Case Report

Cellular Angiofibroma of Vagina Presenting with Secondary Infertility

Jyotsna Naresh Bharti, Biswajit Dey *, Vikram G Raj, Prerna Arora

- *Department of Pathology, Maulana Azad Medical College, New Delhi, India*

Abstract

Background: Cellular angiofibroma, first described in 1997, is known to occur in both genders with equal predilection occurring in middle aged females and older males.

Case Presentation: In this study, a case of vaginal cellular angiofibroma was reported in a 30 year old female presenting with secondary infertility. The case was diagnosed based on morphology and immunohistochemistry and was treated surgically. The interesting feature of the case was the rarity of its incidence at the vagina and its resemblance to other benign and more aggressive tumours in the same site.

Conclusion: Cellular angiofibromas are benign tumours, which rarely occur in vagina. Although middle aged females are affected more, cellular angiofibromas can affect females of reproductive age group and can cause secondary infertility. These tumours need to be distinguished from other benign tumours and aggressive tumours occurring in the same site.

* *Corresponding Author:*
Biswajit Dey, Department of Pathology, Maulana Azad Medical College, New Delhi, Indai
E-mail: drbish25@rediffmail.com

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Introduction

Cellular angiofibroma was first described by Nucci et al. in the year 1997 (1). It is a rare benign tumour of mesenchymal origin and known to occur in the genital regions of both genders predominantly affecting the vulva of middle aged females and inguinal region of elderly males (2). However, it has been rarely reported to occur in the vagina (2, 3). In this study, a case of vaginal cellular angiofibroma was reported in a 30 year old female presenting with secondary infertility.

Case Presentation

A 30 year old female presented with complaint of vaginal foul smelling discharge and mass coming out from vagina for one month. Patient was married for seven years and her last child birth was five years ago, which was uneventful. She complained of inability to conceive for the last six months. Fertility work-up revealed a regular menstrual cycle, with no biphasic temperature curve. Semen analysis of the husband was normospermic. Patient did not complain about any urinary or gastrointestinal disturbances.

Her vital parameters and general examination findings were within normal limits. Abdominal examination did not reveal any guarding, tenderness or rigidity. On gynaecological examination, uterus was midline. Vagina showed a growth on the anterior wall with prolapse of the anterior vaginal wall. The surface of the mass was focally covered with slough and bled on touch. A clinical diagnosis of infected vaginal fibroid causing secondary infertility was made.

The growth was completely excised and sent for histopathological examination. Grossly the growth was polypoidal in shape and measured 10x3x3 cm in size (Figure 1A). Cut section was solid, grey brown without any areas of hemorrhage or necrosis (Figure 1B). Microscopic examination revealed an ulcerated mucosa lined by stratified squamous epithelium with underlying tumour. The tumour was circumscribed and moderately cellular

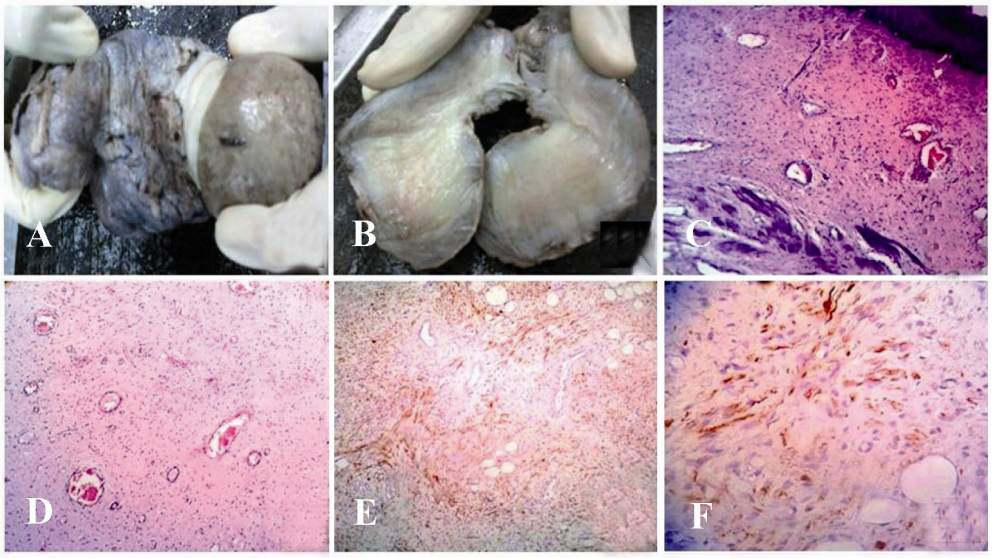

Figure 1. A: Excised polypoidal growth; B: Cut surface was solid, grey brown without any areas of hemorrhage or necrosis; C: Section showing unremarkable stratified squamous epithelium and tumour comprised of spindle and stellate shaped cells with many prominent blood vessels (hematoxylin and eosin stain, 100x); D: Section showing spindle and stellate shaped tumour cells with many prominent blood vessels in a myxoid stroma (hematoxylin and eosin stain, 100x); E: Vimentin positivity in tumour cells; F: Focal desmin positivity in tumour cells

comprised of spindle and stellate shaped cells with many prominent blood vessels (Figure 1C). The stroma was myxoid and partly collagenized (Figure 1D). There was little or no nuclear pleomorphism. No mitotic figures were identified. Immunohistochemically, the tumour cells were strongly positive for vimentin (Figure 1E) and focally positive for desmin (Figure 1F). The tumour cells were negative for smooth muscle actin (SMA) and S-100. Based on morphological and immunohistochemical features, a diagnosis of cellular angiofibroma was made. The postoperative period was uneventful.

Discussion

Cellular angiofibroma is a rare benign tumour originating from the superficial soft tissues of the genital area (2). It occurs in both the genders with equal predilection occurring in middle aged females and older males (4). The sites involved by this tumour are vulva in females and inguinoscrotal region in males (2, 4). Vaginal involvements have rarely been reported (2, 3). Extragenital organs like knee, eyelid, anus retroperitoneum and oral mucosa have also been reported to be involved by this tumour (2). Clinically, it presents as a bleeding polypoidal or nodular mass (3). In this case, the patient presented with a mass coming out from vaginal introtus along with vaginal foul smelling, causing secondary infertility in the patient. Vaginal masses cause secondary infertility by distorting the shape of uterine cavity and cervix.

Clinically and morphologically, cellular angiofibroma can simulate other benign tumours like leiomyoma, angiomyofibroblastoma, spindle cell lipoma and perineurioma (1, 5). In this case, it was clinically confused with leiomyoma. Cellular angiofibroma can also be confused with aggressive vulvo-vaginal tumours like aggressive angiomyxoma and solitary fibrous tumour (1, 5). Although there is some morphologic overlapping of cellular angiofibroma with the above mentioned differentials but bland spindle cells and prominent hyalinized blood vessels along with wispy collagen are distinctive for cellular angiofibroma (1, 2, 5). Moreover, immunohistochemistry plays an important role in differentiating this tumour from other vulvo-vaginal tumours. Cellular angiofibromas are consistently positive for vimentin with variable expression of CD34 and desmin, and negative for smooth muscle actin (SMA) and S-100 (2, 6).

It is of paramount importance to distinguish cellular angiofibroma, which rarely recurs, from more locally aggressive tumours like aggressive angiomyxoma and solitary fibrous tumour. However, it might be difficult sometimes because of

the overlapping features. In the present case, circumscribed nature of the tumour with moderate cellularity and focal positivity for desmin were all in contrast to that of aggressive angiomyxoma. Aggressive angiomyxoma is a hypocellular tumour, which shows diffuse positivity for desmin and has infiltrative margins (7). Negative staining of tumour cells for CD34 helped to rule out solitary fibrous tumour in this case. However, 60% of cellular angiofibromas are also positive for CD34 (4). Solitary fibrous tumour is characterized by areas with the staghorn vascular pattern and dense collagenous stroma in contrast to hyalinized round vessels and wispy collagen of cellular angiofibroma.

Cellular angiofibroma has little tendency to recur. Therefore, a complete local excision with clear margins is an adequate treatment (1, 2).

Conclusion

Cellular angiofibromas are benign tumours, which rarely occur in vagina. Although middle aged females are affected more, cellular angiofibromas can affect females of reproductive age group and can cause secondary infertility. These tumours need to be distinguished from other benign tumours and aggressive tumours occurring in the same site.

Conflict of Interest

The authors declare no conflict of interest.

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