Case Report

Journal Of Clinical & Medical Images Case Reports

Open Access, Volume - 2

Suspected Prostate Malignancy in Elderly Male Results in an Incidental Finding of Cellular Angiofibroma of the Male Pelvis

Nada Shaker[1]*; Anil V Parwani[1]

[1]Department of Pathology, the Ohio State University Wexner Medical Center, USA.

Received Date : May 23, 2022
Accepted Date : June 21, 2022
Published Date : July 06, 2022
Archived : www.jcmimagescasereports.org

***Corresponding Author:** Nada Shaker, Department of Pathology, the Ohio State University Wexner Medical Center, USA.
Email: nada.shaker@osumc.edu

Introduction

Cellular angiofibroma (CAF) is a rare benign mesenchymal tumor that affects both genders in the genitourinary region [1]. Nucci et al. first described this entity in the vulva in female in 1997 [2]. Cellular angiofibroma is generally a slow-growing and form asymptomatic mass that primarily arises in the vulvar-vaginal region in female and the inguinal region in males [3]. Although rare, cases have been reported in the pelvic and extra pelvic regions [4]. It mainly affects women mostly during the fifth decade of life [1]. "CAF" is the applied terminology of this tumor in accordance with World Health Organization [5]. It can easily be misdiagnosed as spontaneous perineal herniation in clinical practice [6]. Detailed pathological examination and correlation with immunohistochemistry workup are imperative to distinguish it from various soft tissue tumors which affect the pelvic regions. Differential diagnosis of this tumor includes both benign and malignant lesions such as spindle cell lipoma, solitary fibrous tumor, angiomyofibroblastoma, leiomyoma, and aggressive angiomyxoma [7]. Treatment with simple local excision is generally curable due to low local recurrence and no chance of metastasizing [8]. Cellular angiofibroma has a good prognosis [9]. However, preoperative distinction of cellular angiofibroma from malignant tumors can be challenging [10]. Periurethral CAF is rare with only two cases reported in the periurethral region [11, 12]. The current study presents a third rare case of cellular angiofibroma affecting the periurethral region in 69-year-old male that measured approximately 7 cm. This case was suspected a prostate malignancy with possible bladder invasion on the radiology imaging.

Case presentation

A 69-year-old male with a past medical history of coronary artery disease, aortic insufficiency, hyperlipidemia, gout, and melanoma who presented with enlarged prostate, elevated PSA of 8.6 and lower urinary tract symptoms of weak stream and mild nocturia. Patient had been followed by urologists for his prostate enlargement since 2011 when he had negative prostate biopsies. His PSA was trended and followed carefully with repeatedly negative prostate biopsies. Last PSA measured 7.13. MRI of the prostate showed an enlarged prostate (130mL) with two PI-RADS 4 lesions on the right and left transitional zones and a substantial periurethral mass of 7-8 cm. (**Figure 1**).

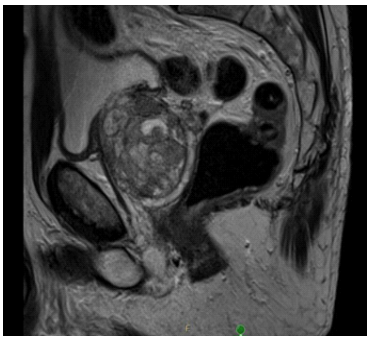

***Figure 1:** Sagittal T2 weighted MRI showed non uniform periurethral mass between the posterior vesicle and compressing rectosigmoid junction. Overall, well circumscribed 7 cm mass with possible bladder invasion.*

Diagnosis:

After review of the pelvic MRI and concern for periurethral mass and enlarged prostate, we performed a cystoscopy, obtained MRI guided TRUS prostate biopsies and periurethral mass biopsy. Cystourethroscopy showed no urethral stenosis or stones. Prostatic enlargement of median lobe was noted. No lesions or neoplasms were identified within the bladder. MRI guided biopsy of prostate showed no significant pathologic change in tissue appearance. Periurethral mass biopsy showed cellular angiofibroma. The large periurethral mass and pathology of cellular angiofibroma were discussed with the patient and he went to the operative room for perineal excision of this mass via an incision in the perineum. The lesion was very close to the right side of the external sphincter and was noted to be adhered to the urethra and was quite vascular. The entire lesion was excised and sent for pathology.

Pathology Gross Description

Three irregular, pale tan to purple-tan soft tissue fragments

Citation: Nada Shaker. Suspected Prostate Malignancy in Elderly Male Results in an Incidental Finding of Cellular Angiofibroma of the Male Pelvis. J Clin Med Img Case Rep. 2022; 2(4): 1186.

measuring 1.2 x 0.5 x 0.4 cm, 2.1 x 1.0 x 0.9 cm, and 2.3 x 2.2 x 0.6 cm. The smaller tissues were somewhat ragged and partially disrupted. The larger tissue was sectioned to reveal tan-white, homogeneous, semi-translucent cut surfaces. There was no gross evidence of necrosis or calcification. Focal hemorrhage was identified.

Pathology Microscopic Description

The microscopic appearance was characterized by a cellular neoplasm composed of uniform, bland, spindled stromal cells with scant, lightly eosinophilic cytoplasm with ill-defined borders, oval to fusiform nucleus. Numerous thick-walled and hyalinized vessels were noted. Scarce component of mature adipocytes was present. (**Figure 2**). The stromal cells were positive for desmin and CD34; and negative for CD117 (**Figure 3, 4**).

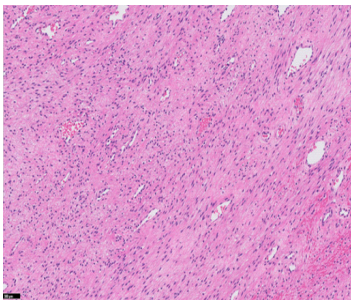

Figure 2: *Hematoxylin-eosin staining shows the cellular spindle cell component and the hyalinized blood vessels.*

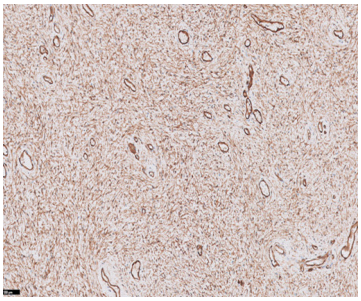

Figure 3: *Iimmunohistochemical staining for CD34 reveals positive staining.*

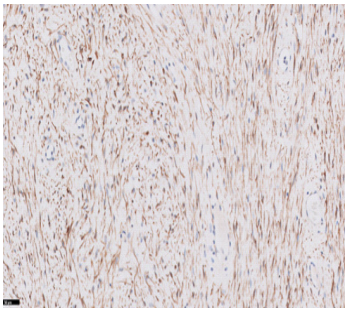

Figure 4: *Immunohistochemical staining for Desmin reveals positive expression.*

Patient Follow-up:

Patient's post-operative course was complicated by urinary retention for which he practiced self-catheterization and took Flomax for incomplete emptying symptoms. He also developed a fever of 102 with urine cultures growing *Serratia macarenes*, thus his UTI was treated with ciprofloxacin. At his post operative 6-month check-up, his PSA was 8.49, prostate estimated at 114g, and he was doing well. Patient follow-up showed no evidence of recurrence.

Discussion

Cellular angiofibroma is a rare benign tumor that has been described in several case studies in children and adults. In terms of the genital and pelvic regions, it is most associated with the labium and vulvovaginal areas in females, and superficially in the scrotum and inguinal canal in males. Cellular angiofibromas have been described in the nasopharynx as well as the female and male genital regions [9, 12, 13]. In 1998 Laskin et al. reviewed angiomyofibroblastomas in the male genital region with six scrotal cases and five inguinal cases. These tumors were superficially located and well circumscribed. After simple excision, one of seven patients on follow up demonstrated recurrent/persistent disease. CAF have been described as more common in women as demonstrated by Mandata et al when they gathered 79 cases of CAF in women from 1997 to 2014. They found that 56% of these CAF cases showed tumor growth in the vulvovaginal and labium region. Similarly, most male cases have been described in analogous male genitalia such as the scrotum and inguinal regions. Unusual location of cellular angiofibroma has been also described in the prostate in a male patient [14].

The diagnosis of cellular angiofibroma can be challenging especially when suspected malignancy is necessary to be ruled out. Valuable information is often provided by MR imaging, especially when ultrasound and/or clinical examinations are inconclusive [15]. Combining conventional and functional magnetic resonance data provides useful diagnostic information in the preoperative characterization of scrotal masses [16]. Generally, diagnosis of CAF is established with biopsy proven pathological examination and immunohistochemical staining. Grossly, CAF presents as a well circumscribed mass without a capsule and often involves superficial tissues. Microscopic images demonstrate bland spindle shaped cells with fusiform nuclei and pale eosinophilic cytoplasm with wispy collagen bundles in the extracellular matrix. Small to medium sized thick walled hyalinized vessels and scattered adipose tissue are also characteristic of the tumor. Cellular atypia and necrosis are uncommon. Sarcomatous transformation in a cellular angiofibroma is reported in only one rare case [17]. Immunohistochemistry findings specific the tumor cells include vimentin, CD99, CD 34, ER/PR, and desmin.

These tumors contain two principle components: the cellular spindle stromal cells and the prominent thick-walled hyalinized blood vessels [18]. The differential diagnosis of this unique tumor includes angiomyofibroblastoma, spindle cell lipoma, solitary fibrous tumor, leiomyoma and aggressive angiomyxoma. CAF is a mesenchymal neoplasm that falls in the spectrum of angiomyofibroblastoma, a well circumscribed less cellular benign neoplasm, and aggressive angiomyxoma, which can be a fast growing locally infiltrative tumor with a tendency to recur when incompletely excised. Angiomyofibroblastoma, cellular angiofibroma, and aggressive angiomyxoma can be distinguished via histologic appearance, immunohis-

tochemistry staining, and the clinical findings. All published cases recommend close follow up of lesions and in the case of rapid increase in size, prompt surgical excision of mass with negative margins is appropriate with the goal of preventing recurrence [19]. In 2013, Emtage et al. described a case of a middle-aged man with voiding symptoms and a large rapidly growing 13.5 cm cellular angiofibroma of male pelvis. This team elected simple surgical resection, but there was no report on follow up to track recurrence or metastasis. A recent case report from Kumar et al in 2018 described a female periurethral mass that had been enlarging to 2-3 cm over 8 years. Simple surgical excision of this mass resulted in cure with no signs of recurrence in the 2 years follow up period.

Conclusion

In summary, CAF in males represents a distinct benign neoplasm with a broad anatomic distribution. Uncommon location in the periurethral region is rarely reported. This is believed to be the third reported case of cellular angiofibroma in the periureteral region. Any lesion in genitourinary areas should be taken with high suspicion of malignancy and the specimen should undergo pathologic and immunohistochemistry examination and appropriate correlation. Once the diagnosis is made, treatment with simple local excision with tumor-free margin is adequate to avoid recurrence of the lesion.

References

1. Aydın Ü, Terzi H, Turkay Ü, Eruyar AT, Kale A. A Giant Vulvar Mass: A Case Study of Cellular Angiofibroma. Case Rep Obstet Gynecol. 2016; 2016: 2094818. [DOI:10.1155/2016/2094818].

2. Nucci MR, Granter SR, Fletcher CDM. Cellular Angiofibroma: A Benign Neoplasm Distinct from Angiomyofibroblastoma and Spindle Cell Lipoma. Am J Surg Pathol. 1997; 21(6): 636-644.

3. Canales BK, Weiland D, Hoffman N, et al. Angiomyofibroblastoma-like tumors (cellular angiofibroma). Int J Urol Off J Jpn Urol Assoc. 2006; 13(2): 177-179. [DOI:10.1111/j.1442-2042.2006.01255.x].

4. Koo PJ, Goykhman I, Lembert L, Nunes LW. MRI features of cellular angiomyofibroma with pathologic correlation. J Magn Reson Imaging JMRI. 2009; 29(5): 1195-1198. [DOI:10.1002/jmri.21747].

5. Fletcher CDM, Unni K, Mertens F. World Health Organization Classification of Tumours. Pathology and Genetics of Tumours of Soft Tissue and Bone. IARC Press; 2002. Accessed October 10, 2021.

6. Kumari R, Tandup C, Agarwal A, Chowdhury A. Rare case of angiofibroma presenting as perineal hernia. BMJ Case Rep CP. 2021; 14(1). [DOI:10.1136/bcr-2020-236661].

7. Dizdar SK, Salepci E, Hascicek SO, Turgut S. Giant Cell Angiofibroma in Sublingual Area: A Case Report and Review of Literature. Med Bull Sisli Etfal Hosp. 2021; 55(1): 134-137. [DOI:10.14744/SEMB.2019.55452].

8. Mandato VD, Santagni S, Cavazza A, Aguzzoli L, Abrate M, La Sala GB. Cellular angiofibroma in women: a review of the literature. Diagn Pathol. 2015; 10: 114. [DOI:10.1186/s13000-015-0361-6].

9. Laskin WB, Fetsch JF, Mostofi FK. Angiomyofibroblastomalike tumor of the male genital tract: analysis of 11 cases with comparison to female angiomyofibroblastoma and spindle cell lipoma. Am J Surg Pathol. 1998; 22(1): 6-16. [DOI:10.1097/00000478-199801000-00002].

10. Kamitani R, Matsumoto K, Fujiwara S, et al. A case of inguinal cellular angiofibroma. IJU Case Rep. 2020; 3(2): 69-71. [DOI:10.1002/iju5.12147].

11. Emtage JB, Parker J, Marcet JE, Finan J, Lockhart JL, Hernandez DJ. A large cellular angiofibroma of the male pelvis presenting with obstructive voiding: A case report and review of the literature. Can Urol Assoc J J Assoc Urol Can. 2013; 7(5-6): E373-375. [DOI:10.5489/cuaj.1222].

12. Kumar P, Singh S, Kumar A, Janoria S. A rare case of cellular angiofibroma affecting the periurethral region in a 38-year-old woman. Case Rep. 2018; 2018. [DOI:10.1136/bcr-2018-225386].

13. Qiu P, Wang Z, Li Y, Cui G. Giant pelvic angiomyofibroblastoma: case report and literature review. Diagn Pathol. 2014; 9: 106. [DOI:10.1186/1746-1596-9-106].

14. Wyn I, Debiec-Rychter M, Van Cleynenbreugel B, Sciot R. Cellular Angiofibroma of the Prostate: A Rare Tumor in an Unusual Location. Case Rep Pathol. 2014; 2014: e871530. [DOI:10.1155/2014/871530].

15. Mathur M, Spektor M. MR Imaging of the Testicular and Extratesticular Tumors: When Do We Need? Magn Reson Imaging Clin N Am. 2019; 27(1): 151-171. [DOI:10.1016/j.mric.2018.08.006].

16. Ntorkou AA, Tsili AC, Giannakis D, et al. Magnetic resonance imaging findings of cellular angiofibroma of the tunica vaginalis of the testis: a case report. J Med Case Reports. 2016; 10: 71. [DOI:10.1186/s13256-016-0861-3].

17. Kandil DH, Kida M, Laub DR, Cooper K. Sarcomatous transformation in a cellular angiofibroma: a case report. J Clin Pathol. 2009; 62(10): 945-947. [DOI:10.1136/jcp.2009.067124].

18. Iwasa Y, Fletcher CDM. Cellular angiofibroma: clinicopathologic and immunohistochemical analysis of 51 cases. Am J Surg Pathol. 2004; 28(11): 1426-1435. [DOI:10.1097/01.pas.0000138002.46650.95].

19. Arena S, Barresi V, Romeo C. Giant Angiofibroma of the Scrotum in a Child. Urology. 2016; 93: e15-16. [DOI:10.1016/j.urology.2016.03.031].