Case report

Ultrasonographic findings and diagnosis of omental dedifferentiated liposarcoma: a case report

Takao Miwa1, Kentaro Otsuji1, Masashi Aiba1, Takahiro Kochi1, Katsuhiro Toda1, Noriaki Nakamura1, Naoki Katsumura1, Tatsuhiko Miyazaki2, and Masahito Shimizu3

1Department of Gastroenterology, Chuno Kousei Hospital, Japan
2Department of Pathology, Gifu University Hospital, Japan
3Department of Gastroenterology, Gifu University Graduate School of Medicine, Japan

Abstract

Liposarcoma is one of the most common types of soft tissue sarcomas and can develop at any site, although omental liposarcoma is extremely rare. Omental liposarcoma has a poor prognosis because the diagnosis is difficult, until it presents as a large tumor causing severe noticeable clinical symptoms. A 51-year-old male with lower abdominal pain was referred to our clinic. Abdominal ultrasonography revealed an ill-defined, solid, heterogeneous, and hypoechoic tumor deep in the lower abdomen. Generally, liposarcomas are hyperechoic, though 20% of liposarcomas present as hypoechoic tumors. This variation might occur depending on the pathological classification. We should consider the possibility of a dedifferentiated component if ultrasonography reveals typical features of soft tissue sarcoma with hypoechoic lesion.

Key words: case report, dedifferentiated, liposarcoma, omental, ultrasonography

Introduction

Liposarcoma accounts for 50% of retroperitoneal sarcomas and 25% of sarcomas of the extremities9. Liposarcomas could develop at any site, but incidence of omental liposarcoma is extremely rare. Liposarcoma has high recurrence rates and low response to existing treatment and radical operation is the only established treatment. According to a large Japanese database, aging, male sex, histological subtype, tumor size and deep tumor location, location in the trunk or head/neck, and intralesional surgical margin were associated with poorer prognosis of soft tissue sarcoma5. Omental liposarcoma develops in the deep abdomen and may have poorer prognosis because the diagnosis seems difficult until it presents as a large tumor that causes severe clinical symptoms, such as abdominal pain, weight gain, and changes in bowel habit. To determine the management, including histological evaluation and operation, evaluation of the malignancy risk is essential.

We report a case of omental dedifferentiated liposarcoma detected by abdominal ultrasonography and its findings.

Case presentation

A 51-year-old male with no relevant medical history, working in a factory that used organic solvents, was referred to our clinic for lower abdominal pain. He was an occasional drinker and consumed one pack of tobacco per day. Blood examination showed no abnormal parameters, including tumor markers. Screening abdominal ultrasonography (LOGIQ E9 with XDclear; C1-6VN convex transducer; GE Medical Systems, Milwaukee, WI, USA) showed a 68 mm × 31 mm × 56 mm, ill-defined tumor, deep in the lower abdomen (Figure 1a). The tumor echogenicity was heterogeneous, hypoechoic at the center, and hyperechoic at the edge. The tumor margin was irregular, and bladder infiltration was suspected. Color Doppler ultrasonography showed color spots reflecting the arterial bold flow (Figure 1b). Contrast-enhanced computed tomography revealed ill-defined omental tumor infiltrating the sigmoid colon (Figure 1c).
The 18F-fluorodeoxyglucose positron-emission tomography showed 18F-fluorodeoxyglucose uptake with standardized uptake value of 15.4 (Figure 1d). These findings suggested malignant omental tumor, and diagnostic resection of the tumor with partial sigmoid colon and bladder serosa resection was performed. The tumor consisted of multinodular yellow lipomatous mass containing discrete, solid non-lipomatous areas (Figure 2a, 2b). Pathological evaluation revealed abrupt transfusion between the well-differentiated liposarcoma and high-grade non-lipogenic area towards the center of the tumor (Figure 3a). The non-lipogenic area revealed high-grade spindle cell neoplasm with pleomorphism and necrosis, including atypical lipoblasts with increased nuclear division (Figure 3b). Additional immunohistochemical staining revealed that MDM2 and CDK4 expression was invariable. The lesion was finally diagnosed as omental dedifferentiated liposarcoma. The patient underwent regular follow-up in our clinic, and reported no recurrence for 10 months. The patient has provided consent to publish this case, and the identity of the patient has been protected.

Discussion

To our knowledge, there are only 15 English case reports on omental liposarcoma, including ours (Table 1). A demographic review of these cases showed that the average age was 50.3 years, and 8 out of 14 (57.1%) patients were male. The classification of liposarcoma was myxoid liposarcoma in 10 out of 15 patients (66.7%), dedifferentiated liposarcoma in 2 (13.3%), well-differentiated liposarcoma in 1 (6.7%), and pleomorphic liposarcoma in 1 (6.7%). The ma-
The major symptom was abdominal pain or abdominal distention in most patients, while some presented with constipation, weight loss, or edema in the leg. The average tumor size was 204 mm, and our case seemed to present the smallest omental liposarcoma reported. The prognosis was poor with recurrence or progression observed in 4 out of 10 patients (40%). In our case, the tumor infiltrated the sigmoid colon. In the colon and rectum, neuronal cell bodies of sensoryafferents were found outside the gut wall. Colon infiltration evoked abdominal pain at an early stage, and this could be the reason for performing abdominal ultrasonography. In fact, screening abdominal ultrasonography proved useful in

Table 1  Characteristics of 15 patients with omental liposarcoma

| No. | Age | Sex | Symptom                  | Size  | Pathological diagnosis          | Treatment        | Prognosis    | Reference |
|-----|-----|-----|--------------------------|-------|---------------------------------|------------------|--------------|-----------|
| 1   | NA  | NA  | NA                       | NA    | Myxoid liposarcoma              | Operation        | NA           | 39        |
| 2   | 54  | F   | Leg edema Abdominal distention | 270 mm | Myxoid liposarcoma              | Operation        | No recurrence | 40        |
| 3   | 83  | M   | Abdominal pain Abdominal distention | 200 mm | Myxoid liposarcoma              | Operation        | No recurrence | 51        |
| 4   | 64  | F   | NA                       | NA    | Myxoid liposarcoma              | Operation        | Recurrence   | 62        |
| 5   | 25  | F   | Abdominal distention     | NA    | Myxoid liposarcoma              | Operation        | NA           | 73        |
| 6   | 45  | M   | Abdominal pain Abdominal distention | NA    | Myxoid liposarcoma              | Operation        | Recurrence   | 83        |
| 7   | 50  | F   | Constipation Abdominal distention | 220 mm | Myxoid liposarcoma              | Operation        | Recurrence   | 90        |
| 8   | 52  | M   | Abdominal pain           | 170 mm | Myxoid liposarcoma              | Operation        | No recurrence | 91        |
| 9   | 52  | M   | Abdominal pain           | NA    | Myxoid liposarcoma              | Operation        | NA           | 91        |
| 10  | 55  | F   | Abdominal distention     | 150 mm | Liposarcoma                     | NA               | NA           | 100       |
| 11  | 34  | M   | Abdominal distention     | 250 mm | Well-differentiated liposarcoma | Operation        | No recurrence | 111       |
| 12  | 63  | M   | Abdominal discomfort Ascites | no clear mass | Pleomorphic liposarcoma        | Conservative     | Progression  | 122       |
| 13  | 65  | F   | Constipation Abdominal pain Abdominal distention | 300 mm | Dedifferentiated liposarcoma | Operation and Adjuvant chemotherapy | No recurrence | 133       |
| 14  | 11  | M   | Constipation              | NA    | Myxoid liposarcoma              | NA               | NA           | 144       |
| 15  | 51  | M   | Abdominal pain           | 68 mm | Dedifferentiated liposarcoma    | Operation        | No recurrence | Our case  |

F: female; M: male; NA: not available.
the detection of the tumor.

The efficacy of ultrasonography in the differentiation between benign soft tissue tumors and sarcomas has been reported. According to these reports, the tumor margin, shape, size, and vascularity indicate the difference between benign and malignant soft tissue tumors. Benign lipoma presents an ovoid shape, well-defined margins, parallel echogenic lines, and avascularity, while liposarcoma presents heterogeneous hyperechogenicity, infiltrated margin, scalloped shape, solid composition, large size, and increased vascularity. However, the usefulness of echogenicity here is controversial. According to some reports, echogenicity is not a significant factor while estimating malignancy risk in sarcomas. On the other hand, some studies showed that low echogenicity is a characteristic of malignancy risk in soft tissue tumors. Liposarcomas can be classified into four types according to the World Health Organization: atypical lipomatous tumor/well-differentiated liposarcoma, myxoid liposarcoma, pleomorphic liposarcoma, and dedifferentiated liposarcoma. Dedifferentiated liposarcoma consists of a multinodular yellow lipomatous (low-grade) area containing non-lipomatous (dedifferentiated) areas. In our case, the lipomatous component on the surface showed high-echogenicity and the solid dedifferentiated component showed low-echogenicity. The surrounding well-differentiated area showed unclear margins, and it was difficult to distinguish the well-differentiated liposarcoma from the intraperitoneal fat in some parts. Based on a previous report, most liposarcomas are usually hyperechoic, although approximately 20% of cases present hypoechoic tumors. In this study, the subtypes of liposarcomas were not mentioned. However, there is a possibility that the dedifferentiated liposarcoma in this study could be assessed as a hypoechoic tumor because of the dedifferentiated component. Dedifferentiated liposarcomas account for almost 10% of the liposarcomas, and this similar rate was noted between hypoechoic tumor in the previous study and the dedifferentiated liposarcoma supports this hypothesis. In another study, most well-differentiated liposarcomas did not show hypoechoic echogenicity, and hypoechoic echogenicity could be the specific property of high-grade liposarcomas rather than low-grade liposarcomas. Echogenicity of liposarcoma should be determined by its pathological component, though there are no discussions explaining the echogenicity variation of liposarcomas in previous reports. From the ultrasonographic findings in our case, dedifferentiated liposarcoma could present as a hypoechoic tumor because of its non-lipomatous area, and the surrounding lipomatous area could present as a hyperechoic lesion.

However, there are several limitations in our case report. First, ultrasonography is useful to estimate the malignant potential of a soft tissue tumor, but it is difficult to make an accurate diagnosis using ultrasonography alone. It could be difficult to differentiate between well-differentiated liposarcoma and fat on imaging in certain cases. Thus, additional imaging, such as contrast-enhanced computed tomography, magnetic resonance imaging, and 18F-fluorodeoxyglucose positron-emission tomography, should be considered before making final diagnosis. Second, ultrasonography should be limited to evaluation of the malignant potential, and pathological confirmation is vital for final diagnosis. Finally, there are no data revealing the difference of echogenicities in liposarcomas according to its classification, and hence further investigation is needed in that direction.

In conclusion, omental liposarcoma is extremely rare and the evaluation of its malignant potential before clinical decision is important. In our case, abdominal ultrasonography combined with other imaging modalities proved useful in evaluating the malignant potential and confirming clinical diagnosis. Additionally, the echogenicity of the dedifferentiated liposarcoma could be heterogeneous, hypoechoic at the center, and hyperechoic at the edge, thus reflecting its pathological component. These findings may add a new insight in the ultrasonographic evaluation of liposarcomas.

**Acknowledgments**

The authors would like to appreciate Dr. Toshihiro Muto and Dr. Soichiro Inoue for their clinical support, and Dr. Nami Asano for her academic support.

**References**

1. Crago AM, Brennan MF. Principles in management of soft tissue sarcoma. Adv Surg 2015; 49: 107–122. [Medline] [CrossRef]
2. Osamura K, Higashi T, Kawai A. Statistics of soft-tissue sarcoma in Japan: report from the bone and soft tissue tumor registry in Japan. J Orthop Sci 2017; 22: 755–764. [Medline] [CrossRef]
3. Ohi M, Yutani C, Shimomukai H, et al. Primary round cell liposarcoma of the omentum. A case report. Acta Cytol 1992; 36: 722–726. [Medline]
4. Okajima Y, Nishikawa M, Ohi M, et al. Primary liposarcoma of the omentum. Postgrad Med J 1993; 69: 157–158. [Medline] [CrossRef]
5. Tsutsumi H, Ohwada S, Takeyoshi I, et al. Primary omental liposarcoma presenting with torsion: a case report. Hepatogastroenterology 1999; 46: 2110–2112. [Medline]
6. Fotiadis C, Zografos GN, Karatzas G, et al. Recurrent liposarcomas of the abdomen and retroperitoneum: three case reports. Anticancer Res 2000; 20(1B): 579–583. [Medline]
7. Alameda F, Coronimas JM, Barranco C, et al. Primitive round cell liposarcoma of the omentum: diagnostic value of ultrastructural study. Ultrastruct Pathol 2020; 15(2): 68–72. doi: 10.2185/jrm.2019-013
2003; 27: 433–437. [Medline] [CrossRef]
8. De U, Jain BK, Sah SP, et al. Primary liposarcoma of the omentum: a case report and review of the literature. Indian J Pathol Microbiol 2003; 46: 638–640. [Medline] [CrossRef]
9. Milic DJ, Rajkovic MM, Pejic VD. Primary omental liposarcoma presenting as an incarcerated inguinal hernia. Hernia 2005; 9: 88–89. [Medline] [CrossRef]
10. Imai A, Onogi K, Sugiyama Y. Omental liposarcoma; a rare complication in neurofibromatosis type 1. J Obstet Gynaecol 2006; 26: 381–382. [Medline] [CrossRef]
11. Meloni F, Fico S, Profili S, et al. Omental Well-Differentiated Liposarcoma: US, CT and MR Findings. Int J Biomed Sci 2009; 5: 302–304. [Medline] [CrossRef]
12. Tomita Y, Kai K, Shirai H, et al. Primary omental pleomorphic liposarcoma identified at autopsy in a living donor kidney transplant recipient. CEN Case Rep 2012; 1: 39–42. [Medline] [CrossRef]
13. Soufi M, Mdaghri J, Benamr S, et al. Giant liposarcoma of the omentum mimicking an ovarian tumor. A case report. Indian J Surg 2012; 74: 423–427. [Medline] [CrossRef]
14. Hightower JL Jr, Dire DJ. Omental liposarcoma presenting as chronic constipation. Pediatr Emerg Care 2014; 30: 483–484. [Medline] [CrossRef]
15. Brierley SM, Hibberd TJ, Spencer NJ. Spinal afferent innervation of the colon and rectum. Front Cell Neurosci 2018; 12: 467. [Medline] [CrossRef]
16. Belli P, Costantini M, Mirk P, et al. Role of color Doppler sonography in the assessment of musculoskeletal soft tissue masses. J Ultrasound Med 2000; 19: 823–830. [Medline] [CrossRef]
17. Morii T, Kishino T, Shimamori N, et al. Differential diagnosis between benign and malignant soft tissue tumors utilizing ultrasound parameters. J Med Ultrason (2001) 2018; 45: 113–119. [Medline] [CrossRef]
18. Chiou HJ, Chou YH, Chiu SY, et al. Differentiation of benign and malignant superficial soft-tissue masses using grayscale and color doppler ultrasonography. J Chin Med Assoc 2009; 72: 307–315. [Medline] [CrossRef]
19. Oebisu N, Hoshi M, Ieguchi M, et al. Contrast-enhanced color Doppler ultrasonography increases diagnostic accuracy for soft tissue tumors. Oncol Rep 2014; 32: 1654–1660. [Medline] [CrossRef]
20. Nagano S, Yahiro Y, Yokouchi M, et al. Doppler ultrasound for diagnosis of soft tissue sarcoma: efficacy of ultrasound-based screening score. Radiol Oncol 2015; 49: 135–140. [Medline] [CrossRef]
21. Morii T, Kishino T, Shimamori N, et al. Preoperative ultrasonographic evaluation for malignancy of soft-tissue sarcoma: a retrospective study. Open Orthop J 2018; 12: 75–83. [Medline] [CrossRef]
22. Fletcher C, Bridge J, Hogendoorn P, et al. WHO Classification of Tumours of Soft Tissue and Bone. IARC Press, Lyon, 2013; 14–43.