A well-differentiated liposarcoma co-existent with leiomyoma

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Background. The coexistence of a well-differentiated liposarcoma (atypical lipomatous tumor) and benign smooth muscle component in a single soft tissue neoplasm is extremely rare. Case report. Histologic and immunohistochemical characteristics of tumor, which represented the recurrence of a retroperitoneal atypical lipoma in a 50-year-old female patient are presented in this paper. The tumor represented the recurrence of a retroperitoneal atypical lipoma. Lipomatous component consisted of a mixture of lipoma-like and sclerosing variant of well-differentiated liposarcoma. Characteristically, the heterologous smooth muscle differentiation was manifested as a macroscopically visible nodular growth in the form of a leiomyoma within the adipocytic component. It consisted of intersecting fascicles of spindle cells, which lacked mitoses and significant atypia, and were immunopositive for smooth muscle markers. Conclusion. This case of well-differentiated liposarcoma with benign smooth muscle is the first reported tumor of retroperitoneal localization. It is also the first one to exhibit the heterologous smooth muscle component as a distinct leiomyoma.

Key words: retroperitoneal neoplasms; liposarcoma; leiomyoma; soft tissue neoplasms.

Introduction

Liposarcoma may contain benign or malignant heterologous mesenchymal elements. Malignant divergent differentiation is mostly encountered in dedifferentiated type of liposarcoma (1, 2), although sporadic tumors composed of other types of liposarcoma and osteo-, chondro-, leiomyo-, and rhabdomyosarcoma have been reported (3–6). However, a well-differentiated (WD) liposarcoma with benign smooth muscle component is extremely rare: to our knowledge only five such cases have been described (7, 8). The case, which was the first to contain the smooth muscle component as a separate leiomyomatous growth, is reported in this paper.

Case report

A 50-year-old female with one-month history of enlarging abdominal mass is presented. Computed tomography (CT) scan demonstrated a soft tissue mass with fat density in the left pelvic region. It extended to the anterior and lateral abdominal wall, posteriorly to the iliac bone, and inferiorly to the left hip joint, displacing the bowel laterally. The patient underwent surgery and was found to have a tumor localized predominantly in the retroperitoneum. Retroperitoneal muscles were unaffected. The tumor was excised with left hemicolectomy. A 3-year follow-up showed no evidence of recurrence or metastasis. Past medical history revealed that eight years prior to the admission the patient had a retroperitoneal atypical lipoma removed in another hospital. The slides of the tumor were not available for the histologic re-review.

Results

The resected tumor was multilobulated mass measuring 20 × 15 × 8 cm, with a yellow cut-surface consistent with mature adipose tissue. It contained sharply demarcated, round, gray-white, and firm nodule showing a whorled pattern, measuring 7 cm in diameter (Figure 1). The left hemicolectomy specimen was unremarkable.

Extensive sampling throughout the tumor was performed for histological examination. The tumor consisted of two distinct components, lipomatous and smooth muscle component.
were characterized by mature fat cells with great variation in cell size and slight nuclear atypia. In addition to lipoma-like tissue, the sclerosing type areas showed irregular, narrow, and broad bands of collagenous tissue, containing atypical stromal cells (Figure 2a). In some parts of the tumor the fibrous component was more prominent. Lipoblasts and mitoses were absent.

The second component corresponding to well demarcated white and firm nodule on gross appearance was found to be a proliferation of spindle cells arranged in fascicles (Figure 2b). The cells exhibited eosinophilic cytoplasm and small uniformly spindled nuclei with only slight focal atypia and no mitoses (Figure 3a). The spindle cells showed immunopositivity for α-smooth muscle actin (Dako, Denmark) (Figure 3b), and desmin (Dako, Denmark), confirming their smooth muscle differentiation. Gross and histologic appearance of smooth muscle component was consistent with a leiomyoma.

The adipocytic component was well-differentiated liposarcoma — atypical lipomatous tumor showing a mixture of lipoma-like and sclerosing subtypes. Lipoma-like areas

There was a patchy dense inflammatory infiltrate consisting of lymphocytes and rare polymorphonuclear leukocytes with occasional lymphoid follicles within the leiomyomatous component (Figure 2b).

Fig. 1 – Gross appearance of a retroperitoneal well-differentiated liposarcoma/typical lipomatous tumor with smooth muscle component in the form of nodular leiomyomatous growth.

Fig. 2 – A part of adipocytic component of the tumor corresponded to well-differentiated liposarcoma, sclerosing-type (a) (HE × 200); the leiomyoma within a lipoma-like component of well-differentiated liposarcoma containing a lymphoid follicle (b) (HE × 40).

Fig. 3 – Benign-looking smooth muscle cells without atypia or mitoses in leiomyomatous component (a) (HE × 200), showing α-smooth muscle actin immunopositivity in smooth muscle component (b) (avidin-biotin complex × 200).
Discussion

We described a tumor consisting of two components: WD liposarcoma and leiomyoma. The diagnosis of WD liposarcoma in the lipomatous component was based on previously accepted criteria (9–11). Since WD liposarcoma lacks the capacity to metastasize, Evans coined the term atypical lipomatous tumor (7), and both are now considered as synonyms (11). Purely lipoma-like and sclerosing subtype of WD liposarcoma may be found in different parts of the same tumor (9), as in the case presented, in which the smooth muscle nature of the second component was confirmed by immunohistochemistry. It displayed a benign appearance according to the lack of mitoses and significant atypia.

Soft tissue tumors showing combined features of adipose and smooth muscle are rare. Several combinations of this dual lineage differentiation within the same neoplasm have been reported. Benign soft tissue tumors composed of mature adipose tissue and benign smooth muscle were recently defined as myxolipomas (12). Other forms of combined smooth muscle/lipomatous tumor included sporadic cases displaying differentiated, although malignant lipomatous and smooth muscle components (4, 5), or malignant smooth muscle component within dedifferentiated component of liposarcoma (1, 2).

The third adipocytic and smooth muscle combination in the same neoplasm represented the admixture of atypical lipomatous tumor and benign smooth muscle tissue. To our knowledge, there have been only two reports combining five tumors showing these features: Evans (7) in 1990 described three cases of atypical lipomatous tumor containing foci of benign smooth muscle, and Zamecnik et al. (8) in 1999 added two more cases. Two of the tumors occurred in the scrotum, and three in the mediastinum, abdomen, and subcutis of the chest. Smooth muscle component was seen only in the primary tumors (3 cases), both in the primary and recurrent tumor (1 case), and in recurrence only (1 case), similar to the tumor of our patient.

This case was the first reported occurrence in the peritoneum. Previously reported cases displayed benign smooth muscle cells in microscopic foci (7), or interspersed within the lipomatous component (8). The case presented here has been unique featuring the smooth muscle component as sharply demarcated leiomyomatous mass. Inflammatory infiltrate in smooth muscle component was its another distinctive feature.

Zamecnik et al. (8) considered the nature of atypical lipomatous tumor with benign smooth muscle component to be in-between myxolipoma and dedifferentiated liposarcoma with leiomyosarcomatous differentiation (8). According to the published cases (7, 8), as well as the presented one, it seems that a leiomyomatous component does not alter the behavior of atypical lipomatous tumor.

In conclusion, this case presented a rare combination of well-differentiated liposarcoma and a leiomyoma in a single neoplasm, which reflected an unusual form of bidirectional mesenchymal differentiation.

References


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Apstrakt

Dobro diferentovan liposarkom sa leiomiomom

Cilj. Uduženost dobro diferentovanog liposarkoma (atipličnog lipomatoznog tumora) i glatkomišićne komponente benignih osobina izuzetno je retka. Prikaz slučaja. U radu su prikazane histološke i imunohistohemijske karakteristike tumora koji je bio recidiv retroperitoneumskog atipličnog lipoma kod bolesnice stare 50 godina. Lipomatozna komponenta se sastojala od lipomu sličnog (lipoma-like) i skleroizgurajućeg tipa jasno izdiferentovanog liposarkoma. Heterologa glatkomišićna diferencijacija manifestovala se makroskopski jasno ograničenim nodusom u vidu lejomioma. Lejomiom se sastojao od ukrštenih snopova vretenastih celija bez mitoza i značajne atipije, koje su ispoljavale imunopozitivnost na markere glatkih mišića. Zaključak. Ovaj slučaj dobro diferentovanog liposarkoma sa benignom glatkomišićnom komponentom prvi je opisani tumor retroperitoneumske lokalizacije, a takođe je i prvi sa heterolognom glatkomišićnom komponentom u obliku jasnog lejomioma.

Ključne reči: retroperitonealne neoplazme; liposarkom; leiomiom; meka tkiva, neoplazme.

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