient recalled that she first recognized the mass in 1996, which was soft, nontender, and almond sized. Since the presumptive clinical diagnosis at a primary clinic was of a Bartholin gland cyst at that time, the clinician recommended a regular follow up. The mass then increased in size extremely slowly over the ensuing eight years, and she forwent regular gynecologic examinations. However, one year before this presentation she started regularly taking pomegranate seed oil as a health food, and thereafter, the mass grew

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Fig. 1. Pedunculated vulval mass.

more rapidly. On examination, a pedunculated, globular mass of $22 \times 21 \times 8$ cm was seen arising from the middle portion of the right labia majora. The mass was nontender with overlying engorged skin (Fig. 1). Other external genitalia and a bimanual pelvic examination were normal. For further diagnostic investigation, computed tomography (CT) was carried out, and revealed a mass originating from labia majora, but not extending toward the perineum or pelvic fossa. The clinical diagnosis was vulvar leiomyoma with cystic degeneration or of a rapidly growing vulvar lipoma. Under this impression, she underwent surgical excision of the huge mass on February 17, 2005. Surgical margins were free of tumor in frozen section, and its cut surface had a glistening, pale, gelatinous appearance. It was well demarcated and focally infiltrative, but not encapsulated. Tissue sections were stained for hematoxylin-eosin (H&E), estrogen receptors (ER), progesterone receptors (PR), vimentin, S100, smooth muscle specific actin (SMA), desmin, CD34, and factor VIII related antigen. H&E staining showed the neoplasm was paucicellular and consisted of bland spindle-shaped cells set within a copious myxoid matrix containing variably sized vessels (Fig. 2). Neoplastic cells were frequently clustered around vessels in whorls. Despite a careful search, no mitotic figure was identified. Immunohistochemistry revealed strong positivity for vimentin, desmin, and SMA, but negativity for S100. CD34 was focally positive in spindle cells and strongly positive in vessels; Factor VIII

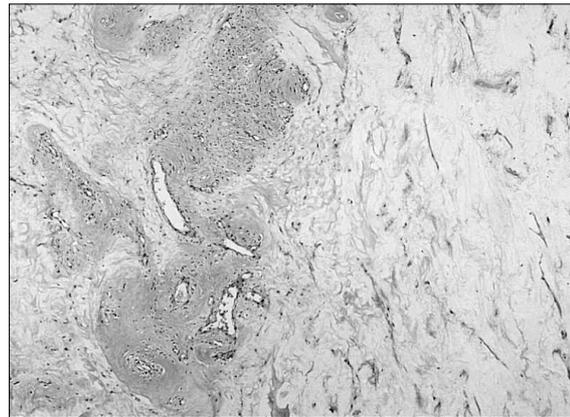


Fig. 2. Aggressive angiomyxoma was paucicellular with bland spindle-shaped cells set in a copious myxoid matrix. Vascular components were conspicuous and composed of variably sized vessels (H&E, $\times 100$).

related antigen positivity was confined to vessels; ERs were stained positively in about 60%; and PRs were strongly positive in $>90\%$ of neoplastic cells. Based on its histological features and immunohistochemical staining findings, a diagnosis of AA was confirmed. The patient had no postoperative complications and was discharged with a restored anatomy.

DISCUSSION

Steeper and Rosai first reported an AA of the female pelvis and perineum in 1983.¹ The tumor characteristically grows slowly and insidiously, and is focally infiltrative. Tumors measuring between 3 and 60 cm have been reported.⁵ Clinically AA is often mistaken for a vulvar lipoma, abscess, Bartholin's cyst, Gartner's duct cyst, vaginal cyst, vaginal prolapse, or levator hernia. The final diagnosis of AA is always confirmed by histology. Differential diagnoses on inspection of the cut surface of the tumor include myxoma, myxoid neurofibroma, pseudo sarcoma, myxoid liposarcoma, and myxoid leiomyosarcoma.

AA is usually a deep infiltrative lesion rather than a polypoid one. Our case is distinct from previously reported cases in that she presented with polypoid mass arising from vulva. The tumor had a dominant myxoid histological pat-

tern and prominent vascularity, which are characteristics of AA.

The treatment of choice is wide surgical excision to avoid recurrence. Amezcua et al reported that immunohistochemistry showed positivity for ER (10/12) and PR (11/12) in 12 cases of AA.⁶ These findings suggest that circulating hormone may influence the behavior of these tumors. Htwe et al reported a 41-year-old woman who presented with an AA of the left vulva at 18 weeks of gestation. An immunohistochemical examination of this tumor showed strong positivity for PR but negative for ER.⁷ Ribaldone et al also reported case of AA increasing in size during pregnancy with positivity for ER and PR.³ These findings indicate that AA has a hormonal dependency and provide a rationale for management with GnRH agonists. McCluggage et al suggests that GnRH agonists may be of value in managing cases of AA, either primary or recurrent, which are not amenable to surgical excision.⁴

Our patient had taken pomegranate seed oil as health food for one year, and the tumor grew more rapidly during this period. Pomegranate seed oil is known to contain estrogen. Moneam et al obtained estrogen from pomegranate seeds by absorption high-performance liquid chromatography (HPLC),⁸ and Okamoto et al reported pomegranate extract alleviated depressive state and bone properties in an ovariectomized mice menopausal syndrome model.⁹ These results indicate that pomegranate oil has estrogenic activity.

She had no history of exposure to other estrogenic sources. There were no other causes presumed to aggravate tumor except pomegranate seed oil. Therefore, sudden rapid growth of AA may be due to pomegranate seed oil.

In conclusion, our case is implying that external administration of estrogenic substance may be a contributory factor to the sudden accelerated growth of AA. Although the past medical history in this case relies on an unverified obser-

vation by the patient alone, the unique clinical course and impressive gross appearance of AA may help clinicians with diagnosis in practice. Moreover, Given positive estrogen and progesterone receptor status of this tumor, we will consider hormonal treatments in the case of a future relapse.

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석류씨 오일 복용 후 갑자기 커진 공격성 혈관점액종 1예

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김미경¹ · 신은경¹ · 심인숙¹ · 이선옥¹ · 주 웅¹ · 김승철¹ · 성순희² · 한운섭²

공격성 혈관점액종은 주로 여성의 외음부와 회음부에서 발생하는 드문 연부조직 종양으로 원인은 분명치 않다. 공격성 혈관점액종은 에스트로겐의 자극 하에 증가하는 것으로 보고되고 있으며, GnRH 유사체가 치료제로 사용되기도 한다. 저자들은 외인성 에스트로겐을 함유하는 것으로 알려진 석류씨 오일 복용 후 크기가 급격히 증가한 공격성 혈관점액종 1예를 경험하였기에 문헌 고찰과 함께 보고하는 바이다.

중심단어 : 공격성 혈관점액종, 석류씨 오일, 외음부
