

Adrenal Leiomyoma: A Rare Tumor Presented as an Incidentaloma in a Patient with AIDS

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ABSTRACT

Adrenal leiomyoma is an extremely rare and benign tumor, usually presents as an incidental finding. Generally, this tumor is a non-functional large adrenal mass. The majority of leiomyomas of the adrenal gland are called as “incidentalomas” because they are found in absence of clinical symptoms and represents an unexpected finding during an abdominal ultrasound or computed tomography (CT) scan. Here we describe a 40-year-old man seropositive for the human immunodeficiency virus (HIV), under highly active antiretroviral therapy who presents a heterogeneously left adrenal mass in the context of a routine ultrasound abdominal scan. A CT abdominal scan confirms this finding. Laboratory results revealed that the tumor was non-functional. Because the possibility of adrenal carcinoma, the patient underwent a successful left adrenalectomy. Histopathology examination and immunohistochemistry techniques confirm the diagnosis of leiomyoma. Adrenal leiomyoma should be included in the differential diagnosis of unilateral, non-functional adrenal tumors in HIV patients.

KEYWORDS

Adrenal Leiomyoma; Incidental Finding; HIV; AIDS.

INTRODUCTION

Incidental adrenal tumors are often diagnosed during abdominal ultrasonography or computerized tomography (CT) studies for other pathologies. In human immunodeficiency virus (HIV) infected patients, leiomyomas are described with more frequency than in the general population. Also, in AIDS patients, these tumors have been well described in association with Epstein-Barr virus (EBV) infection [1].

We report the case of an HIV-seropositive male with an inci

idental adrenal mass that was diagnosed as a leiomyoma on histopathological and immunohistochemical (IHQ) studies. A literature review on adrenal leiomyoma was also made, including Pub Med, Medline and Embase for articles published until December 2015.

MATERIAL AND METHODS

A 40-year-old HIV seropositive man with diagnosis of AIDS

since 2012 account of his history of toxoplasmic encephalitis. He received a complete antiparasitic treatment and after, he was started on antiretroviral therapy. In this moment, the CD4 T-cell count was 11 cell/ μ L and the plasma viral load was 46 120 copies/mL (\log_{10} 4, 70). Two years later, in a routine medical examination, he referred a colic pain on the left lumbar fossa. An abdominal ultrasound scan showed a large size of the left adrenal gland. A computed tomography scan of the abdomen revealed an enlarging mass of 43 x 57 mm on the left adrenal gland (Figure 1).

Figure 1: Computed tomography of abdomen showing a large left-sided adrenal mass.

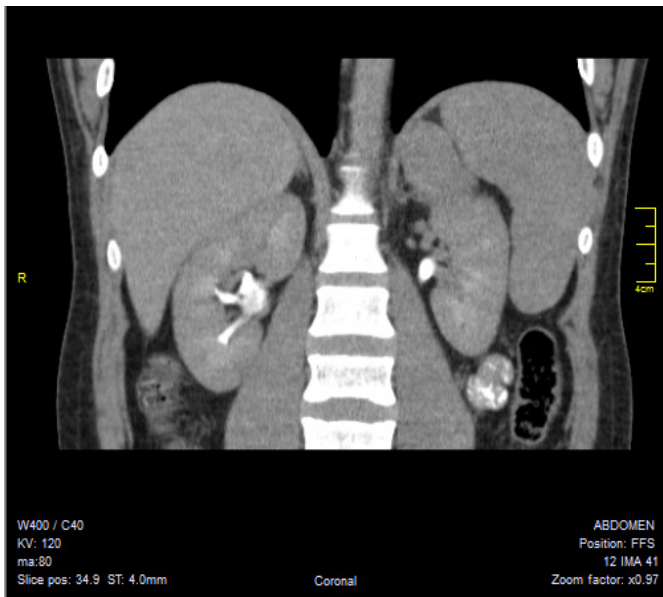
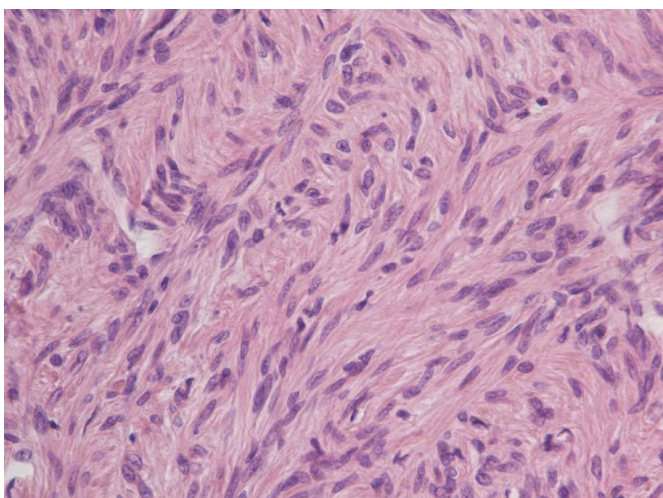


Figure 2: H-E 40X: Histopathology of the surgical piece showing a fusocellular proliferation of bland spindle cells with cigar-shaped nuclei, eosinophilic cytoplasm and areas of hyalinosis, compatible with smooth muscle tumor.

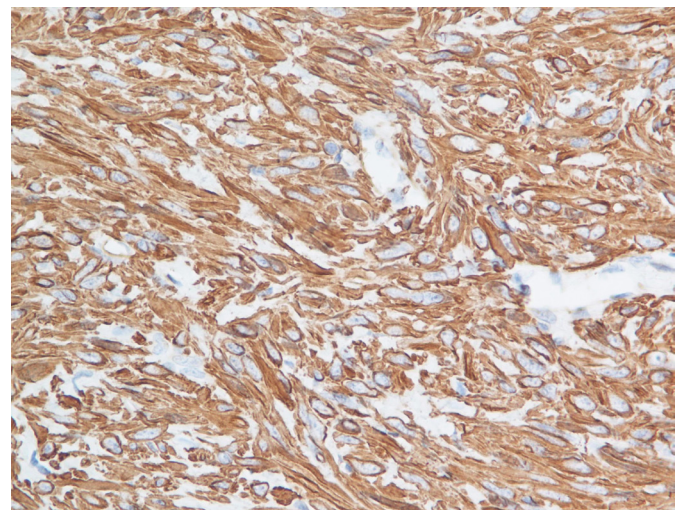


Functional tests including a serum cortisol level at 08:00 h and at late-night were normal and ruled out the presence of a cortisol-producing adenoma. The patient was normotensive repeatedly during admission to exclude an asymptomatic

pheochromocytoma. The patient underwent a left adrenalectomy because of the size of the lesion and the radiological appearance; macroscopic pathological examination of the surgical piece showed a well-circumscribed and encapsulated large mass; histopathological analysis revealed the smooth muscle origin of the tumor.

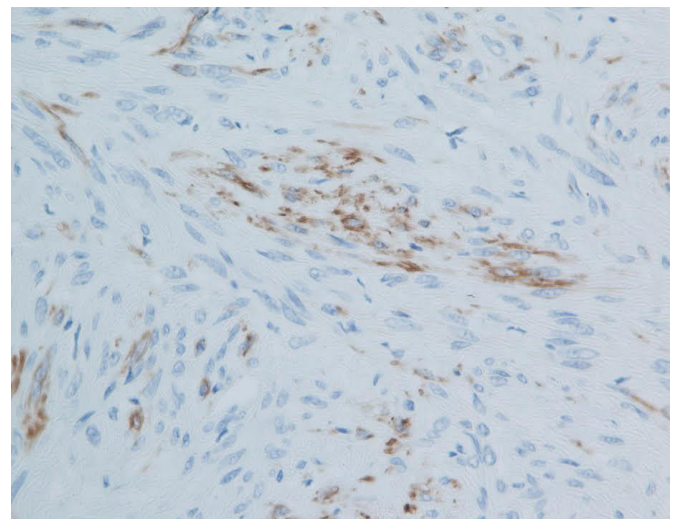
Histologic feature includes a mesenchymal tumor characterized by the fusocellular proliferation of spindle cells with cigar-shaped nuclei, eosinophilic cytoplasm with areas of hyalinosis, compatible with the diagnosis of smooth muscle tumor (Figure 2).

Figure 3: The tumor cells showing immunoreactive for specific anti-smooth muscle actin.



Diagnosis was confirmed by a positive immunostaining for smooth muscle actin. The tumor cells showed strongly immunoreactive for specific antismooth muscle actin and partial desmin expression (Figure 3 and 4)

Figure 4: Partial immunoreactive for desmin with diffuse cytoplasmic staining within the tumor cells.



With no expression of CD34, CD31, S-100, LANA and keratin,

consistent with the histopathological diagnosis of leiomyoma.

The patient’s postoperative course was normal and he was discharged two weeks after in a good clinical condition.

RESULTS AND DISCUSSION

Leiomyomas of the adrenal glands are very rare tumors except in AIDS population. They are benign tumors composed of smooth muscle cells. An adrenal incidentaloma is defined as a clinically unapparent adrenal tumor, generally of 1 cm or more, that is discovered during radiological studies performed for other causes [2, 3]. Adrenal incidentalomas are diagnosed in approximately 1% of abdominal CT scans [4]. Also, in autopsy studies, asymptomatic adrenal masses are discovered in up to 9% of patients [5]. Diagnosis criteria for adrenal incidentaloma have been well established and include test to determine the functional tumor status and the histopathology [6]. Adrenal leiomyomas have a wide range of age at presentation, with a median of 38 years and female predominance (66, 6%). These tumors are typically an adrenal unilateral solitary mass lesion with a median size of 5 cm [7]. The widespread in

the use of modern imaging techniques is associated with an increased frequency of adrenal incidentaloma. The majority of these masses are adrenal cortical tumors (adenomas or carcinomas) or adrenal medullary tumors (pheochromocytomas) [7]. The most common sites of leiomyomas are the uterus and the gastrointestinal tract [8]. Adrenal leiomyomas are rare tumors and only a few cases have been reported in the medical literature. Until December 2015, only 19th cases of adrenal leiomyomas have been reported; only 8 of them affected HIV infected patients [7] (Table 1). In this aspect, infection with HIV facilitates the development of smooth muscle tumors [9].

EBV have been well documented in association with AIDS-related smooth muscle tumors, especially in pediatric population and HIV seropositive patients. The EBV genome have been detected by polymerase chain reaction (PCR) in biopsy smears, especially in children [10].

Adrenal leiomyoma should be included in the differential diagnosis of unilateral non-functional and incidental finding in HIV/AIDS patients [11-16].

Table 1: Epidemiological and clinical characteristics of patients with adrenal leiomyoma reported in the medical literature.

Case No	Reported cases	Age (years)	Sex	Tumor characteristics		HIV status	EBV status
				Functional	Size (mm)		
1	Lin et al [8]	31	Female	No	110 x 90 x 70	Positive	Negative
2	Nishida et al (11)	48	Female	No	55 x 45 x 50	Negative	Negative
3	Alteer et al (3)	40	Male	No	49 x 55 x 60	Positive	Positive
4	Chang et al (12)	53	Female	No	55 x 45 x 35	Positive	Negative
5	Radin et al (13)	28	Female	NA	30	Positive	Negative
6	Jimenez-Heffeman et al (10)	2	Male	NA	70 x 50 x 50	Positive	Positive
7	Mouchet et al (14)	10	Female	NA	50 x 40 x 30	Negative	Negative
8	Gibbs et al (1)	49	Female	No	30 x 35 x 20	Negative	Negative
9	Jacobs et al (5)	65	Female	No	50 x 31 x 42	Negative	Negative
10	Parola et al (15)	35	Female	NA	35	Positive	Negative
11	Goldman et al (16)	72	Male	No	90 x 70 x 60	Negative	Negative
12	Dahan et al (17)	32	Male	No	30	Positive	Negative
13	Rosenfeld et al (18)	11	Female	NA	L: 50, R: 30	Positive	Negative
14	Demirel et al (19)	15	Male	No	L: 40 x 50 x 35	Negative	Negative
					R: 80 x 50 x 30		
15	Chao et al (20)	40	Female	No	NA	Negative	Negative
16	Jurczak et al (21)	56	Male	No	72	Negative	Negative
17	Al-Masri et al (22)	38	Female	NA	NA	Negative	Negative
18	Parelkar et al (23)	11	Female	No	L: 43 x 35 x 30	Negative	Negative
					R: 90 x 70 x 40		
19	Meher et al (7)	42	Female	No	120 x 100 x 80	Negative	Negative
20	Our report	40	Male	No	100 x 60	Positive	NA

* Modified of Meher D et al [7].

NA: Not available

Conflicts of Interests: The authors declare that there is no conflict of interest.

Patient Consent: Written informed consent was obtained from the patient prior to publication this manuscript [17-23].

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