Primary ovarian leiomyoma of the ovary is an extremely rare tumor; only approximately 70 cases have been reported worldwide [1]. The lesions are usually found incidentally, with the majority being discovered in perimenopausal and postmenopausal women (85%) [2,3]. They are difficult to diagnose preoperatively based on ultrasonographic results, because they are usually small in size (<3 cm in diameter) and rarely induce serious clinical symptoms [1,3]. We present a case of asymptomatic primary leiomyoma of the left ovary of a pregnant woman.

A 41-year-old woman, gravida 3, para 1, ectopic 1, had a history of a right tubal ectopic pregnancy, and a left ovarian echogenic mass measuring about 3.5 × 2.4 × 2.5 cm was observed using ultrasonography 1 year prior to this presentation. She received medical treatment with two doses of methotrexate, and the ectopic pregnancy had terminated at the 6-week follow-up examination (tested by β-hCG). Follow-up laboratory test results showed that CA-125 was within the reference range limits. Intrauterine pregnancy was noted 1 year after the ectopic pregnancy. The patient underwent an uneventful routine prenatal examination.

On acute severe variable fetal distress during labor, the patient received emergency cesarean section and was delivered of a female infant weighing 2,930 g. During this procedure, the opportunity was taken to review the left ovary, and a fasciculated grey-white left ovarian tumor that was firm and solid measuring 4.5 × 4.4 × 3.2 cm, suggesting fibroma/thercoma or leiomyoma, was discovered, and a left oophorectomy was performed. No ascites fluid was noted in the abdomen. Following meticulous examination of the uterus, three small intramural leiomyomas of approximately 1 cm in diameter, were further discovered. The patient was discharged 5 days after surgery, and her recovery period was uneventful. The final histopathologic examination revealed ovarian leiomyoma. The mass consisted mostly of interacting bundles of smooth muscles (Figure 1) and a thin layer of ovarian tissue over the cortex (Figure 2). Hemorrhage with focal infarction and hyalinization degeneration were noted (Figure 3). Immunohistochemically, these cells proved positive for desmin and the smooth-muscle origin of the tumor (Figure 4). Rare mitoses were seen.

Ovarian leiomyoma was first described in 1862, and a giant ovarian leiomyoma measuring 24 × 20 × 14.5 cm was reported in 1899 [3]. Clinically, most of the patients were asymptomatic, and the tumors were discovered incidentally or following mild complaints of lower abdominal pain because the majority of the tumors measured <3 cm in diameter [3]. However, Meigs syndrome secondary to ascites [4–6],
Hydroureteronephrosis due to the larger size of some tumors [6,7], hydrothorax [5], or acute symptoms due to torsion or necrosis [8] were the reported symptoms in some cases. Most ovarian tumors are solid, but secondary degeneration changes due to hemorrhage, hyalinization, calcification, and cyst formation may occur to some extent in the more common uterine leiomyomas [2,3,8,9]. Ovarian leiomyoma is often found in perimenopausal and postmenopausal women; however, in our reported case, the small echogenic tumor was found incidentally on ultrasonography during an ectopic pregnancy. The tumor pathology revealed spindle smooth muscle cells with interacting bundle ends, thinning of the ovarian tissue with stretched corpus luteum, with some degree of degeneration by hemorrhage and hyalinization, which may be related to the pregnancy.

Uterine leiomyomas are very common. It is important to distinguish between primary ovarian leiomyomas and parasitic leiomyomas (pedunculated subserous leiomyomas), which become attached to the ovary after detaching from the uterus. Ovarian leiomyomas must also be distinguished from leiomyomatosis peritonealis disseminata, which may be multiple and situated at the surface of the ovary [2,10] and also from intravascular extensions, which may be present loosely attached in the lumina of the ovarian hilar vessels [11]. Primary ovarian leiomyomas also have a number of features in common with sex-cord stromal tumors such as fibroma and thecoma. In practical terms, differentiation is possible by staining for α-smooth muscle actin (SMA); where thecoma does not stain intensively but both leiomyomas and fibroma stain strongly positively. Fibroma may be distinguished by testing for immunoreactivity using anti-α-inhibin and anti-SMA antibodies with which a leiomyoma will test negative [12]. Leiomyomas also need to be distinguished from leiomyosarcomas, diagnosis of which is established by the presence of four or more mitoses per 10 high-power fields for the malignant counterpart [13]. Hence, fibroma and thecoma of the ovary, parasitic leiomyoma, intravenous leiomyomatosis, leiomyomatosis peritonealis disseminata, and leiomyosarcomas should all be considered in the differential diagnosis [13].

Several theories have been proposed regarding the potential origin of these tumors. One researcher proposed that the origin is from an area of the endometriosis of the ovary, which apparently contains occasional smooth muscle cells; however, most cases were not associated with endometriosis [14]. Another researcher
proposed that the undifferentiated germ cells in the ovarian stroma might be stimulated to differentiate into smooth muscle with ultimate tumor formation [15]. However, the consensus of most recently reported research is that primary ovarian leiomyomas originate from the walls of blood vessels in the ovarian hilus or from the smooth muscle fibers near the attachment of the ovarian ligament [3–5,8,16].

Four cases have been reported in pregnant women. Moore was the first author to publish a case of leiomyoma of the ovary of a pregnant woman [17]. Zorlu et al reported a request for the termination of a pregnancy at 6 weeks of gestation and incidentally found a solid primary ovarian leiomyoma [18]. However, in both of these reported cases, no information was available about the effects of the gravid state on the tumor or vice versa [16, 18]. Kohno et al presented a 32-year-old woman who had a large pelvic mass and rapid growth noted by the 16th week of gestation. A subsequent laparotomy at the 20th week of gestation found the mass to weigh 11 kg and measure 23 × 23 × 20 cm, and the left ovarian leiomyoma contained extensive degeneration with hyalinization and edema [9]. Our case exhibited concomitant uterine leiomyomas, as in some cases reported previously [3]. The tumor mass of the leiomyoma enlarged from 3.5 × 2.4 × 2.5 cm to 4.5 × 4.4 × 3.2 cm over the course of 1 year, including during a full term of pregnancy. Increased concentrations of the progesterone and estrogen hormones in the peripheral blood could have been a factor in stimulating the growth of these tumors.

References