

## Lymphedema as the First Manifestation of Hodgkin's Disease

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### Summary

Two cases of *Hodgkin's* disease are described, initially manifested with unilateral lymphedema of the lower extremity. The appearance of pronounced lymphedema of a lower extremity, as the first clinical manifestation and presenting sign of *Hodgkin's* disease is extremely rare. The surgical treatment of this condition consisted in removal of the involved lymph nodes of the inguino-femoral area and at the same time in omental transposition of a lymphatic communication of the afflicted area with the peritoneal cavity. The procedure was successful in ameliorating the lymphedema. No episodes of postoperative cellulitis occurred and good functional capacity of the extremity was achieved.

### Introduction

*Hodgkin's* disease is a chronic malignant disease of unknown etiology, and it is usually manifested by progressive enlargement of the lymph nodes, spleen and liver. On some occasions, other organs and tissues are, also involved. The clinical picture in the initial cases is characterized by pruritus, and as the disease progresses, fever, anemia, night sweats, cachexia and weight loss, also become evident. Some patients present with peripheral lymph node (e.g. axillary, cervical or inguinal) enlargement. In certain cases there is evidence of lower extremity edema of venous etiology i.e. due to pressure of the inferior vena cava by the enlarged lymph nodes. The initial presentation with unilateral lower extremity edema of lymphatic etiology as in the two cases presented here, is extremely rare. Asymptomatic patients may have their adenopathy for extended periods of time with waxing and waning of lymph node size. Old x-ray films, in retrospect, may reveal that evidence

of mediastinal widening has been present for several years.

### Case Reports

The first case involves a 47 year old white male who was admitted because of a generalized, painless, pitting edema of the entire left lower extremity, of about one month's duration. The arterial and venous system of the left lower extremity was found to be patent, with the utilization of ultrasonographic and angiographic methods. A 3 x 4 cm hard, immobile and slightly painful mass was felt by palpation of the left iliac fossa. The rest of the physical examination elicited no abnormal findings. The patient was afebrile. The hematological examination was unremarkable, with the exception of an increased erythrocyte sedimentation rate (ESR) of 95 and 107 mm, on the first and second hour, respectively. The chest x-ray and the radiographic visualization of the large intestine following a barium enema, were normal. The intravenous urogram showed an irregularity of the left margin of the urinary bladder, probably due to external pressure (see Fig. 1). The lymphangiographic work-up displayed large and irregular left inguinal nodes with filling defects, which gave the impression of a neoplastic infiltration (see Fig. 2). The afferent lymph vessels were found to be dilated.

The patient was operated and a vertical incision was made along the left iliac fossa, which extended into the thigh. Following opening of the peritoneum a mass was encountered which consisted of enlarged lymph nodes. The iliac and inguinal lymph nodes were removed and biopsies were taken of some pre-



**Fig. 1** Irregularity of the left margin of the urinary bladder

aortic nodes. Next followed, the transposition of the greater omentum in the area that was occupied by the removed lymph nodes. The

histologic examination of the removed nodes showed *Hodgkin's disease*. The postoperative course was marked by a dramatic decline in the edema, which completely disappeared within a few days. The patient underwent chemotherapeutic treatment and currently (eight months postoperatively) he is doing well.

The second case concerns a 51 year old, white female, who was admitted because of painless, pitting edema of the entire left lower extremity of about six months duration. The personal and family history were unremarkable. The patient was gravide 7, with 3 normal deliveries and 4 spontaneous abortions. Menopause was at the age of 41.

The physical examination showed an afebrile patient, with marked edema of the entire left lower extremity. The ultrasonographic and angiographic work-up showed good patency of the arterial and venous system of the extremity: The gynecological examination was unremarkable. The hematologic exam. depicted a moderately increased ESR of 35 and 68 mm on the first and second hour, respectively. The radiographic visualization of the large intestine, and the intravenous urogram were all considered normal. The lymphangiographic work-up exhibited a left iliac lymph node en-



**Fig. 2** Left inguinal and iliac nodes with filling defects





**Fig. 3** Left inguinal and iliac lymph nodes with filling defects

largement (see Fig. 3), with filling defects, which gave the impression of neoplastic infiltration and the afferent lymph vessels were shown to be dilated.

During the surgical operation on the left inguino-femoral area, the vertical, as well as horizontal line, of left iliac and inguinal nodes were found to be enlarged. The area was thoroughly dissected with removal of the subcutaneous fat, lymph nodes and the aponeurosis. A corrugated plastic graft consisting of Dacron was implanted through the femoral ring, in order to facilitate lymph drainage to the abdominal cavity and the absorption of lymph by the greater omentum. The histologic examination of the removed lymph nodes showed *Hodgkin's* disease with moderate nodular sclerosis. The postoperative decrease of edema was remarkable with complete disappearance within a few days. The patient underwent chemotherapeutic treatment.

### Discussion

In these two cases, which are very rare, *Hodgkin's* disease became manifest by appearance of unilateral lymphedema of a lower extremity. In order to alleviate the lymphedema, it was decided in the first case to transpose the greater omentum in the area of the removed lymph nodes. In the second case, it was decided, to utilize a corrugated plastic graft consisting of Dacron which was implanted through the femoral ring to expedite the lymphatic drainage into the abdominal cavity and its subsequent omental reabsorption.

The four procedures frequently utilized in the surgical management of lymphedema are: 1) the Charles procedure, which is total excision of subcutaneous tissue, with free skin grafting, 2) omental transposition, 3) buried dermal flap and 4) subcutaneous excision of the lymphedematous tissue beneath skin flaps (2).

The introduction of an omental flap into the involved extremity has been advocated (3, 4) however, there has been considerable criticism of the theoretical basis of this procedure (5, 6).

In the two described cases of *Hodgkin's* disease, with the rare manifestation and presenting symptom of unilateral lymphedema of a lower extremity, the results were very encouraging with respect to subsiding of the lymphedema.

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