BAOJ Cancer Research & Therapy

Anurag Mehta, et al, BAOJ Cancer Res Ther 2017, 3:3
3: 040

Case Report

Primary Leiomyosarcoma of the Seminal Vesicle- a Rare Event

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Summary

Primary malignancies of the seminal vesicle are rare and early detection is a problem due to the nonspecific nature of symptoms. Leiomyosarcoma of the seminal vesicle is an extremely rare neoplasm which has a poor prognosis due to late diagnosis and difficult surgical resection. Only eight cases have been reported in indexed literature so far. Radiology can be of immense importance in determining the site of origin. Though rare, their existence must be kept in mind at the time of diagnosis and therapy, as complete surgical resection is the treatment of choice.

Abstract

Background

Soft tissue sarcomas of genitourinary tract are rare with the most common site being paratesticular region, kidney and prostate. Primary sarcomas of the seminal vesicle are extremely rare with only eight cases of primary leiomyosarcoma reported thus far in literature. These tumors are detected late due to nonspecific symptoms.

Case Presentation

Herein we report the case of a middle aged male who presented with the complaints of increased frequency and urgency of micturition along with longstanding intermittent constipation. Lab workup showed a serum PSA level of 0.53ng/L. Radiological evaluation revealed a large pelvic mass measuring 10.1 x 9.9 x 13.3 cm replacing the prostate. Surgical resection of the pelvic mass along with cystoprostatectomy and segmental resection of rectum was performed. After histopathological examination, a final diagnosis of leiomyosarcoma of the seminal vesicle was given.

Results and Conclusion

We report the ninth case of primary leiomyosarcoma of the seminal vesicle. Though rare, their existence must be kept in mind during diagnosis and therapy as radical surgical excision is the treatment of choice. Due to paucity of literature, such case reports offer the only resources for clinicians encountering them in their practice.

Key Words: Soft Tissue Sarcoma; Genitourinary; Primary Leiomyosarcoma; Seminal Vesicle; Prostate

Introduction

Soft tissue sarcomas (STSs) of genitourinary tract are rare tumors, representing about 2.1% of all sarcomas [1]. The most common site for STSs in genitourinary tract is paratesticular region, kidney and prostate [2]. Leiomyosarcomas of the seminal vesicle are extremely rare. To our knowledge, only eight cases have been reported so far in English literature [3-9]. All these cases have been summarized in (Table 1). The diagnosis of such tumors has been described as difficult due to obscuration of the primary site of origin by extensive involvement of the neighbouring organs. Scarce literature suggests that radical surgery is the treatment of choice [3-7]. Herein is reported, a case of primary leiomyosarcoma of the seminal vesicle which was clinically thought of as a prostatic tumor.

Case Presentation

A 75 year old male with no known comorbidities, presented to Rajiv Gandhi Cancer Institute and research centre (RGCIRC) , Delhi with a history of lower urinary tract symptoms (LUTS) such as increased frequency and urgency of urination. He also gave a history of constipation and bleeding per rectum for the last two years. He had undergone surgery 15 years back for haemorrhoids and was taking symptomatic medication for the same. There was no other significant medical history. The general physical and systemic examination were non contributory.

The lab workup at our hospital showed a total PSA value of 0.53ng/L. Colonoscopy revealed a normal study. Positron emission tomography-

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Sub Date: September 6th, 2017, **Acc Date:** September 20th, 2017, **Pub Date:** September 22nd, 2017.

Citation: Anurag Mehta, Prerna Chadha, Poojan Agarwal and Gurudutt Gupta (2017) Primary Leiomyosarcoma of the Seminal Vesicle- a Rare Event. BAOJ Cancer Res Ther 3: 040.

Table No. 1 Previously reported cases of leiomyosarcoma of the seminal vesicle

Researchers	Clinical Presentation	Pathological Resection Margin	Initial Treatment	Adjuvant Therapy
Schned et al[3]	Asymptomatic	R0	Surgery	Not given
Russo et al[4]	Asymptomatic	R1	Surgery	Not given
Russo et al[4]	Asymptomatic	R1	Surgery	Not given
Amirkhan et al[5]	Rectal pain	R0	Surgery	Not given
Muentener et al[6]	Urinary tract symptoms	R0	Surgery	Not given
Upreti et al[7]	Urinary tract symptoms	NA	Surgery	Not given
Agrawal et al[8]	Urinary tract & rectal symptoms	NA	Surgery	CT and RT
Cauvin et al[9]	Rectal symptoms	R1	Surgery	RT

R1:marginal; R0: wide margin; CT: chemotherapy; RT: radiotherapy; NA: not available

computed tomography (PET/CT) showed a large mildly metabolically active solid cystic mass lesion measuring approximately 10.1 x 9.9 x 13.3 cm replacing the prostate. Bilateral seminal vesicles were also not visualized separately. The mass lesion was pushing the urinary bladder anteriorly and bowel loops posteriorly. The left lung showed a mildly metabolically active pleural based nodule measuring 2.5 x 2.4 cm. Surgical resection of the pelvic mass along with cystoprostatectomy and segmental resection of the rectum was performed, and the specimens were sent to the lab.

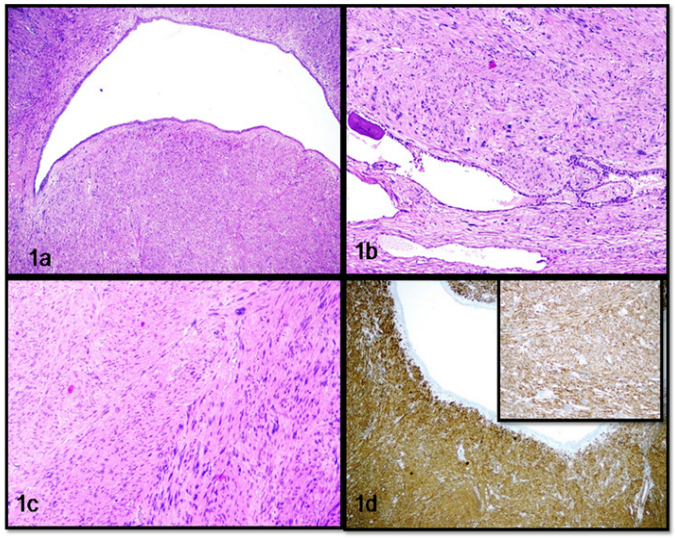
Pathological Findings

Three separately sent containers were received, labelled as a) pelvic mass? prostate; b) urinary bladder and c) rectum. The pelvic mass comprised a single piece of nodular tissue measuring 10 x 8.5 x 8 cm along with an attached fold of peritoneum measuring 8 x 5 cm. The cut surface of the mass was a fleshy grey white color having whorled appearance. Few dilated lumina were identified within the tumour largest measuring 7 cm in maximum dimension. Serial sectioning of the peritoneal fold revealed remnant seminal vesicle measuring 2 x 0.7 x 0.7 cm.

The specimen sent as "urinary bladder" comprised an unremarkable urinary bladder (measuring 10 x 8 x 3 cm) along with prostate (measuring 4 x 3x 1.8 cm). The right seminal vesicle measured 2 x 1.6 x 0.7 cm while the left seminal vesicle could not be identified. Bilateral vas were unremarkable. The cut section of prostate was unremarkable. The specimen of "rectum" comprised an unremarkable rectal segment measuring 8 cm in length. No tumor deposit was seen on gross.

Histopathology

Sections from the mass showed a well circumscribed solid tumor composed of spindle shaped cells arranged in interlacing fascicles (figure 1c). The individual tumor cells have indistinct cell borders with moderate eosinophilic cytoplasm, plump spindled to ovoid vesicular nuclei having blunt ends and conspicuous nucleoli. Paranuclear cytoplasmic vacuolation is seen . Focal areas show high grade nuclear atypia with bizarre hyperchromatic nuclei having prominent nucleoli along with multinucleate cells. A thin walled vasculature is seen interspersed throughout the tumor. Perivascular lymphomononuclear inflammation is evident .Areas of necrosis are evident. Mitosis is 3-4/10 High power field (HPF) in areas of highest cellularity. Intratumoral dilated lumina are lined by seminal vesicle lining epithelium comprising columnar cells with bland nuclei (figure 1a). Periphery of the tumor shows remnant seminal vesicle. The tumor also appears to involve prostate superficially (figure 1b).The tumor reaches upto the inked surgical margin. On immunohistochemistry (IHC) the spindled tumor cells are positive for SMA and SMMH (figure 1d) while being negative for CD21, CD23, CD34 and CD117 (figure 2b). The epithelium lined lumina express CK (figure 2a). Ki-67 labelling index is 15% in the areas of highest cellularity. A final diagnosis of leiomyosarcoma, FNCLCC grade 2, arising from the left seminal vesicle was given. Sections from the urinary bladder, rectum, right seminal vesicle and bilateral vas were free of tumor. Aside from the superficial involvement, the bulk of the prostate was unremarkable.



Legends

Fig1 Leiomyosarcoma of the seminal vesicle

1a: Dilated lumina lined by seminal vesicle lining epithelium. The muscular coat is replaced by tumor

1b: Tumor with minimal peripheral involvement of prostate

1c: Tumor arranged in intersecting fascicles

1d: Immunohistochemistry stain for SMA showing strong and diffuse positivity. Inset shows SMMH positivity

Discussion

Primary malignancies of the seminal vesicles are extremely rare while primary mesenchymal neoplasms are rarer [10]. Only 8 cases of primary seminal vesicle leiomyosarcoma have been reported in English literature so far [3-9]. This case represents the 9th case. Most of the patients have presented in the age range of 37-68 years with urinary or rectal symptoms [9]. Our patient was a 75 year old who also presented with lower urinary tract symptoms along with intermittent long standing constipation. Due to the non-specific nature of symptoms, the initial presentation can be late leading to a late diagnosis. Radiological imaging such as PET/CT and magnetic resonance imaging (MRI) can be useful to determine the organ of origin, in order to avoid an extensive resection procedure. However, if the mass is large, like in this case, imaging may not be able to locate the site of origin due to extensive involvement of the neighbouring organs.. The gross findings of an unremarkable prostate with a separately present tumor mass along with peripherally compressed seminal vesicle were of paramount importance. Definitive diagnosis of a leiomyosarcoma was done on histopathology. The differentials considered were of a leimoyosarcoma arising from adjacent organs (prostate, bladder, rectum) and secondarily invading the seminal vesicle along with prostatic stromal sarcoma, Gastrointestinal stromal tumor (GIST), Solitary fibrous tumor (SFT) and follicular dendritic cell (FDC) sarcoma. However, the epicentre of the mass and pattern of displacement of adjacent organs favoured a seminal

vesicle origin. Strong and diffuse immunohistochemical expression of SMA and SMMH, negativity for CD34, CD117, CD21 and CD23 ruled out the other differentials. Leiomyosarcoma of the seminal vesicles has a poor prognosis, possibly due to a delayed diagnosis and difficult surgical resection. The treatment of choice is complete surgical resection [8], while response to chemotherapy or radiation has not been documented due to scarce literature. However, few researchers have used adjuvant treatment in their cases [9]. In this case, the inked surgical margin was positive, and for such cases adjuvant radiotherapy or chemotherapy may be considered.

Conclusion

Herein is reported the ninth case of primary leiomyosarcoma of the seminal vesicle. Though rare, their existence must be kept in mind during diagnosis and therapy as radical surgical excision is the treatment of choice. A multimodality treatment may be considered in select patients such as those with a positive resection margin. Adjuvant radiotherapy or chemotherapy with docetaxel and gemcitabine may be of help in these patients. In view of paucity of literature, such case reports may be the only resources for clinicians who encounter them in clinical practice.

Acknowledgements

The authors thank the technical staff of the laboratory medicine department for their skilful assistance. This study did not receive any funding.

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