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FACTITIOUS LYMPHEDEMA OF THE UPPER-LIMB

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Abstract

A case of factitious lymphedema experienced in a 24 year old female (cashier of a supermarket) caused by a tourniquet was reported. Edema had developed in the dominant hand during the course of physiotherapy for shoulder-arm-neck syndrome. As the possibility of thoracic outlet syndrome was considered, she had also received surgery. But the exploratory surgery in this case did not cure the edema eternally. Edema began to recure from the 8th day after surgery. When a plaster cast was applied and the patient was unable to gain access to the limb to apply a tourniquet, the edema markedly improved.

The patient had been recognized as having an occupation related ailment, and had been receiving compensation benefits from two and a half years ago. It was felt that hysteria loomed in the background of this case. As both surgery and physiotherapy is ineffective to this lymphedema, the surgeon should discontinue treatment and refer the patient for psychiatric care.

CASE REPORT

The patient is a 24 year old female referred to this hospital with complaint of recurrent chronic lymphedema of unknown cause. After graduating from senior high school, she was employed as a cashier at a supermarket where she tapped the keys of a cash-register.

About 3 years ago she began experiencing numbness in the nape, shoulders and arms for which she received treatment, but as her condition aggravated, 6 months later she was admitted to a hospital for detailed examinations. She was diagnosed as shoulder-arm-neck syndrome, and her condition was recognized as an occupation related ailment. She received compensation benefits from then; Temporary relief permitted her to return to work, but before long

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(about one year after her onset of shoulder-arm-neck syndrome) she had began to develop edema in her right upper limb (dominant hand). She was hospitalized for treatment 3 times, and during this time her condition when through episodes of improvement and aggravation.

Two years after her first episode of edema, she was admitted to our hospital. As findings suggestive of thoracic outlet syndrome were noted on angiography, exploratory surgery was performed, but it was found there were no signs of oppression of the subclavian vessels. Division of the pectoralis minor muscle and excision of the 1st rib were performed. The edema practically disappeared by the day after surgery, but began to recur from the 8th day and reached almost the presurgical state. Physiotherapy was administered, but as there was hardly any improvement, she was discharged after 4 months, but physiotherapy was continued as an outpatient.

While following her condition as an outpatient by examination a few times a week, circumferential ecchymotic linear bands were noted in the mid-portion of her upper arm. She was immediately readmitted to the hospital, and plaster casts were applied to her trunk and the affected limb. Her edema completely subsided by the next morning. As the patient was unable to

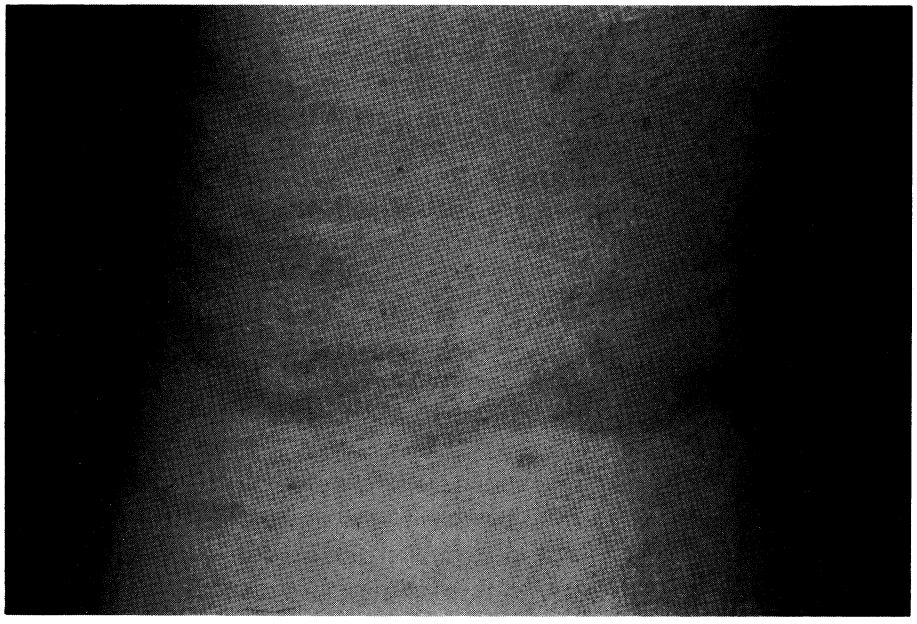

Fig. 1. This photograph shows the mid-lateral aspect of the dominant right upper arm, the hand is to the bottom, the shoulder to the tip. Note the transverse, circumferential ecchymotic linear bands.

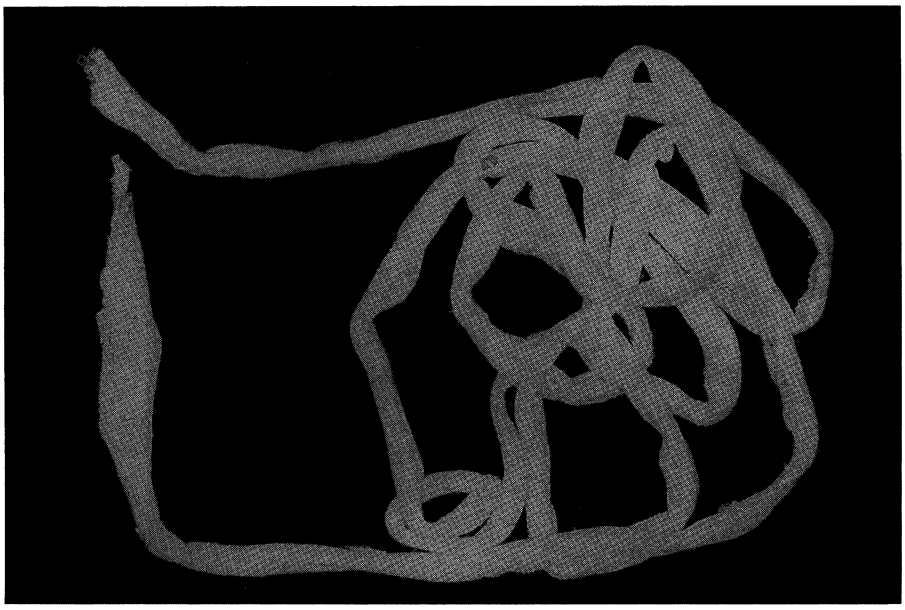

Fig. 2. This old elastic bandage was used to provide a turniquet effect by herself.

apply a tourniquet, attempts had been made to constrict her upper arm by pushing toilet paper into the cast from the gap at the end. Also an old elastic bandage which had no doubt been used as a tourniquet was found in her possession (Fig. 1–4). Thus, it was concluded that the edema was self-induced.

Our psychiatrist found that hysteria was at the base of the patient's factitious lymphedema.

DISCUSSION

There are various causes for edema of the limbs. The congenital causes include lymphatic aplasia, hypoplasia[1] and Milroy's disease,[2] but in these conditions, the degree is more severe in the lower extremities than the upper. The acquired causes include tumor, surgery, irradiation and filariasis which induce lymphatic obstruction and cause secondary edema. Thoracic outlet syndrome induces venous obstruction which in turn causes upper limb edema.

In congenital lymphedema, aplasia or hypoplasia of the lymph channels can be demonstrated by lymphangiography, and in acquired lymphedema, it will show chronic obstructive pattern at the site of lymph stasis, and dilated channels more distally. In the case of thoracic outlet syndrome, the site of

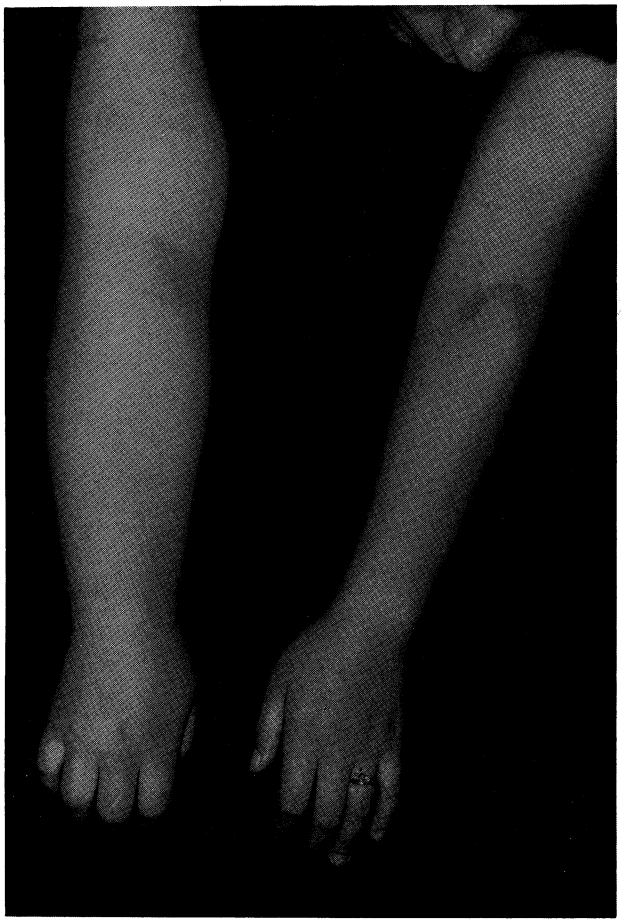

Fig. 3. This photograph was taken before applying plaster cast. Note the termination of the swelling in the midportion of right upper arm.

venous blockage can be ascertained by abducting and extending the limb during venograph. In factitious lymph-edema caused by intermittent application of a tourniquet, distension of the superficial lymphatics and extravasation of the contrast medium may be seen distal to the site of constriction.[3] Further, so-called broken windowpane pattern which is effect of increased collateral circulation will be seen.[4]

According to R. J. Smith,[4] factitious lymphedema has been noted to develop following blows to the hand and repeated irritation of the skin, in addition to the application of tourniquets. It is said that in cases with recurrent episodes of edema or chronic edema following a comparatively minor

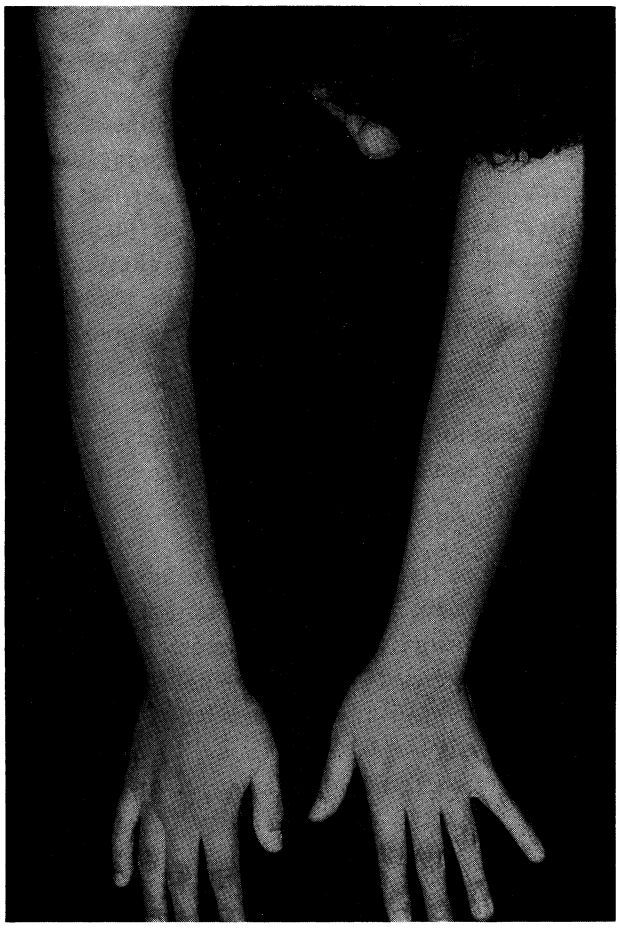

Fig. 4. This photograph was taken immediately after the plaster cast applied for one night was removed.

injury of the hand, the possibility of it being self-induced should be considered.

Characteristically, factitious lymphedema shows signs of tourniquet compression (constriction and circumferential discoloration) and the presence of edema distal to this site. Careful attention should be paid by the physician as the patient will attempt to conceal the constricted part from his eyes. It is considered that practically all cases reported to date showed evidence of psychopathology.[5–10] There are many cases such as ours who induce peculiar symptoms by the application of a tourniquet and visit a number of hospitals, even going to the extent of submitting to surgery. Cases such as this also

referred to as having Munchausen's syndrome.[11–13] This description includes hysteria, schizophrenia, masochism and abnormal character. Our case is considered to have hysteria.

K. Tsuge[14] has succeeded in alleviating severe factitious lymphedema by the use of a plaster cast. For the purpose of confirming diagnosis, the authors applied a plaster cast from the trunk to the hand so that a tourniquet could not be applied. It is considered a characteristic of factitious lymphedema to find that a plastic cast is much more effective in alleviating edema than any type of physiotherapy. In the author's case, when the plaster cast was removed, there was invariably recurrence of edema the next morning.

Patients with factitious lymphedema usually first visit a surgeon, but it is not possible to improve the edema even by physiotherapy on surgery. Therefore, once a definite diagnosis has been established, the surgeon should not attempt any further treatment, and refer the patient to a psychiatrist.

SUMMARY

One case of factitious lymphedema of the upper limb which developed during the course of treatment for shoulder-neck-arm syndrome was reported. This patient was receiving workmens accident compensation benefits. She was considered to have hysteria based on psychopathological evaluation. In recurrent chronic lymphedema, when the cause is not clear, the possibility of it having been self-induced should be considered. The application of a plastic cast is more effective in reducing swelling than any type of physiotherapy.

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