

A case of multifocal liposarcoma?

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A 53-year-old man presented with a grade 2 liposarcoma in his left thigh and grade III liposarcomas in the mediastinum and omentum. Later, a grade 2 tumor was discovered in the musculature of his right thigh. In addition to recurrences, he developed lesions that were

considered metastatic. He died 3 years later of multiple lesions. The case may represent a multifocal liposarcoma, which has been previously reported in only 35 cases.

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Liposarcoma may occur simultaneously in several sites not representing metastases. The multicentric, multifocal manifestation has been even called sarcomatosis (Siegmund 1934). The condition is rare. We found 35 cases in the literature (Siegmund 1934, Ackerman 1944, Enzinger and Winslow 1962, Reszel et al. 1966, Georgiades et al. 1969, Kagawa et al. 1985).

We report a case of liposarcoma where the primary manifestations presumably were multifocal with metastases in the later course.

Case report

A 53-year-old polar explorer had to withdraw from participation in an expedition in February 1985 because of pain in his left distal thigh. In June 1985, a subfascial tumor with a diameter of 5 cm was found. An abdominal examination revealed a mass extending to the umbilicus. A chest radiograph showed a mass in the left lower anterior mediastinum. In a computed tomographic study, all the masses had a low density, which was consistent with a lipomatous tumor. The mediastinal mass had a diameter of 8 cm; and the abdominal mass, between the gall bladder and iliac fossa, had a diameter of 10 cm. The mass in the left thigh was located extramuscularly close to the femur. A biopsy of the mass in the thigh showed tissue with abundant myxoid intercellular substance containing a plexiform capillary network with predominantly monovacuolar lipoblasts of varying size and with slight to moderate nuclear atypia, indicating a myxoid liposarcoma of histologic grade 2 (Broders 1964).

A sternotomy was performed, and the mediastinal tumor, weighing 250 g, was removed; next, the incision was widened to a laparotomy to remove the omental, 500-g tumor. And finally, the operation was finished with a wide excision of the tumor in the left thigh, which

weighed 420 g. The histologic preparations of the mediastinal and abdominal tumors demonstrated a myxoid liposarcoma, as described above. In addition, areas dominated by small, immature, usually nonvacuolated tumor cells (round cells) with a moderate nuclear atypia compatible with liposarcoma, grade 3, were found. The histologic examination of the tumor in the thigh showed a purely myxoid liposarcoma, grade 2.

The patient was followed clinically and radiographically at 3-month intervals. Fifteen months later, a CT scan showed a recurrence, a mass 4-cm in diameter, in the mediastinum and another mass, 5 cm in diameter, in the left thigh. The tumors were removed respectively with a marginal and a wide excision. The histologic examination again demonstrated a purely myxoid liposarcoma, grade 2, at both sites. Postoperatively, the patient received radiotherapy, 50 Gy, to both locations.

For a year, he had no signs of further recurrences. Then, he presented with a mass, 3 cm in diameter, in his right distal adductor longus muscle. The CT scan showed a low density mass that, in contrast to the previous masses, took up contrast medium avidly. The tumor was removed by an adductor longus myectomy in October 1987. The histologic preparations again confirmed a liposarcoma, grade 2. There were, however, areas of large, more mature tumor cells than what had been found earlier.

Soon after this operation, the patient developed numbness of his left lower trunk and leg, as well as weakness of the right lower extremity. A myelographic investigation showed no compression of the spinal cord. The patient was found to have a lesion corresponding to the L1-2 level with a pyramidal tract affection, and with deep sensitivity on the left side and analgesia on the right side. The partial Brown-Séquard syndrome was considered to be due to postirradiation myelopathy. The MRI study was consistent with an irradiation injury from Th-2 to Th-12.

Two and one-half years after the primary manifestations, a low attenuating tumor mass, 2.5 cm in diameter, was found in the left proximal adductor musculature. A compartmental excision of the left adductors was performed in March 1988. Once again, the histologic picture was that of a purely myxoid liposarcoma. No other recurrences were found. Neurologically, the patient's symptoms had become slightly aggravated, with spasticity of the right lower limb, and he now used a crutch. His anal sphincter control was normal.

In July 1988, he presented with muscle weakness and pain in the groins radiating to the anterior thighs. A lumbar CT examination showed a pathologic fracture of the body of L-4 with compression of the dural sack, verified by a myelogram. He was in good physical condition, and accepted further operative treatment. A laminectomy of L3–4 was performed with internal fixation using the Dick apparatus.

After the patient recovered from this procedure, 10 days later, the body of L-4 was removed by a marginal excision and an anterior fusion from L-3 to L-5 was performed using a bone block from the left iliac crest. The histologic preparations showed vertebral infiltration of a purely myxoid liposarcoma compatible with metastasis from the soft tissue tumors described above. The patient's walking ability improved, and the groin pain subsided. He was discharged to a rehabilitation center.

One month later, he had increasing abdominal pain and constipation, and pain radiating to the lower extremities. A CT examination revealed large tumor masses in the right and left abdomen, and he died 2 months later, i.e., 3 years and 3 months after the primary admission. At autopsy, large abdominal tumor masses were found infiltrating the larger omentum and retroperitoneal tissue. Tumor infiltration was also found in the remaining lumbar column compressing the medulla and the nerve roots. The histologic preparations of the omental and dural tumors showed a purely myxoid grade 2 liposarcoma.

For this case presentation, all the histologic specimens taken from several sites of the tumors were reviewed.

Discussion

Siegmund (1934) reported on the multicentricity of liposarcomas, and called the condition sarcomatosis. The condition is rare. Ackerman (1944) reported 1 case and referred to four previous reports. In a series of 222 liposarcomas of the extremities and limb girdle, 5 cases were found to be multicentric (Reszel et al. 1966). Enzinger and Winslow (1962) found 20 cases among

their 103 liposarcomas where a liposarcoma of the lower extremity was followed by "secondary tumors" at various sites, including the thoracoabdominal area and the opposite thigh. Mediastinal liposarcomas are rare; and in a series of 8 cases (Cicciarelli et al. 1964), all the tumors had solely a mediastinal location.

Our patient developed liposarcomas in four different locations—in both thighs, in the mediastinum, and in the abdomen. Certain features support a multifocal origin of the disease. The tumor in the thigh could have been a primary one. But metastasis to the omentum would be rare. Further, the liposarcomas removed from the mediastinum and omentum were of the mixed type, with myxoid and round cells, whereas the other tumors were purely myxoid. Of course, focal variations are common in myxoid liposarcomas, and small foci of less differentiated areas with round cells may be lost when a tumor is examined. The tumor in the lumbar spine was considered metastatic because it could not be identified in the earliest radiographs and because it was of the same histologic type as the tumors removed from the lower extremities. In other similar studies, almost the same regional distribution and clinical course were reported with similar conclusions (Enzinger and Winslow 1962, Kagawa et al. 1986).

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