

## PRIMARY OVARIAN LEIOMYOMA: A CASE REPORT

Benzina Intissar\*<sup>1</sup>, Talib Sarah<sup>1</sup>, Edahri Yassine<sup>2</sup>, Amina Lakhdar<sup>2</sup> and Kharbach Aicha<sup>1</sup><sup>1</sup>Department of Gynecology and Endocrinology, Maternity Souissi, University Hospital Center Ibn Sina, University Mohammed V, Rabat, Morocco.<sup>2</sup>Department of Gynecology and Endocopy, Maternity Souissi, University Hospital Center Ibn Sina, University Mohammed V, Rabat, Morocco.

\*Corresponding Author: Benzina Intissar

Department of Gynecology and Endocrinology, Maternity Souissi, University Hospital Center Ibn Sina, University Mohammed V, Rabat, Morocco.

Article Received on 02/03/2021

Article Revised on 22/03/2021

Article Accepted on 12/04/2021

## ABSTRACT

**Introduction:** Primary ovarian leiomyoma is a rare benign tumor of the ovary seen in women between 20 and 65 years old. It is usually diagnosed incidentally but their most clinical manifestations are abdominal pain and palpable mass, and it is mostly misdiagnosed as malignant tumor. **Case:** A 40-years-old female patient with history of 5 pregnancies, 2 parities, 3 abortions, pancreatic cancer in her father, presented chronic pelvic pain. Transvaginal ultrasonography found a right adnexial mass. She underwent a laparotomy which revealed un ascites. Unilateral ovariectomy was performed. Pathological examination revealed an ovarian leiomyoma. There was no recurrence after 6 months' followup. **Conclusion:** This rare tumor of the ovary should be considered in the differential diagnosis of solid ovarian masses. An immunohistochemical analysis may be useful for definitive diagnosis.

**KEYWORDS:** Ovary, Leiomyoma, Pathological examination, Immunohistochemistry.

## INTRODUCTION

Leiomyoma is one of the rarest solid tumors of the ovary, which occurs in 20 to 65-year-old women. It accounts for 0.5–1% of all the benign ovarian tumours.<sup>[1]</sup> Most of them are small and unilateral. The majority of these tumors are discovered incidentally, with about 80% of the cases occurring in premenopausal women.<sup>[2]</sup> Patients are usually asymptomatic, and the tumor is most commonly diagnosed unintentionally by histological examination of ovarian tissue after an ovariectomy for solid ovarian mass. The probably origin of these smooth muscle include ovarian hilar blood vessels, but other possible origins are cells in the ovarian ligament, smooth muscle cells or multipotential cells in the ovarian stroma, undifferentiated germ cells, and cortical smooth muscle metaplasia.<sup>[3,4]</sup> We report a case of a primary ovarian leiomyoma and discuss its diagnostic and therapeutic features.

## Case

The patient is a 40 years old woman, having as a background a deceased father by neo pancreas, 5 pregnancies, 2 parities, 2 children, 3 abortions, admitted to our university hospital (Ibn Sina, Rabat, Morocco) with a history of chronic pelvic pain evolving for two years.

Abdominal examination revealed the presence of sensitivity in the right iliac fossa, on vaginal examination; a mobile mass was detected in the right lower abdomen. Vital signs were normal.

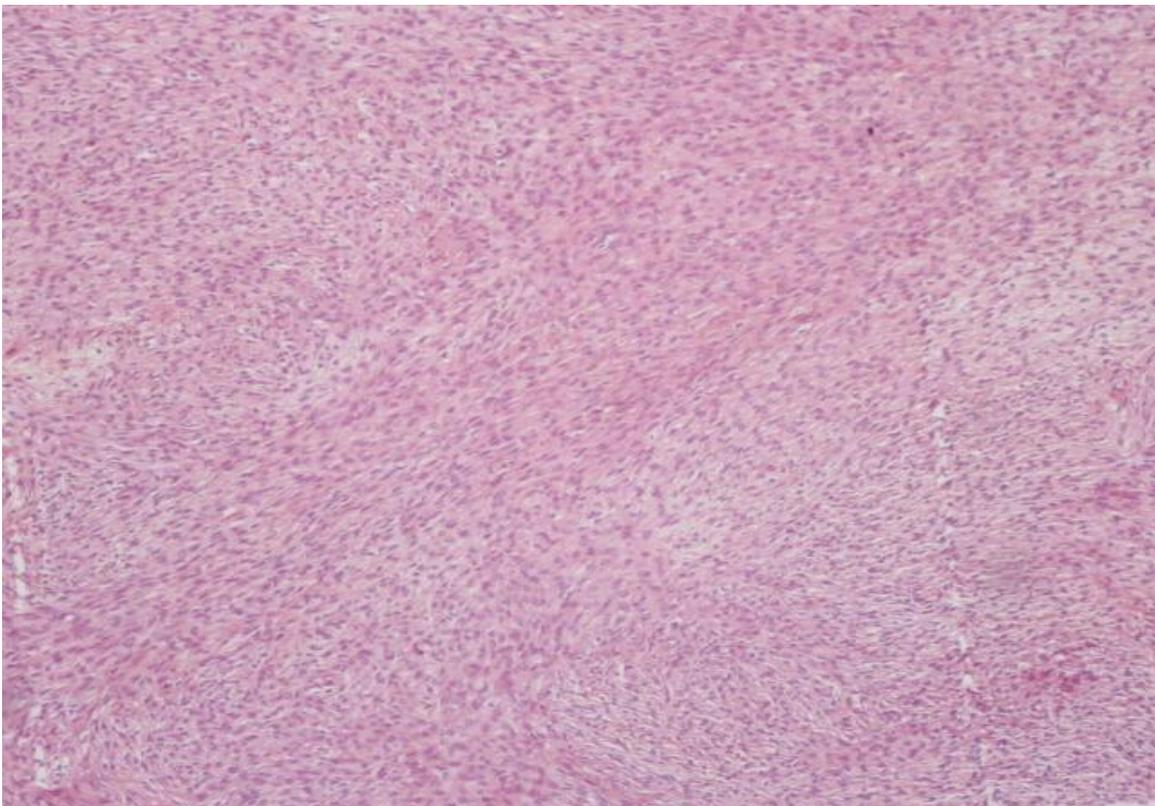
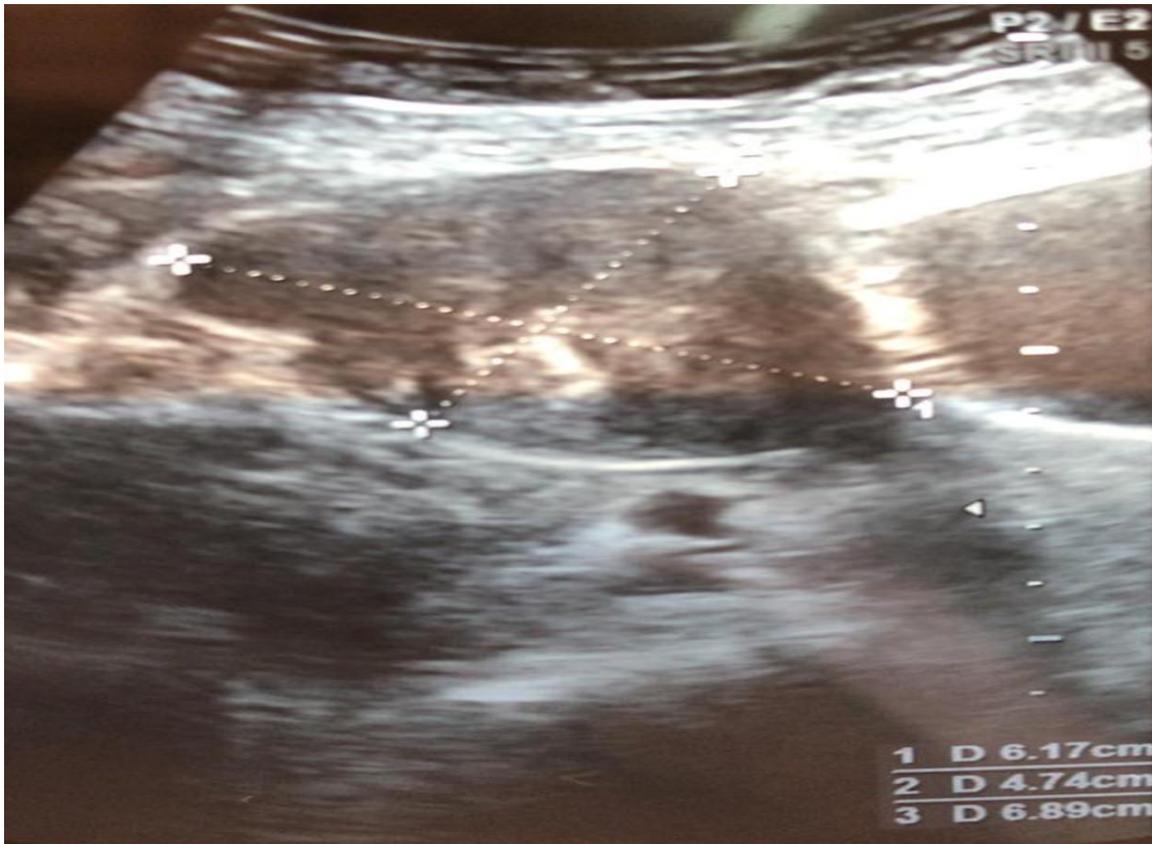
Tumor markers (AFP, CA125, CEA, and HE4) were all in the normal range.

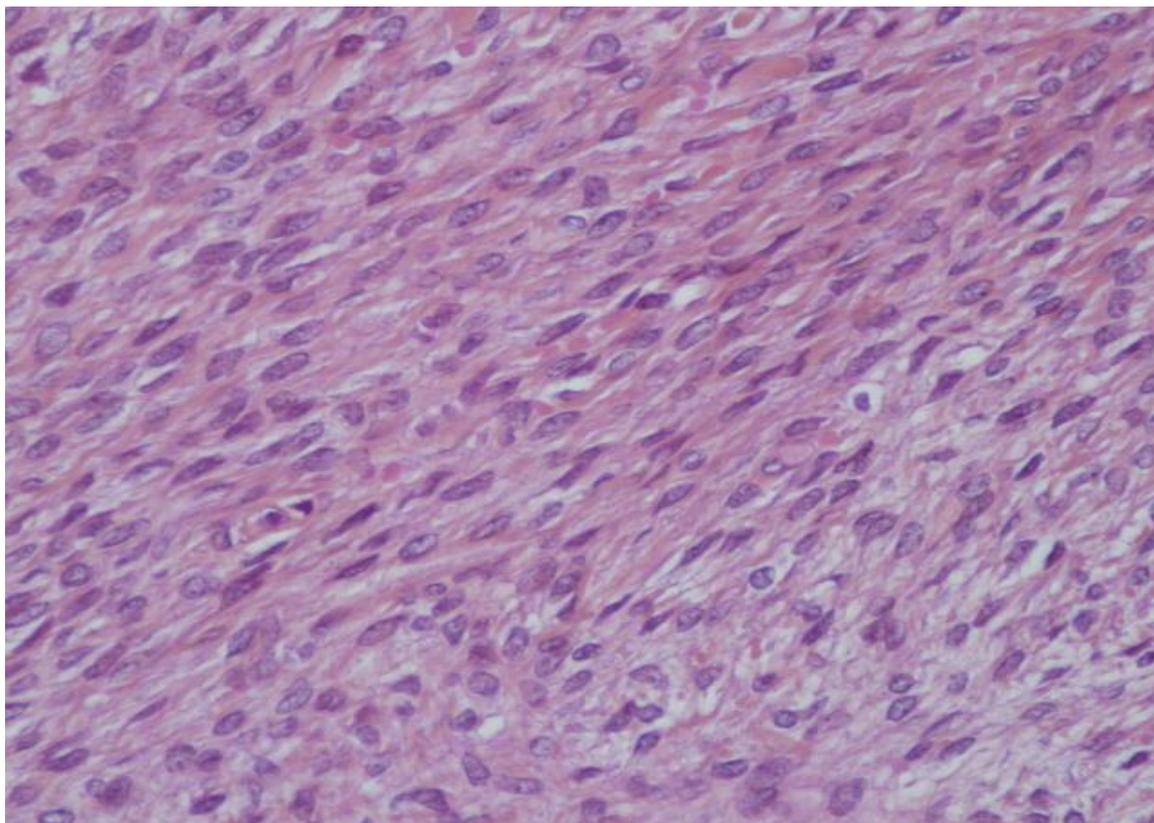
Pelvic ultrasonography revealed an inhomogeneous solid left ovarian mass with a dense content and regular contours measuring 69.62.47 mm, the right ovary was normal. The anteverted, pre-reflected uterus, measuring 87.58.50 mm, presents a regular contours and homogeneous echostructure. Pathologic blood flow was not detected in Doppler ultrasonography (**figure1**).

A laparotomy was performed under general anesthesia and revealed firstly ascites and secondly a solid, firm, irregular right-sided ovarian tumor with a smooth surface measuring 65.50.40 mm in diameter. The tumor was distinctly separated from the uterus and exhibited no adhesion to or infiltration of the surrounding structures. The left adnexa was normal. Right oophorectomy was done, postoperative recoveries were smooth and the patient was discharged home after 4 days after the operation.

Pathologic examination of the surgical specimen number 181105825 macroscopically showed a smooth external surface, its sectional slice shows a gray-white, homogenous appearance and of firm consistency and

microscopically a proliferation of fusiform cells arranged in intersecting bundles and not showing any Cytonuclear atypias corresponding to a fibrothecoma. (figure 2,3).





The ascites fluid showed no suspicious cells.

Postoperative follow-up performed at 3 months, 6 months was normal.

## DISCUSSION

Primary ovarian leiomyoma is a very rare benign tumor, that is one of extra-uterine localisations of leiomyoma which were reported in few cases: vulva,<sup>[5]</sup> urethra, retroperitoneum,<sup>[6]</sup> bladder, etc. The majority of these

tumors are discovered incidentally during routine pelvic examination, during surgery, or after ovariectomy. Most of them are small, usually less than 3 cm in diameter.<sup>[7]</sup> Most of the patients are asymptomatic or, as in our case, have only complaints of lower abdominal pain. In contrast, giant ovarian leiomyomas can be presented with ascite, hydrothorax, hydronephrosis, or slightly elevated levels of tumor marker, CA 125,<sup>[1,4,8]</sup> in our case, it is presented with ascites.

They are usually unilateral, but bilateral ovarian leiomyomas have been reported in female aged between 16 and 25 years.<sup>[9]</sup> Our patient was 40 years old and had unilateral ovarian leiomyoma. Ovarian leiomyomas often coexist with uterine leiomyomas, but sometimes they can be a secondary origin. That is, subserosal pedunculated uterine leiomyoma can lose its attachment to the uterus and connect to the ovary. There was no associated uterine leiomyomas in our case.

Oestrogen may play a role in the development of ovarian leiomyomas. Another possible mechanism suggests that tumor may arise in developmentally abnormal ovaries.<sup>[10]</sup> In our case, normal ovarian tissue was present histologically. This suggests that the tumor can originate from the smooth muscle cells in the walls of blood vessels, in the cortical stroma, in the hilus, in the corpus luteum, or in the ovarian ligament.

Differential diagnosis can be difficult from ovarian thecoma and/or fibromas of the ovary. Because ultrasonography visualizes ovarian leiomyomas as intrapelvic solid tumors, isoechoic with the myometrium, it is difficult to distinguish them from pedunculated uterine leiomyomas and other solid ovarian tumors. MRI is often a useful adjunct to ultrasonography for the purpose of diagnosing indistinct pelvic masses.<sup>[11]</sup>

In addition to ultrasound, ascites puncture and cytological analysis are an integral part of the diagnostic approach. The non-hemorrhagic character of ascites and the absence of neoplastic cells in the sample (as is the case in our observation) contribute to reinforcing the suspicion of a benign tumor, but the sensitivity of this cytological examination is only 70%.<sup>[12]</sup>

It remains to discuss the contribution of tumor markers. The increase in CA 125 in ovarian fibroma was reported for the first time by O'Connell in 1987 and all cases subsequently published were associated with an increase in CA 125 with an average of 1,200 IU / l and extremes of 286 IU / l and 2,360 IU / l. In general, the rates

recorded in ovarian fibroma are much higher than those observed in ovarian cancers.<sup>[12]</sup>

However, this is not a good screening test because CA 125 can also be increased in other pathological situations.<sup>[12]</sup>

To confirm diagnosis of an ovarian leiomyoma, identification of the smooth muscle nature of the tumor is required. In the present case, pathologic examination revealed fibrothecoma. The diffuse strong positive staining for SMA is characteristic of leiomyoma. Thecoma could also be considered in differential diagnosis, but it does not express SMA.<sup>[1,3,7]</sup> Ovarian leiomyomas must also be differentiated from leiomyosarcomas. For this aim, pathologists use criteria such as mitotic count, cytological atypia, and tumor necrosis.<sup>[3]</sup> In our case, none of these criteria were detected.

Most of the patients undergone a salpingo-oophorectomy or an oophorectomy with or without hysterectomy despite the young age of affliction.<sup>[13]</sup> In the present case, we performed unilateral oophorectomy. Because the tumor was indistinguishable from the ovary, the tumor was also unilateral and benign.

## CONCLUSION

Ovarian leiomyoma is a rare ovarian tumor. This tumor is difficult to be diagnosed by preoperative imaging examination. The choice treatment is ovariectomy with intraoperative frozen section, pathology and immunohistochemistry can be used for diagnosis. The prognosis of this disease is good.

## Declarations

**Consent for publication:** Written informed consent was obtained from the patient for publication of this case report and any accompanying images.

**Competing interests:** The authors declare that they have no competing interests.

**Funding:** No funding to declare.

**Authors' contributions:** CE wrote the paper. SA, AN, AL,NZ, AB contributed by correction of this paper. All the authors read and approved the manuscript.

## REFERENCES

1. Doss BJ, Wanek SM, Jacques SM. Ovarian leiomyomas: clinicopathologic features in fifteen cases. *Int J Gynecol Pathol*, 1999; 18: 63–8.
2. Blue N.R, Felix J.C, Jaque J. Primary ovarian leiomyoma in a premenarchal adolescent: first reported case. *J Pediatr Adolesc Gynecol*. 2013. pii:S1083-3188(13)00242-8.
3. Tomas D, Lenicek T, Tuckar N, Puljiz Z, Ledinsky M, Kruslin B. Primary ovarian leiomyoma associated with endometriotic cyst presenting with symptoms of acute appendicitis: a case report. *Diagn Pathol*, 2009; 4: 25.
4. Mine Islimye T, Emine O, Fatma Y, Necmettin O, Umit I. Primary ovarian leiomyoma: A case report. *Int J Surg Case Rep.*, 2014; 5(10): 665–668.
5. Ammouri S, Elkarkri C, Zerai N, Lakhdar A, Baydada A, Vulvar leiomyoma: a case report, *Pan African Medical Journal*, 2019; 32: 208.
6. Ziouziou I, Bennani H, Zouaidia F, El Ghaouti M, Haddan A, Mahassini N, Karmouni T, El Khader K, Koutani A, Iben Attya Andaloussi A. Retroperitoneal leiomyoma: a case report. *Prog Urol*, 2014 Apr; 24(5): 262-5. Epub 2013 Jul 30. PubMed.
7. Erdemoglu E, Kamaci M, Bayram I, Güler A, Güler Sahin H. Primary giant leiomyoma of the ovary: case report. *Eur J Gynaecol Oncol*, 2006; 27(6): 634–635.
8. Güney M, Ozsoy M, Oral B, Mungan T, Kapucuoğlu N. Unilateral primary ovarian leiomyoma in adolescent: a case report. *Arch Gynecol Obstet*, 2007; 275(June (6): 507–510.
9. Wei C, Lilic N, Shorter N, Garrow E. Primary ovarian leiomyoma: a rare cause of ovarian tumor in adolescence. *J Pediatr Adolesc Gynecol*, 2008; 21(1): 33–36.
10. Lim S.C, Jeon H.J. Bilateral primary ovarian leiomyoma in a young woman: case report and literature review. *Gynecol Oncol*, 2004; 95(3): 733–735.
11. Yasushi K, Noriyuki T, Masako S, Kaei N, Isao M. Magnetic resonance imaging findings in leiomyoma of the ovary: a case report. *Arch Gynecol Obstet*, 2005; 273: 298–300.
12. Cissé C.T, Ngom P.M, Sangare M, Ndong M, Moreau J. Fibrome de l'ovaire associé à un syndrome de Demons-Meigs et à une élévation du CA 125. *Journal de Gynécologie Obstétrique et Biologie de la Reproduction*, 2004; 33(3): 251-254.
13. Parvin R, Mohsen H, Marzie B, and Farnoosh M. Large ovarian leiomyoma in young woman. *Adv Biomed Res.*, 2014; 3: 88.10.4103/2277-9175.128001.