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Lymphedema: Results of Surgical Treatment in 64 Patients (1936 – 1964)

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Summary

Patients with peripheral lymphedema are usually treated without operation. In some, however, operations are required in order to reduce swelling, ensure comfort, and improve function and appearance. Because new surgical approaches are available for the treatment of peripheral lymphedema, we reviewed our previous operative experiences for these conditions at the Mayo Clinic to provide a reference to which the newer procedures may be compared.

Sixty-four patients underwent operation for peripheral lymphedema between 1936 and 1964. Follow-up information was obtained on 56 patients. Seventeen patients required three procedures to alleviate the swelling in an extremity. Morphologic aspects of the excised tissue were also evaluated in these cases. Results were considered excellent in 13, good in 22, fair in 8, and poor in 13. Complications of operation consisted primarily of wound infections, hematomas, and necrosis of skin flaps.

Most patients with idiopathic or secondary lymphedema can be treated satisfactorily by medical measures (1-4). In a few patients, however, medical therapy alone will not reduce the swelling enough for the patient to be comfortable either esthetically or functionally. For these patients, surgical means have been used to alleviate the swelling. Recently, significant advances have occurred in the surgical treatment of lymphedema with the introduction of a newer "physiologic" procedure aimed at restoring lymph-channel continuity by means of autotransplantation (5). Because of the availability of this new procedure, we reviewed our overall experience with the earlier surgical procedures in an effort to establish a reference for the evaluation of the newer surgical procedure.

* Dr. Harrison died July 5, 1975.

Our Series

The results of the surgical treatment of 64 patients with lymphedema seen at the Mayo Clinic between 1936 and 1964 were reviewed. The type of lymphedema, incidence of preoperative and postoperative cellulitis and lymphangitis, previous surgical treatment, type and number of surgical procedures, complications, and results of surgery were considered. Follow-up information was obtained on 56 of the 64 patients either by direct examination or by letter to the patient or to his physician.

Of the 64 patients (39 women and 25 men), 45 had idiopathic lymphedema (13 of the congenital type, including 2 with Milroy's disease) and 19 had secondary lymphedema. Fifty-nine patients had involvement of the lower extremity, and five (four postmastectomy and one congenital) had involvement of the upper extremity. Fourteen patients had bilateral swelling; 25 had involvement of the left leg, 3 of the left arm, 20 of the right leg, and 2 of the right arm. The swelling was extreme in 59 and moderate in 5.

Twenty patients had had 39 previous surgical procedures on the involved extremity, including the Homans and Kondoléon procedures, other excisional types of surgery, lumbar sympathectomy, and ligation of varicose veins. There was either no relief or only temporary relief of the swelling from these procedures.

Before their surgical treatment at the Mayo Clinic, 31 of the 64 patients had two or more episodes of cellulitis and lymphangitis, and 2 had single episodes of phlebitis of the legs. Eight of the 31 patients had trichophytosis.

In all 64 patients, the indication for surgery was the huge size of the limb; 41 patients had limbs that often interfered with function, and 23 patients had recurrent episodes of cellulitis and lymphangitis.

A total of 135 procedures were done on the 64 patients; these procedures included the Homans, the modified Charles, and the Macey procedures, as well as other plastic operations. Seventeen patients needed three procedures or more in order for the size of the limb to be reduced effectively (Table 1).

Clinical and Surgical Findings

Lymphedema Praecox

Thirty-two patients (24 women and 8 men) had lymphedema praecox (50 % of the series). The average age of the patient at the onset of swelling was 19.2 years, whereas the average age at surgery was 33 years. The swelling was located in the lower extremity in all 32 patients; 9 had bilateral involvement. Twenty-eight patients had severe swelling, whereas four had moderate swelling. In all patients, the indication for surgery was the size of the limb (Fig, 1 through 5).

Surgery. – Thirty-three limbs of 32 patients (1 had bilateral swelling) were operated on for a total of 73 operations; these included the single, double, and triplestage Homans operations, the modified Charles procedures, the Macey procedures, and the lesser plastic procedures (Table 1). In general, the modified Charles procedure was used when the lymphedema was more severe. The Charles procedure was done on three patients in this group. Eleven patients required three surgical procedures or more to reduce the size of the limb effectively, and in many of these patients, the thigh was involved as well, necessitating surgery in this location as well as on the lower leg (Table 2).

Results of Surgery. – Twenty-nine of the 32 patients were traced from 1 to 22 years, the average being 6 years. Three subsequently died; their deaths were unrelated to either the operation or the disease. Thus, the results of surgery are based on operations on 30 limbs of 29 patients.

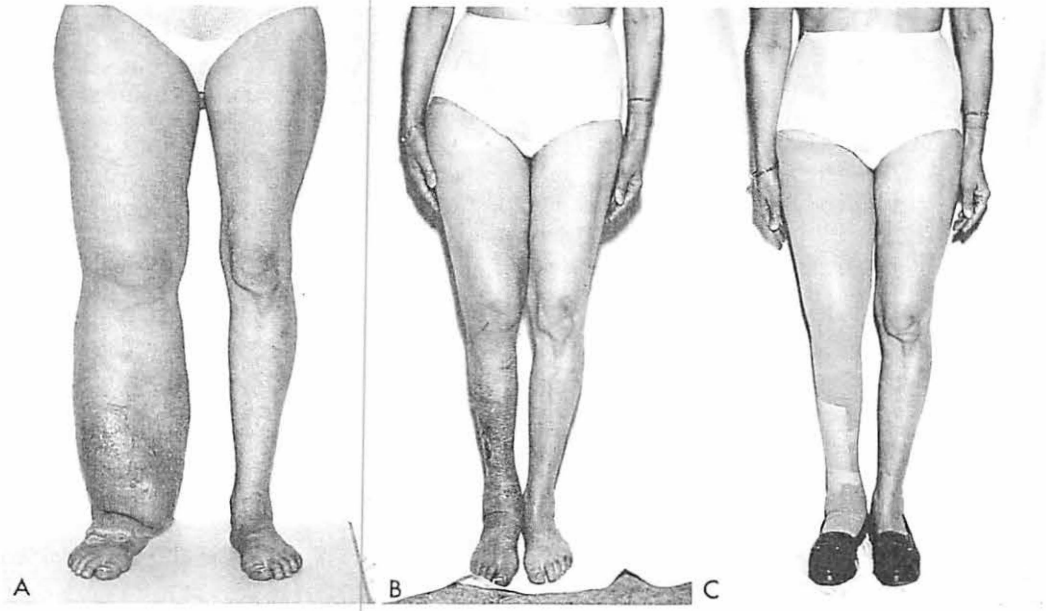

Fig. 1 Lymphedema praecox of 16 years' duration in a 35-year-old woman.
A, Preoperative view. B, Two months after first- and second-stage Homans procedure from ankle to knee. Excellent result. C, Patient wearing fitted elastic stocking, 2 months after operation.

Table 1 Surgical Procedures for Lymphedema

Procedure	Idiopathic lymphedema			Secondary lymphedema		
	Praecox (32 patients)*	Congenital (11 patients)*	Late form (2 patients)	Obstructive (11 patients)	Inflammatory (8 patients)	Total (64 patients) †
Homans						
Single-stage	10 (9) ‡	5 (4)	2 (2)	4 (4)	5 (5)	26 (24)
Double- stage	28 (14)	10 (5)	2 (1)	8 (4)	2 (1)	50 (25)
Triple -stage	24 (8)	3 (1)	3 (1)	3 (1)	0 (0)	33 (11)
Macey	4 (4)	0 (0)	0 (0)	3 (1)	0 (0)	7 (5)
Charles	3 (3)	2 (1)	2 (2)	1 (1)	2 (2)	10 (9)
Minor plastic operations	4 (4)	0 (0)	2 (2)	2 (2)	1 (1)	9 (9)
Multiple operations (3 or more)	... (11)	... (1)	... (2)	... (3)	... (0)	... (17)
Total	73 (53)	20 (12)	11 (10)	21 (16)	10 (9)	135 (100)

* One patient had bilateral involvement
† Five patients also had lymphangiograms
‡ Numbers in parentheses represent patients

Table 2 Sites of Surgery for Lymphedema

Site	Idiopathic lymphedema			Secondary lymphedema		Total
	Praecox	Congenital	Late form	Obstructive	Inflammatory	
Lower leg	48	15	4	8	5	80
Thigh	11	2	3	0	2	18
Lower leg and thigh	11	2	2	3	3	21
Foot	3	0	2	2	0	7
Forearm	0	1	0	1	0	2
Upper arm	0	0	0	2	0	2
Arm	0	0	0	5	0	5
Total	73	20	11	21	10	135

The results were classified as follows: (1) excellent if the patient had no residual swelling or if the size of the leg was smaller than normal or was normal; (2) good if the patient had slight residual swelling; (3) fair if the size of the extremity was reduced, although with persistent swelling; and (4) poor if surgery had not benefited the patient or if the size of the extremity had increased.

The results of surgery were excellent in 8 patients, 1 of whom underwent bilateral surgery; good in 12; fair in 4; and poor in 5 (Table 3). In 70 %, therefore, the results were excellent or good, and in 83 %, the operation helped reduce the size of the involved limb. Of the five patients with poor results, two had a single-stage Homans procedure only and did not return for a necessary second stage, and one patient had three subsequent procedures at another institution, with an excellent result. Thus, only 2 of 27 patients treated adequately had poor results, or more than 90 % had satisfactory surgical results.

Influence of Adequate Support on the Results of Surgery. – *Ghormley* and *Overton* (6) noted that the wearing of adequate elastic support increased the degree of benefit after operation. Of our 25 patients with excellent, good, or fair results, 20 wore adequate elastic support after operation (Table 4).

Congenital Lymphedema

Eleven patients (7 men and 4 women) had congenital lymphedema (17.2 % of the series of 64 patients): 9 the simple type and 2 with Milroy's disease. The onset of swelling for most of the patients was at birth to 1 1/2

Table 3 Results of Surgical Treatment of Lymphedema

Type of lymphedema	Followed patients	Result			
		Excellent	Good	Fair	Poor
Praecox	29*	8*	12	4	5
Congenital	9	4	3	1	1
Late form of idiopathic	2	0	0	1	1
Obstructive	10	0	6	2	2
Inflammatory	6	1	1	0	4
Total	56	13	22	8	13

* One patient had bilateral involvement

Table 4 Correlation Between Results of Surgery in Lymphedema Praecox and Wearing of Adequate Support

Result	Support		Total
	Adequate	Inadequate	
Excellent	8	1	9
Good	8	4	12
Fair	4	0	4
Poor	4	1	5
Total	24	6	30*

* One patient had bilateral involvement

years of age, whereas the average age at surgery was 26.5 years. All 11 patients had huge extremities.

Surgery. – Surgery was done on 11 lower extremities and 1 upper extremity; one patient had both lower extremities involved. A total of 20 operations were performed (Table 2), including a Homans procedure and two Charles procedures on the same patient (Table 1).

Results of Surgery. – Of the 11 patients, 9 were traced for an average of 9 years; the range varied from 1 year to 19 years. Four patients had excellent results, three had good results, one had a fair result, and one had a poor result (Table 3).

Idiopathic Lymphedema

Two patients (a male and a female, 3.1 % of the series) had lymphedema that was neither congenital lymphedema nor lymphedema praecox. Each had severe swelling in a lower extremity.

Secondary Lymphedema

Eleven patients (seven women and four men) had obstructive lymphedema (17.2 % of the series). The various causes are listed in Table 5. The average age at the onset of swelling was 37 years, and the average age at surgery was 45 1/2 years, reflecting the late onset of swelling. Four patients had swelling in the arm after radical mastectomy (Fig. 6), and seven had swelling in the lower extremity. The swelling of these patients tended to be severe, more so than in other groups.

Table 5 Causes of Obstructive Lymphedema

Cause	No. of patients
Postmastectomy	4*
Mass removed from groin or lower abdomen	2†
Radical lymphadenectomy in inguinal area for lymphosarcoma	1
Incision and drainage of thigh abscess	1
Fracture of femur	1
Removal of bone tumor and postoperative radiation therapy	1
Questionable malignancy (late onset of lymphedema: age 50 yr)	1
Total	11

* Two patients received postoperative radiation therapy

† One patient received postoperative radiation; bilateral leg swelling.

Surgery. – Twenty-one total operations, including one Charles procedure, were done on the 11 patients with secondary lymphedema, three operations or more being necessary on each of 3 patients (Tables 1 and 2).

Results of Surgery. – As expected, the results in this age group of patients were not as good as in the previous groups. There were 10 patients with adequate follow-up; 1 had died from an unrelated cause 3 years after surgery. Six of the 10 patients had good results, 2 fair results, and 2 poor results (Table 3).

Inflammatory Lymphedema

Eight patients (12.5 % of the entire series), three women and five men, had inflammatory lymphedema of the lower extremity after an initial episode of cellulitis and lymphangitis. Indications for surgical treatment were the large size of the involved limbs and multiple severe recurring cellulitis in seven and the size alone in one patient.

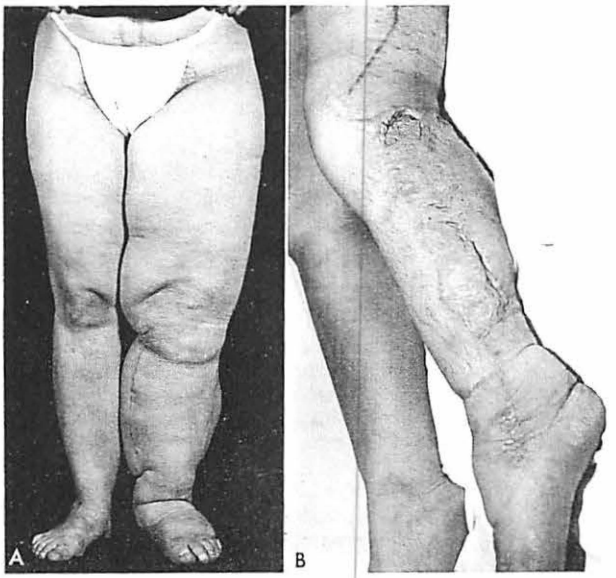

Fig. 2 A 43-year-old woman with lymphedema praecox since age 13 years.
A, Recurrent lymphedema 15 years after two-stage Homans procedure to leg.
B, One year 9 months after multiple procedures to thigh and calf, including Charles procedure. There is adequate reduction in size of limb, but warty excrescences are present in skin graft; this occasionally is seen after the Charles procedure.

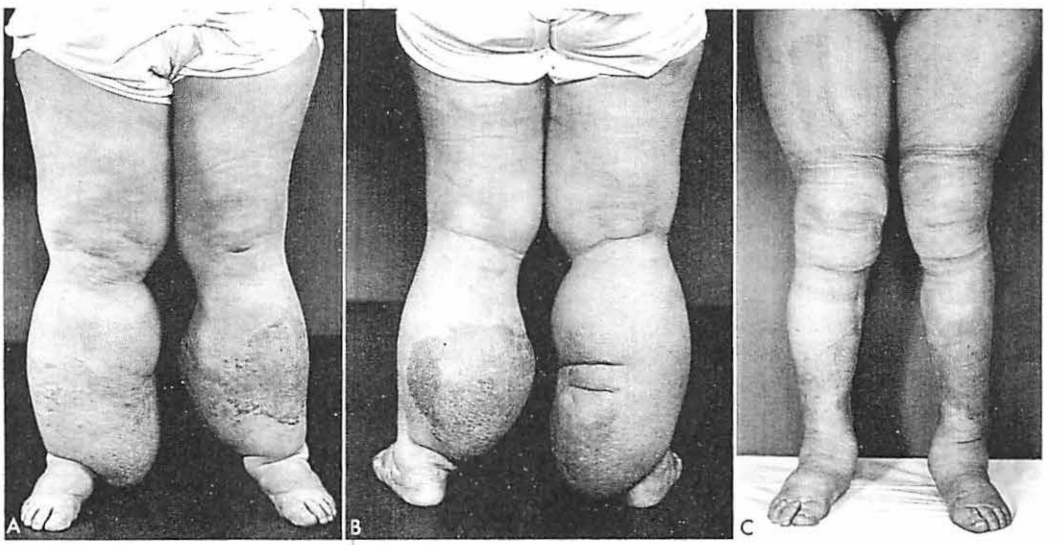

Fig. 3 A 43-year-old woman with severe lymphedema praecox (since age 16 years) complicated by multiple episodes of lymphangitis and cellulitis.
A, Preoperative anterior view.
B, Preoperative posterior view.
C, Postoperative view (3 years) after first-stage Homans procedure to medial aspect of both legs from ankle to knee. Excellent result; no further episodes of lymphangitis or cellulitis.

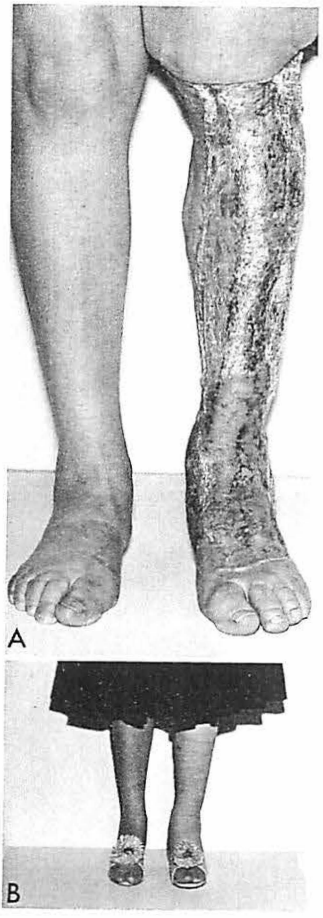

Fig. 4 A 32-year-old woman with severe lymphedema praecox of 10 years' duration. Two previous first- and second-stage Homans procedures did not effectively reduce swelling of limb.
A, Six months after Charles procedure, with removal of 1 lb of lymphedematous tissue.
B, Ace bandages on affected limb. Size of limb is normal. Excellent result.

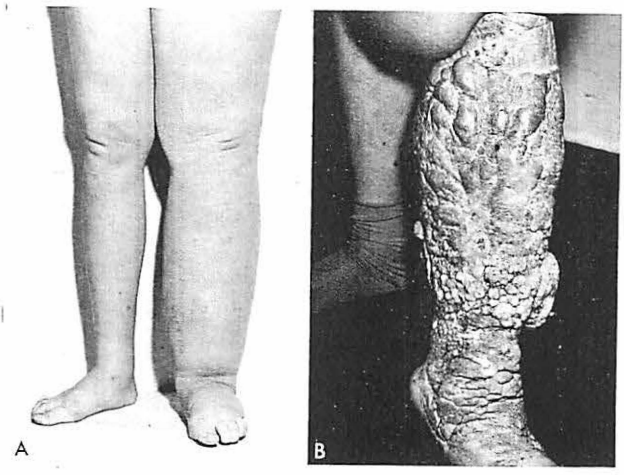

Fig. 5 A 29-year-old man with lymphedema praecox of 15 years' duration.
A, Preoperative view.
B, Eight years after Charles procedure. Severe nodular excrescences of skin graft of calf and foot, with recurrence of lymphedema. Poor result.

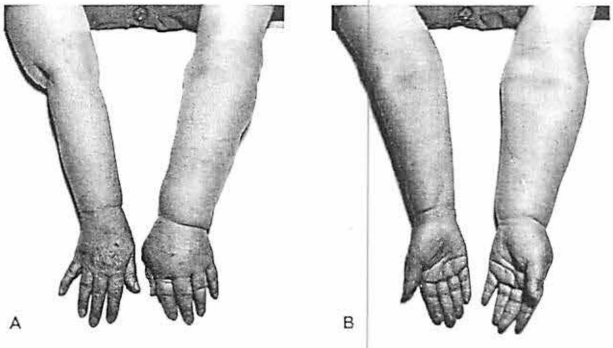

Fig. 6 A 43-year-old woman who developed severe postmastectomy lymphedema of left arm in 1937 after radical mastectomy for carcinoma of left breast. She had two Homans procedures to her arm, with removal of a total of 1,175 g of lymphedematous tissue. Result was fair although another modified Homans procedure on dorsal part of arm from elbow to wrist was later performed. Patient is alive and well, without evidence of recurrent carcinoma, and has had a good result after the last procedure.
A, Dorsum of arm.
B, Volar aspect of arm.

Tabele 6 Complications of Surgery for Lymphedema

Type of lymph-edema	Patients	Oper-ation	Hema-toma	Infection	Necrosis	Pulmonary embolus	Phlebitis	Miscellaneous	Death	Total complications
Praecox	32	73	19(2)*	1(0)	3(0)	2	1	1 (paresthesia) 2 (keloids)	0	29
Congenital	11	20	2(1)	0(0)	8(4)	0	0	None	0	10
Idiopathic	2	11	2(1)	1(0)	1(1)	0	0	1 (tight heel cords)	0	6
Obstructive	11	21	2(1)	3(2)	0(0)	0	1	1 (paroxysmal atrial tachycardia)	0	7
Inflamm-atory	8	10	4(0)	2(1)	0(0)	0	0	1 (lt. ulnar paresthesia)	0	7
Total	64	135	29(5)*	7(3)	12(5)	2	2	6	0	59

*Number in parentheses represents those requiring grafts.

Surgery. – Six of the eight patients were followed up from 1 to 21 years, the average being 12 years. A total of 10 operations, including two Charles procedures, was done (Tables 1 and 2). The poorest results have occurred after surgery for inflammatory lymphedema. In most of these patients, the cellulitis and lymphangitis continued unabated after the operation, and the swelling eventually returned.

Complications of Surgery

The most common complications were related to the wound and consisted of hematomas, necrosis of skin flaps, and wound infections (Table 6). The operative mortality rate of 0 % in the 135 surgical procedures established the safety of these radical operations. Occasionally, tight heel cords have been seen after the Charles procedure, but this was not a problem in our series. A more distressing finding on several occasions was verrucous hyperplasia of the skin grafts around the foot, necessitating removal of the warty nodular skin and regrafting.

Pathologic Findings

Review of the formalin-fixed gross tissues excised from the extremities revealed no apparent difference among the various types of chronic lymphedema. Each type had roughening and the epidermal surfaces that clinically were "pigskin-like" (Fig. 7A). On cut section through the tissues, the dermis was usually firm and grayish, as was the deep fascia, whereas the intervening subcutaneous fat lobules were light yellow and at times projected between the fibrous septae (Fig. 7B). In several specimens that were examined in the fresh state, lymph exuded from the dermis and subcutaneous tissues on compression,

Table 7 Gross Thickness of Excised Formalin-Fixed Tissues

Type of lymphedema	No. of patients measured	Dermis, cm Range	Dermis, cm Average	Subcutaneous, cm Range	Subcutaneous, cm Average
Praecox	28	0.2 – 0.8	0.4	1.0 – 7.0	2.5
Congenital	15	0.2 – 1.0	0.5	1.2 – 5.4	2.5
Inflammatory	5	0.2 – 0.8	0.4	2.2 – 3.2	2.5
Obstructive	8	0.2 – 0.7	0.4	0.8 – 8.0	3.7

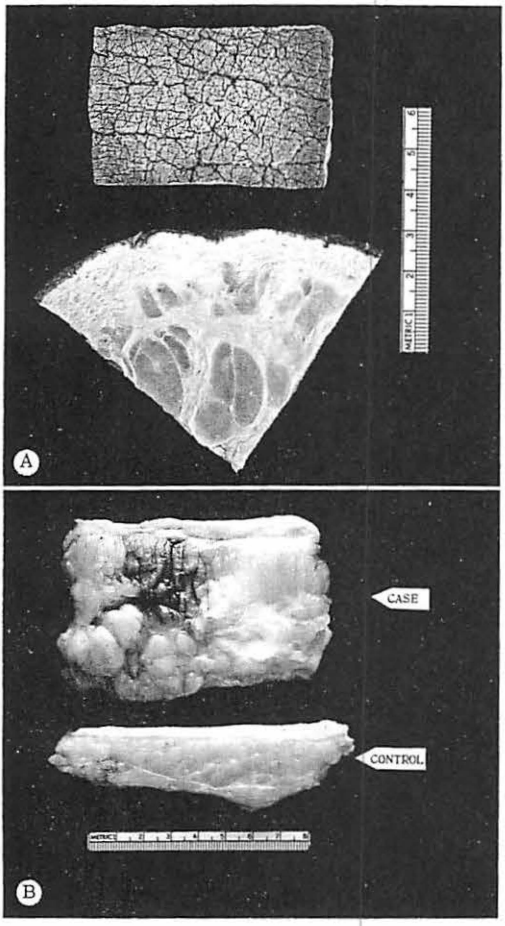

Fig. 7 Gross appearance of lymphedema.
A, Congenital lymphedema (40 years) showing marked irregularity and thickening of skin surface above, and dermal subcutaneous fibrosis outlining fat lobules with trabecular bands.
B, Lymphedema praecox. Skin and subcutaneous tissue seen in cross section and compared with nonlymphedematous tissue from patient without lymphedema.

although none was "sponge-like". Measured dermal and subcutaneous tissue thicknesses were comparable in all types (Table 7 and Fig. 8).

Microscopically (Table 8), the epidermis usually was undulated and had a atrophic changes with thinning in areas overlying the widened dermal papillae and variable prominence of epidermal pegs. Some hyperkeratosis was common (Fig. 9A), which occasionally filled keratin pits. Only one specimen had lymphedema verruca formation; this specimen was from a patient with congenital lymphedema.

Dermal edema with separation of collagen or elastic fibers was observed in most specimens and was similar (moderate to severe) in the various types of lymphedema. Dermal fibrosis of a moderate or severe degree was more frequent in inflammatory lymphedema (89 %) but was present in about two-thirds of the other three types.

Perivascular (lymphatic or venous) inflammatory cell infiltration (lymphocytes, plasma cells, and at times a few eosinophils) was noted in some specimens of each of the various types but was observed more frequently in inflammatory lymphedema and in the praecox type.

Subcutaneous fat was abundant in all specimens and rarely had an obvious edematous appearance. Prominent subcutaneous fibrous septae were seen in about half of the specimens.

Lymphatic channels were difficult to assess in routine section (hematoxylin and eosin, elastic-van Gieson, and Mallory-Heidenhain stains). Some sections of each type of lymphedema had few deep lymphatic channels and

Table 8 Histopathologic Changes in Chronic Lymphedema

Type of lymphedema	No. of patients	Epidermal atrophy	Dermal		Perivascular inflammation	Subcutaneous		Lymphatic vessels			Thickened blood vessels
			Edema	Fibrosis		Prominent septae	Abundant fat	Few deep	Dilatation	Prominent deep	
Praecox	32	22	16	20	29	14	32	15	19	5	16
Congenital	15	15*	10	10	14	8	15	6	11	5	8
Inflammatory	9	6	6	6	9	4	9	3	3	1	4
Obstructive	9	8	6	6	7	4	9	5	7	1	4

* One patient with verrucous change.

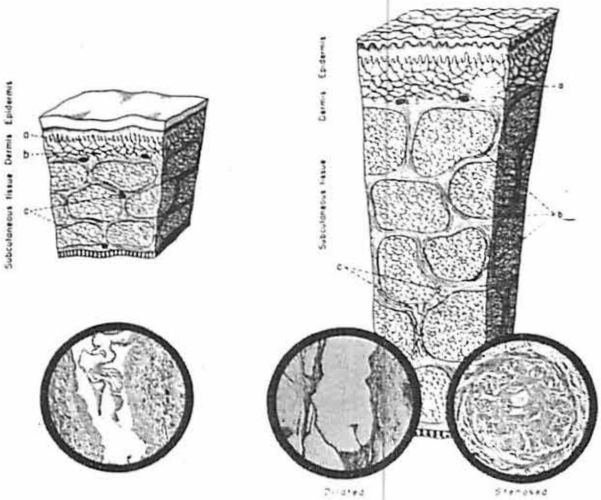

Fig. 8 Normal tissues (*Left*) compared with findings in lymphedema (*Right*). Superficial (a) and deeper (b) layers of dermal lymphatic networks and deep subcutaneous lymphatic vessels (c).

infrequent focal obliteration. Superficial dermal lymphatic vessels of the subpapillary network level or of the lower superficial network at the level of the appendages could be identified only if they were dilated, and this was a more frequent finding in obstructive and congenital lymphedema than in the others. Focal dilatation (lymphangiectasis) of deep, subcutaneous lymphatic channels, usually adjacent to septal blood vessels and small nerves, was seen in one-third of the specimens of congenital lymphedema (Fig. 9B) and 16% of those of praecox lymphedema (Fig. 9C), but was also noted in one specimen of inflammatory and one of obstructive lymphedema (Fig. 9D). In four specimens of congenital, four of praecox, and one of obstructive lymphedema, the dilated deep lymphatic channels had thin walls with little smooth muscle and were slightly multilocular, somewhat resembling focal cavernous lymphangioma.

Thickening of smooth muscle coats and narrowing of the lumen of subcutaneous blood vessels, which at times had an edematous appearance, were noted especially in veins and in a comparable incidence in the various types of lymphedema.

Comment

In our series of 64 patients with lymphedema of various types who had a total of 135 operations, including the Homans, Charles, and other procedures, indications for surgery were usually the huge size of the extremity or recurring cellulitis that in time became disfiguring. We agree that surgery is not indicated for mild lymphedema, but it should not be delayed until the involved extremity becomes massive. This delay increases the magnitude of necessary surgery, increases the postoperative morbidity, and decreases the ultimate benefit from surgery. Multiple procedures may be necessary to reduce the size of the extremity effectively. In addition to evaluating the clinical procedures, we evaluated the morphologic aspects of the excised tissue.

Of the total group, the results of surgery were excellent, good, or fair in about 75 %. In 72% of patients with congenital lymphedema or lymphedema praecox, the results of surgery were good or excellent, and the swelling was effectively reduced in 85 % of these patients. Generally, we do not recommend surgery for patients with secondary lymphedema unless the size of the limb prevents unsurmountable problems to the patient. In patients with inflammatory lymphedema, surgery should be undertaken with great reservation and only after maximal control of the complicating infection has been achieved.

The need for adequate postoperative support was reemphasized by our study because,

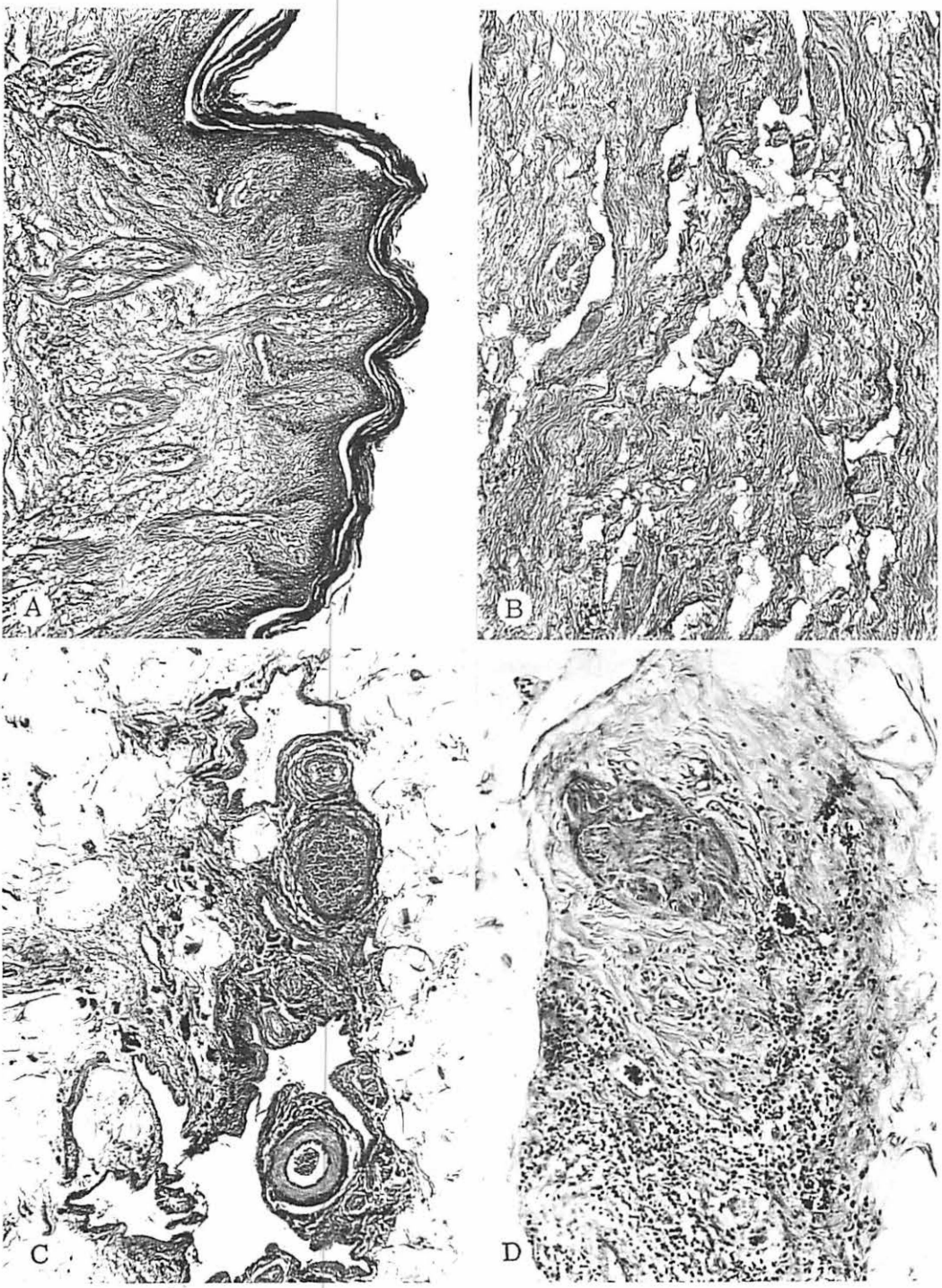

Fig. 9 **A**, Congenital lymphedema with hyperkeratosis, epidermal thickening, and edematous separation of dermal fibroelastic tissues. (Hematoxylin and eosin; x85.)
B, Marked focal lymphangiectasis resembling cavernous lymphangioma (same case as in Figures 7A and 9A). (Hematoxylin and eosin; x95).
C, Lymphedema praecox with irregular dilatation of deep lymphatic vessels (lymphangiectasis). (Hematoxylin and eosin; x95.)
D, Obliteration of deep lymphatic vessels due to sclerosing lymphangitis in lymphedema complicated by right recurrent episodes of lymphangitis and cellulitis. (Hematoxylin and eosin; x112.)

although surgery reduces the size of the extremity, it does not affect the basic lymphatic defect. Adequate support prevents a recrudescence of swelling and allows tissue shrinkage.

Surgery also has been helpful in reducing or eliminating recurring episodes of cellulitis and lymphangitis. Intermittent antibiotic therapy as described by *Babb* et al. (1) should be prescribed for all patients with recurring lymphangitis.

Although extensive, the excisional operations employed in our series are very safe; there were no operative deaths. After surgery for lymphedema, no extremity will be exactly like its mate but the return of physiologic function and reduction of size of the involved extremity far outweigh cosmetic impairment, which may be lessened by the use of adequate elastic support.

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