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Case Report

Primary ovarian leiomyoma with predominant cystic change

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ABSTRACT

Ovarian leiomyoma is a rare benign tumor, seen mostly in women 20-65 years old. The case of a 51-year-old woman with a large tumor in the pelvic cavity is reported. The dominant feature of the tumor was its cystic component, but an irregular, solid component was recognized along the dorsal margin of the tumor. The latter area showed signal intensity as low as muscle on T2-weighted images and significant contrast enhancement; however, it did not show restricted diffusion, which implied that it was benign. The large tumor was resected because malignancy could not be ruled out, and it was pathologically diagnosed as ovarian leiomyoma with predominant necrosis. When a low-intensity component without malignant features is seen on magnetic resonance imaging, leiomyoma should be included in the differential diagnosis despite its rare occurrence in the ovary.

Case report

A 51-year-old gravida 0 para 0 woman who had been complaining of abdominal fullness for 1 year was referred to our hospital because a large tumor was found in her pelvic cavity on ultrasound examination at a clinic. She had a history of surgery for a uterine cervical polyp. Complete blood count and biochemical tests showed no abnormal results, and levels of the tumor markers CEA, AFP, and SCC were within normal ranges. Contrast-enhanced computed tomography (CT) showed a tumor in the pelvic cavity measuring 18.5 cm in its longest diameter, and the tumor seemed to stem from the left ovary (Fig. 1a). The interior of the tumor included a predominant cystic component with coarse calcification and thin septa, and an irregular solid component was also seen along the dorsal side of the tumor (Fig. 1b). Magnetic resonance imaging (MRI) showed the cystic tumor with high intensity, in addition to numerous septum-like structures with low intensity on T2-weighted imaging (T2WI) (Fig. 2a). The dorsal solid component demonstrated significant contrast enhancement (Fig. 2b), but diffusion was not restricted (Fig. 2c). The tumor was unlikely to be malignant, although her attending doctor did not consider a specific malignant diagnosis in the differential; he was concerned about malignancy because of the large size and necrotic features of the tumor. Consequently,

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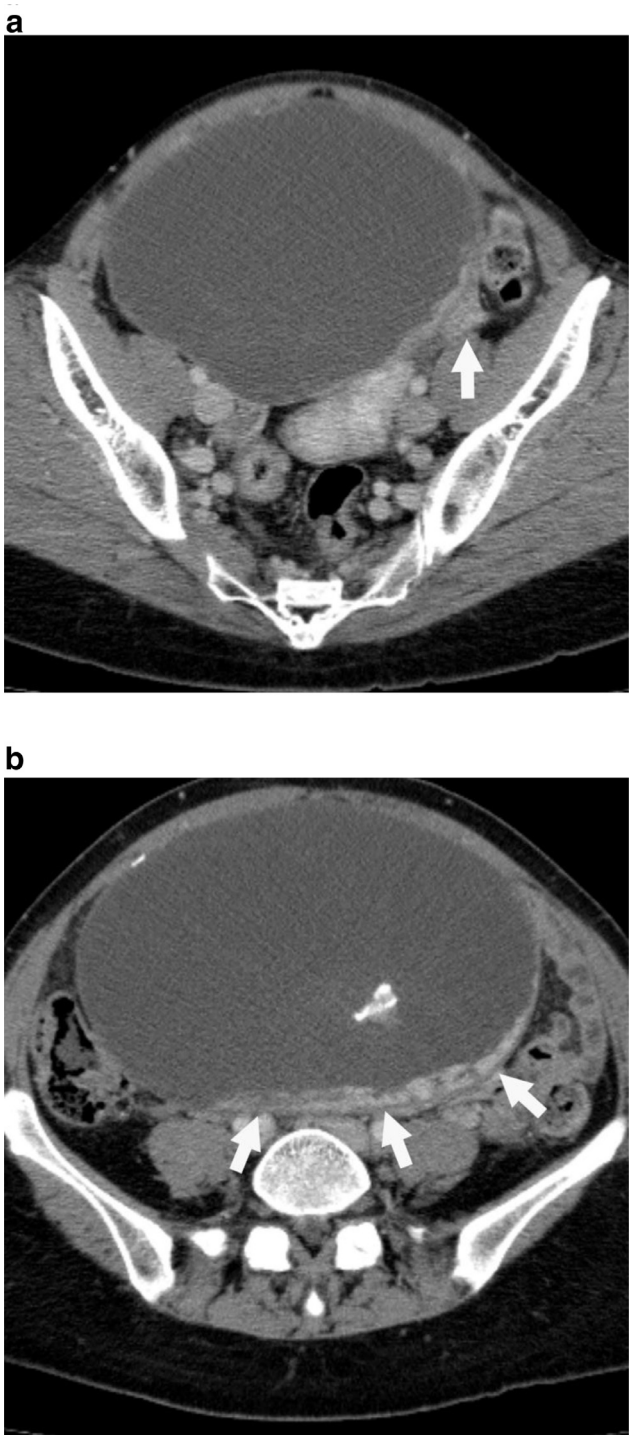

Fig. 1 – An axial abdominal contrast-enhanced computed tomography (CE-CT) scan shows a large cystic tumor. The left ovarian vein (white arrow) is recognized on the left side of the tumor, which suggests that it stems from the left ovary (a). The lower level of the CE-CT scans shows high attenuation along the dorsal side of the tumor (white arrows) (b).

the tumor was removed surgically. The intraoperative findings showed the tumor originating from the left ovary with counterclockwise rotation of its pedicle about 180°. On intraoperative rapid diagnosis, it was difficult to identify malignancy because of the vast amount of necrosis inside the tumor. Because the possibility of malignancy was deemed very low, only bilateral salpingo-oophorectomy was performed. The microscopic findings of the solid component of the dorsal part (Fig. 3a) showed proliferating spindle cells (Fig. 3b) that stained positive for alpha-SMA immunofluorescence. The histopathological diagnosis was primary ovarian leiomyoma.

Discussion

Primary ovarian leiomyoma (OL) is a very rare benign tumor that accounts for about 1% of all benign ovarian tumors, occurring mainly in 20 to 65-year-old women [1–3]. The tumor is usually small in size, less than 3 cm in diameter, and asymptomatic, typically detected during routine physical examination, surgery, or autopsy. In the present case, there was a well-circumscribed, large tumor in the pelvic cavity. The frequency of large primary OL has not been clear; however, there have been several previous cases of a large primary OL, including one with a maximum diameter of approximately 25 cm [4–6]. The tumor seems to grow rapidly during early pregnancy because estrogen might be a significant factor in the growth of OL [4,7]. Accordingly, the chances of encountering large OLs could be higher during early pregnancy. When the tumor is large, such as over 10 cm, some patients present with abdominal pain and discomfort [5,8]. Torsion could also be a complication, since an OL with the longest diameter of 14 cm was twisted [9]. Although the established management of twisted primary OL has not been reported, the tumor should be extracted, as in torsion of other types of ovarian tumors [10,11].

On CT and MRI, the tumor showed a predominant cystic component with coarse calcification and thin septa. This could be explained by degeneration of the mass, such as due to bleeding and necrosis, and, in fact, the resected tumor showed significant necrotic changes on macroscopic examination. The necrosis could have resulted from its large size and torsion of the tumor based on the intraoperative findings. The large degenerated OL showed polycystic change with some septum-like structures with low-signal intensity on T2WI [12], which was very different from the typically reported signal pattern of primary OL with a low-signal intensity on T2WI [12–14]. However, the tumor along the inner dorsal side showed low-signal intensity similar to muscle on T2WI and significant contrast enhancement. This solid component actually showed proliferating spindle cells microscopically. Even though the solid component was a minor part, it could lead to including OL in the differential diagnosis due to its typical signal intensity on T2WI.

The differential diagnosis includes the ovarian tumors with a solid component that shows low intensity on T2WI and cystic change. Ovarian fibroma and thecoma are more frequent stromal ovarian tumors showing low-signal intensity

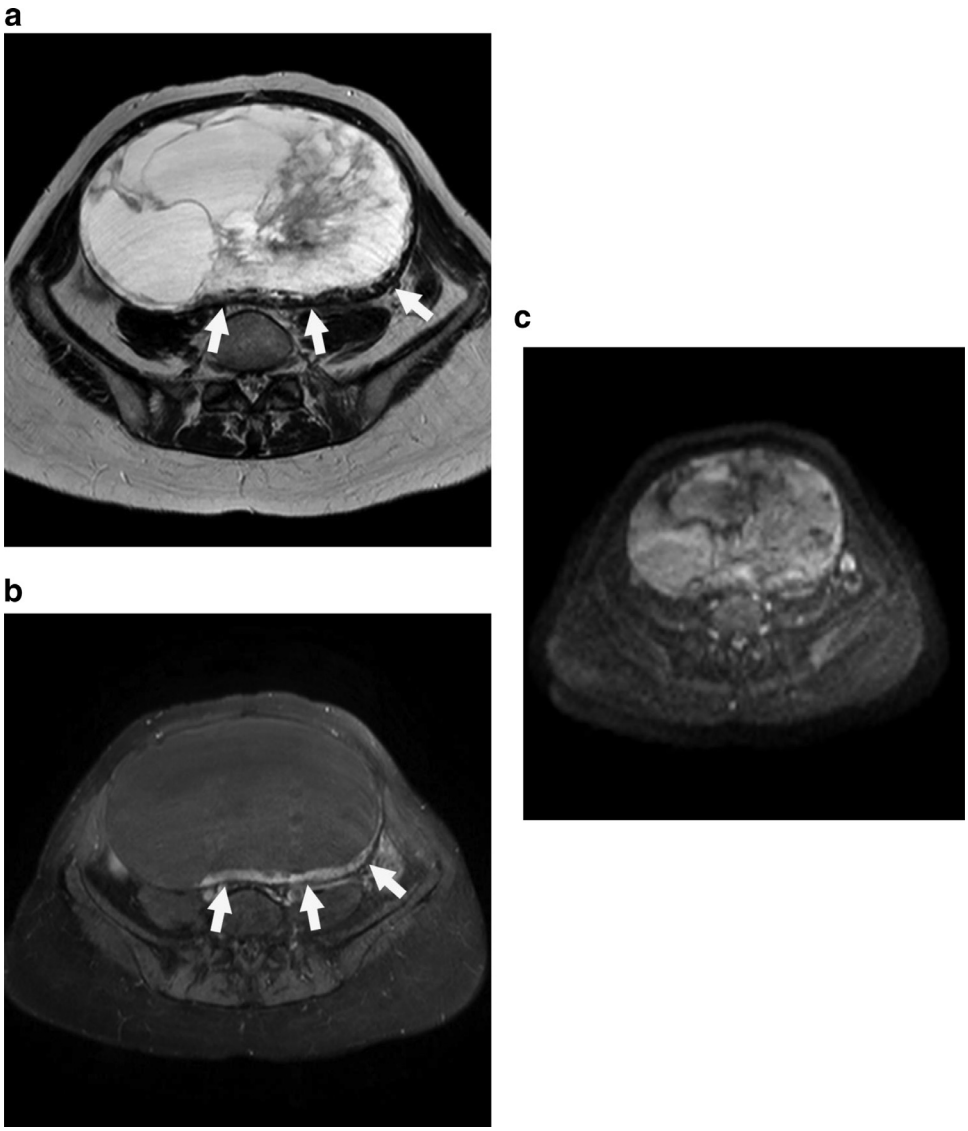

Fig. 2 – An axial magnetic resonance image shows the solid component with low-signal intensity along the dorsal side of the tumor on the T2-weighted image (white arrows) (a), which demonstrates significant contrast enhancement on the fat-saturated contrast-enhanced T1-weighted image (white arrows) (b). It does not show restricted diffusion on diffusion-weighted imaging (c).

on T2WI, which demonstrates cystic change as well [10,15]. A benign Brenner tumor also usually shows low-signal intensity on T2WI [16] and is associated with cystic change [17]. In addition, large cystic lesions of the ovary in the 50s-60s usually consist of serous cyst adenoma and mucinous cyst adenoma [18–21]. However, a solid component showing low intensity on T2WI is not noted in these tumors. Including a primary OL in the differential diagnosis is important because it would affect the surgical procedure. Unilateral salpingo-oophorectomy or only oophorectomy would be selected, as in the previous reports of OLs [22,23], and gynecologists could take these surgical procedures into consideration before operation if a primary OL is in the differential diagnosis. Although there have been no reports about whether a smaller primary OL should be surgically excised, abdominal pain or discomfort could be a factor suggesting the need for surgery. The frequency of necrotic change of OLs has not been clear. Large leiomyomas in other anatomic regions showed necrosis, such as a uterine leiomyoma, an adrenal leiomyoma, and a primary liver leiomyoma, measuring 11 cm, 8.5 cm, and 20 cm in diameter, respectively [24–26]. Likewise, OLs could become necrotic when the tumor is large. The final histological diagnosis requires immunohistochemical analysis. The proliferating spindle cells were positive for α-SMA staining, and, therefore, the final diagnosis in the present case was primary OL.

In conclusion, when an ovarian tumor with a cystic component is encountered, the solid component that shows low-signal intensity like muscle on T2WI could lead to including primary OL in the differential diagnosis, despite it being very rare.

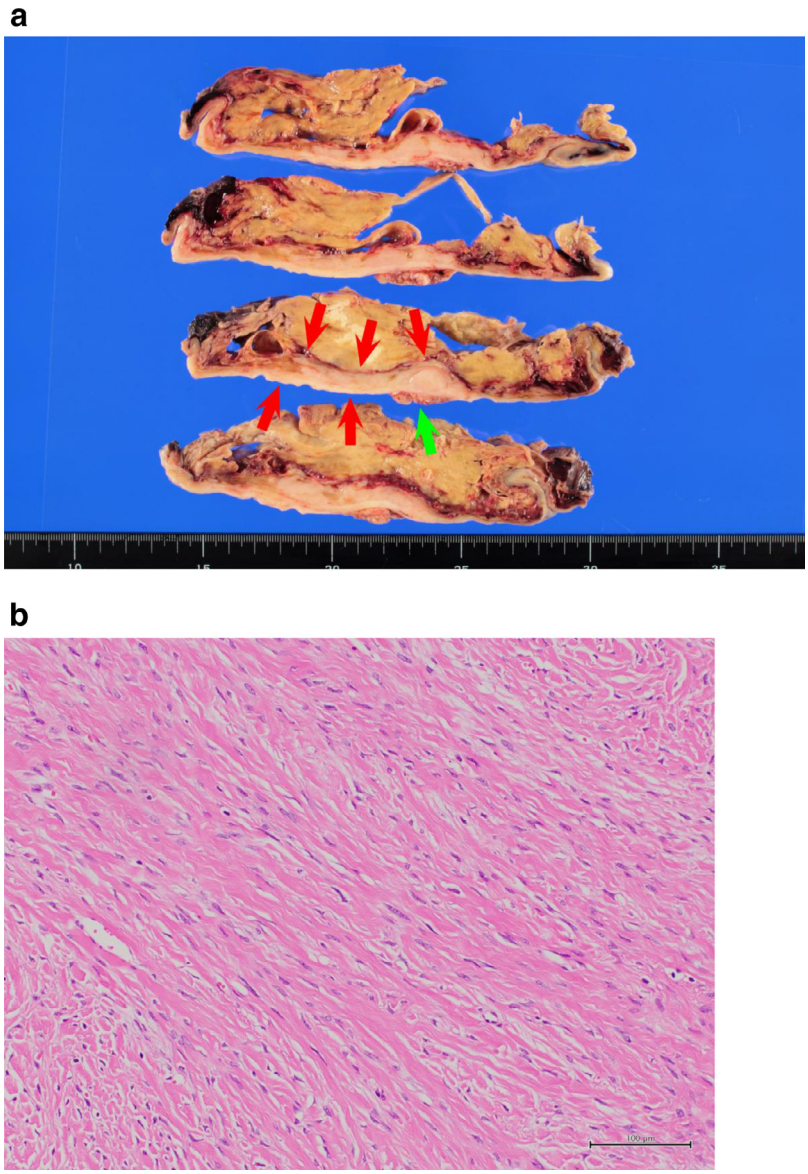

Fig. 3 – In the 4 continuous cross-sectional views of the tumor, the left ovarian duct is attached to it (green arrow), suggesting that it stemmed from the left ovary. The solid component is noted along the inner edge (red arrows) (a). Microscopically, this area contains many proliferating spindle cells (b: hematoxylin and eosin stain, original magnification x 20) showing positive staining for α-SMA. (For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article.)

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