

ELEPHANTIASIS NOSTRA*

BY GEORGE P. MULLER, M.D., AND CLAUS G. JORDAN, M.D.

OF PHILADELPHIA, PA.

FROM THE SURGICAL DIVISION OF THE UNIVERSITY HOSPITAL

ELEPHANTIASIS, called Herculasian disease by the ancient Greeks, has always been a very spectacular affection. Up until the Middle Ages, as Virchow¹ tells us, it was often confused with leprosy due to its similarity in certain phases with the tissue hypertrophy of lepra. Just as the man with elephantiasis was banished from society by certain ancient people who thought him to be a leper, so did certain African tribes make saints out of the victims of this disease because they thought it a special favor of the gods to have a much enlarged scrotum or leg. Even today in more civilized countries we often see an elephant man exhibited in a circus or side show. Sir Frederick Treves² has written a vivid and sad story of an elephant man.

Up to the beginning of this century elephantiasis was a medical curiosity and the only rôle the surgeon played was to undertake the last step of amputating the affected part when it threatened to become larger than its owner. With the rapid rise of surgery this disease has received its share of attention and many different operations have been devised.

For the proper understanding of the treatment one must know first the nature and cause of this disease. As we will see subsequently there is only one elephantiasis but there are innumerable causes for it. Elephantiasis is a chronic inflammatory fibromatosis of the dermal and hypodermal tissues, either preceded or accompanied by venous and lymphatic stasis. Mere venous and lymphatic stasis will not produce true elephantiasis; there must be a second element which is the infection. In the past twenty years there has been much controversy as to the rôle lymphatic obstruction and infection play as etiological factors. Matas³ believes both are of equal importance. Unna⁴ and Saboroud⁵ believe that infection alone by the streptococcus can produce elephantiasis. Kuntzen,⁶ who writes from Payre's clinic, believes that lymphatic obstruction plays the major rôle, the erysipeloid streptococcal infection being secondary and acting only to intensify the course of the disease.

According to the numerous causes we could classify this disease into many varieties. In order to avoid confusion, however, we can distinguish between the elephantiasis which is caused by the filaria and that which is not. In the true filarial elephantiasis the lymph channels are obstructed by the parasites, thus producing fertile soil for the invasion of the streptococcus. It is now generally recognized that the tropical elephantiasis is frequently not

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caused by filaria. The reason for the greater frequency of elephantiasis in the tropics is the greater exposure of the skin to infection.

In northern latitudes the disease is most often called elephantiasis nostra, distinguishing it from the tropical variety, but there are authorities who give the by-name of nostra only to those cases in which no apparent cause can be detected. Any factor, such as an injury involving the circumference of a limb at its root, or tumors, be their origin carcinoma, tuberculosis or syphilis, involving or pressing on the lymphatics, may ultimately cause elephantiasis, by causing lymphatic stasis. Very often a phlebitis or an inguinal bubo or the cicatricial contraction after herniorrhaphy will with a superadded streptococcal infection produce the disease. In certain cases as for example Milroy's disease, heredity must also be considered an important factor in the etiology. Then we have the true surgical elephantiasis which follows radical breast amputations, and which Halstead⁷ believes is due to infection of the flaps at the time of operation, but which many others believe is nothing but a sign of the recurrence of the malignancy. Kuntzen, who has studied forty-seven cases of the non-tropical variety, includes under the heading of genuine elephantiasis nostra only those cases which begin insidiously in young people at the age of puberty and in which the disease always involves the lower extremity or the external genitalia. It is his belief that because it begins at puberty or occasionally near pregnancy that there is some endocrine factor involved in its production. Kuntzen further emphasizes the localizing element in these cases. As the lymph flows by muscular and valve action the hydrostatic element in the production of this disease is very evident. It is seldom that we find elephantiasis nostra in other than the dependent parts of the body, such as leg, scrotum, or penis. Concerning the rôle of infection, Kuntzen, after careful study of his forty-seven cases, found that in only one case did the typical erysipeloid attack precede the lymphoedematous swelling. He believes that the erysipeloid infection has nothing to do with the cause of elephantiasis but that it is of grave importance for the subsequent course of the disease. It is our belief that without the erysipeloid attacks there may be lymphoedema but there cannot be that true fibromatosis so characteristic of elephantiasis.

A very interesting study of sixty cases of elephantiasis has been made in Porto Rico by Suarez.⁸ He took cultures from what he believed to be the infectious foci of ten cases and got nine positive cultures of hemolytic streptococcus and hemolytic staphylococcus. On the other hand, he took fifty cultures from the tissues of the affected limbs and got only one positive culture.

The typical course of elephantiasis might be described as follows: Recurrent attacks of elephantoid fever, with chills, a temperature around 103°, general malaise, a painful red swollen limb, with enlarged regional lymph-nodes, the entire picture very closely resembling erysipelas. In fact, one of our cases was first treated as erysipelas. After from two to eight days the fever subsides and the patient feels well but the swelling of the part does

not disappear entirely. After about a year or a few months seldom oftener than every month the attack will recur and the swelling subsides less each time. The dermal and hypodermal tissues will hypertrophy and the underlying soft tissues will unite with the skin to form one brawny hard induration and the typical elephantoid proportions will develop. The whole process extends over a period of years and once the erysipeloid habit is developed it will not stop until the elephantiasis is fully developed. There are certain cases where the constitutional symptoms are lacking, but when these patients are carefully questioned they always remember their attacks of redness and swelling. In a fully grown elephantiasis one observes a pillar-like extremity or a scrotum as big as a head. The skin is hard, does not fold or pit, and shows certain trophic disturbances such as nail changes, hyperkeratosis and scaling. Occasionally there will be ulcerations with considerable lymph drainage. The microscopical picture shows a typical hypertrophy of the skin and subcutaneous tissue with an increase in the collagen bundles. The deep fascia is often very much thickened. The vessel walls are hypertrophied and show considerable perivascular round-cell infiltration. The lymph-vessels themselves do not show much change. Interesting and of great importance for the success of the surgical treatment is the fact that the tissue hypertrophy and sclerosis stops with the deep fascia. The disease involves only the structures of the superficial venous and lymphatic system.

The diagnosis of elephantiasis is rather simple. It is easily differentiated from leprosy because the latter is a systemic disease of slow progress and long standing. Recently, Reichert,⁹ of San Francisco, and Golden, in Porto Rico, have taken soft-tissue X-rays and they find a very marked dense trabeculation of the soft parts and in the case of filarial elephantiasis they find the calcified filaria.

The treatment of this disease should be both medical and surgical for each one of them alone does not produce very permanent results. The medical treatment consists of:

- (a) Baking and massage (usually pre- and post-operatively).
- (b) Elevation of limb (usually pre- and post-operatively).
- (c) Compression of limb by elastic bandages.
- (d) Injection streptococcus polyvalent serum.
- (e) Vaccine therapy.
- (f) Fibrolysin.
- (g) Foreign-protein therapy (typhoid, aolin, and whole blood).

There are several cases reported in the literature, especially in more recent times, which have been treated very successfully with vaccine and anti-streptococcus serum alone and it is worth while to employ this treatment in every case possible. A recent case in our own series has shown this. This medical treatment, if it can stop the acute attacks, might be sufficient. In more advanced cases, however, if once the elephantiasis proper has developed, medical treatment is of importance only so far as it helps to prepare

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the patient for operation and again post-operatively helps to prevent those very disastrous erysipeloid attacks. The sixteen cases which have been treated by Aubin¹⁰ with streptococcus anti-serum have apparently shown some good immediate results. It is, however, a question as to the permanent results. Preventive treatment is important, namely, to prevent infections of the extremities such as ulcers and phlebitis, also to be careful in the removal of inguinal glands in hernia operations, and not to apply circular bandages in cases where there is danger of venous or lymph stasis.

The earliest reference to any surgical treatment other than amputation was made in 1851, when Carnochan and Morton in this country proposed the ligation of the main artery. This operation, besides risking gangrene, did not produce the desired results. In 1900, Mikulicz and others started to excise large cuneiform pieces of tissue from such limbs and they obtained occasional good results. Finally, Lanz, of Holland, starting with the idea that lymphatic stasis was the underlying evil, proposed a very radical operation for the establishment of new lymph-channels. He made an incision down to the bone, trephined the bone and placed strips of deep fascia into the trephine holes. Lanz obtained excellent results, but his operation was too radical. It is from this operation and from one which Handley proposed in 1908 that the Greek surgeon Kondoleon¹¹ adapted his operation. Handley proposed to establish new lymph drainage by placing silk threads in the subcutaneous tissues, starting at the wrist and extending up into the healthy tissues of the shoulder. The immediate results of his operation were very successful, but later investigations showed that the silk thread was represented by nothing more than fibrous strands and had ceased to give lymphatic drainage.

With Lanz's operation in view, Kondoleon, in 1912, proposed the following operation. Based on the very reasonable theory that in elephantiasis the superficial lymphatic system is diseased and blocked, while the deep lymphatic system is still sound and functioning, he proposed by the removal of the deep aponeurosis covering the muscles to open the way for the formation of new lymph channels between the deep and superficial lymphatic systems. His operation consists of an incision from the trochanter to the external malleolus, down to the deep fascia and the removal of a wide strip of this fascia. The same procedure is repeated on the inner side of the limb. The Kondoleon operation was first performed in this country by Matas in 1912 and he modified it slightly by excising considerable portions of skin and subcutaneous tissue and by sewing the deep fascia against the muscle in order to prevent the reformation of this fascia. Sistrunk¹² at The Mayo Clinic further modified the operation by removing much larger proportions of skin and subcutaneous tissue, thus trying to reestablish the normal proportions of the limb. Payre,¹³ in Germany, who claims to have originated his operation at about the same time as that by Kondoleon, employs an operation similar to that used by Sistrunk. Payre, who has done the same number of operations as Kondoleon, namely, twenty, stresses the pre-operative treat-

ment especially. He advises long-continued bed rest, often as long as four to five weeks, with bandaging and elevation of the limb in order to shrink it as much as possible. The smaller the leg at the time of operation the more tissue can be removed and the better the result. He also lays emphasis upon the fact that as much as possible of the skin should be removed in order to have a sufficient pressure of the skin upon the underlying tissues. Payre, and his co-worker, Kuntzen, take great care in preparing the skin of the affected limb, cleaning it every day and applying sterile bandages for almost a week preceding operation. Sistrunk and Auchincloss,¹⁴ in New York, have independently advocated the correction of the deformity and the removal of as much as possible of the diseased tissues as one of the important features of a modified Kondoleon operation.

The actual technic of the operation consists of an elliptical incision of the skin, removing all excess skin; reflection of skin flap at least six centimetres laterally and then removing a large section of subcutaneous tissue and deep fascia; exposing the muscle and bringing it into direct contact with the skin.

If we now review critically the results of the surgical treatment, we find that the splendid improvement which the originators of this operation had hoped to obtain has not been fulfilled. Kondoleon, who reviewed his twenty cases, in 1924, stated that some of them were improved for as long as two years and then there was a recurrence of attacks. He states, however, that after the operation in spite of the attacks the limb or scrotum always returns to normal size again, and he believes this to be definite proof of the establishment of new lymph-channels by the operation. Summarizing his experience with the operation, he believes there is definite improvement after operation but not a "restitutio ad integrum." Again, Kuntzen, in reviewing twenty-two cases, shows that their immediate results were very good, but that the permanent results are not so encouraging. Out of sixteen cases of the lower extremity, two were completely cured; six were much improved, four very little improved and three were not improved at all. The cases showing the largest improvement were cases of secondary elephantiasis of long standing. The results of the radical operation for elephantiasis of the scrotum and penis have been better. Sistrunk, after ten years of experience, feels that the operation has definite value. Auchincloss, in Porto Rico, has had the same good immediate results, but he also has had many recurrences. The great factor in recurrence is the reappearance of the erysipeloid attack.

CASE REPORTS.—CASE I.—Mrs. M. L., aged fifty-three years, white. First admitted September 12, 1930, to Doctor Muller's service, Misericordia Hospital. Tremendous swelling of right leg. Eighteen years ago, when three months pregnant, the patient noted a generalized swelling of right foot and leg. This swelling would pit on pressure. The swelling persisted during the remainder of the pregnancy but did not become worse. Bed rest after parturition improved the leg, but getting up produced the same swelling. In spite of all palliative treatment, during the last eighteen years, the size of the leg became

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gradually worse and in December, 1929, it attained its present proportions. (Fig. 1.) The patient was at that time unable to walk or even to perform her household duties on account of the size of the leg. It began to ulcerate and discharge as much as a quart daily of clear white lymph. The patient has never been farther south than Washington. She states that three months prior to her initial swelling she fell and injured her spine. At the time of her examination by us there was, however, no evidence of injury. Her previous medical history was negative. No elephantiasis in family, apparently only one abortive attack of erysipelas. Mother of four children. Physical examination entirely negative except for her right leg. The lower leg is most markedly elephantoid. The



FIG. 1.—(Case I.) Elephantiasis of right leg. Anterior view. Before operation.

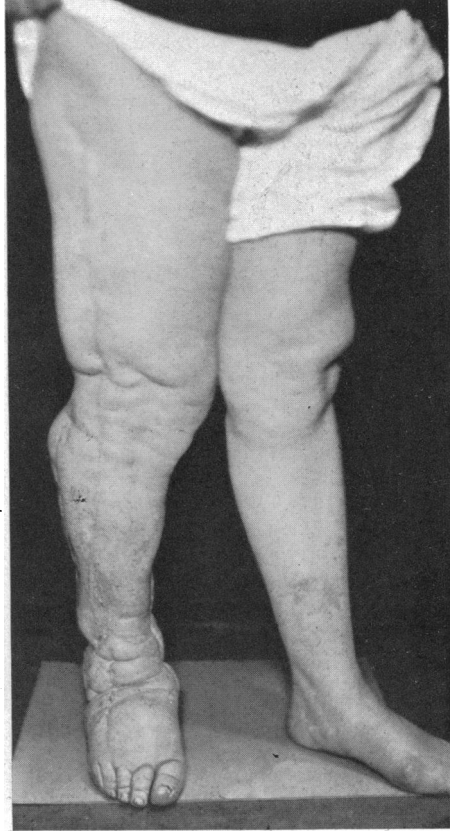


FIG. 2.—(Case I.) Elephantiasis of right leg one and a half years after operation.

skin has become very much thickened, it does not pit on pressure, and in the ankle region is covered with numerous wart-like thickenings. The patient was prepared for a week and then the first-stage Kondoleon operation was performed. The skin, subcutaneous tissue, fat, deep fascia were removed from the trochanter to the external malleolus. From trochanter to knee a strip about two and one-half inches was removed and from the knee down the strip varied from two and one-half to five inches. A month later the second-stage Kondoleon on the inner side of the leg was performed. The healing of both scars progressed slowly but steadily and with the help of a small skin graft she was finally discharged eleven weeks after her first admission. The patient was admitted three times during the last one and one-half years for minor plastics on the same leg and at the present time her right leg has almost the same dimensions as her left one. (Fig. 2.)

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She is again able to do her housework and can walk. She still has a small granulating area over her external malleolus, but this is healing steadily. For the patient's comfort it is also necessary that she still wears an elastic bandage on her right leg. She has never had any erysipeloid attacks during the last ten years.

CASE II.—W. S., boy, aged thirteen years. First admission, September 27, 1930, Doctor Muller's service, University Hospital. Recurrent painful swelling of both legs. Without previous disease or ill health the boy's legs became painful and swollen about a year before first admission. The condition lasted only for a few weeks in the right leg, but remained for about two months in the left leg. The painful swelling with redness

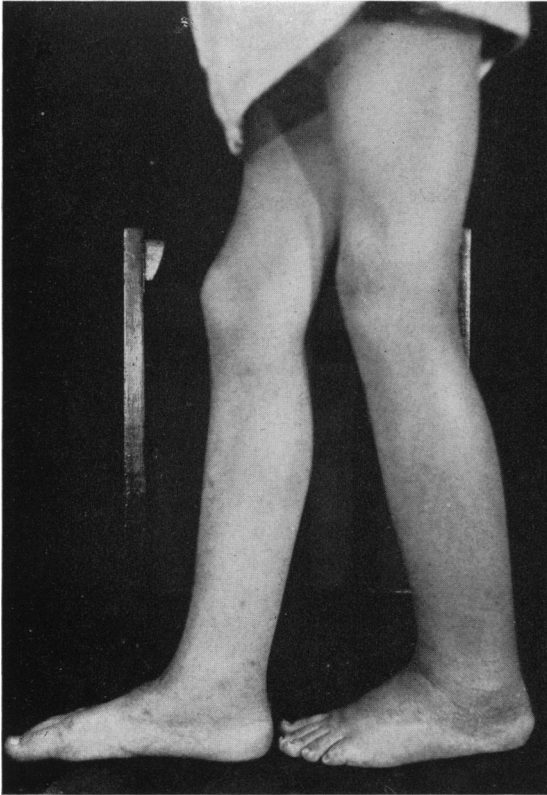


FIG. 3.—(Case II.) Elephantiasis nostra of left leg before operation.

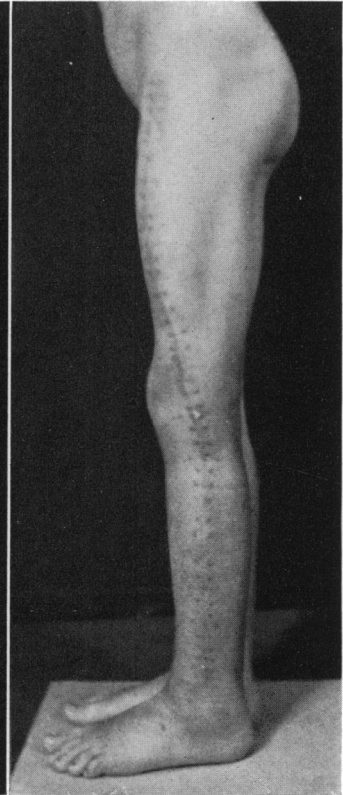


FIG. 4.—Four months after operation.

and fever returned about every three to four months in the left leg. In the interim, with otherwise good health, the left leg remained, however, somewhat swollen but was not as large as during acute attacks. On September 25, the patient had another attack, which began with chills and fever and was followed about eight hours later by redness and swelling and severe pain. This pain and redness started at the internal malleolus and ascended up the leg as high as the iliac crest. There were no history of trauma and no hereditary factor present. Patient had scarlet fever in June, 1928. Tonsils had been removed several years previous. Physical examination was negative except that the left foot and leg were swollen considerably. The skin of the foot and ankle was thick, dry, coarse and scaling with a tendency to eczema and ulceration. The foot and leg were not tender. A diagnosis of elephantiasis nostra due to repeated attacks of lymphangiitis was made. Pre-operative treatment was started and with the leg elevated the swelling

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decreased considerably. The picture shown (Fig. 3) was not taken till two weeks after admission. Kondoleon operation October 16, 1930. The tissues of the lower leg had a decidedly bony feel. The boy's wound healed perfectly and he was discharged November 10. In December, 1930, the patient returned for the second-stage Kondoleon operation. (Fig. 4.) The swelling at this time was much less. Seen in follow-up clinic April, 1931, there was still slight swelling over the external malleolus of left leg. He was going to school, was still wearing an ace bandage. Had not had any attacks since operation. On June 11, 1931, he woke up with a sudden pain in the right foot, followed by a chill and swelling of his right foot this time. The red area spread up the knee in four days. He was admitted with a temperature of 103° and he was treated and diagnosed as erysipelas by the receiving ward intern who did not know his previous history. The left leg at this time was perfect. About the right knee there was an area of redness simulating erysipelas and the entire leg was very much swollen. Ultra-violet light was

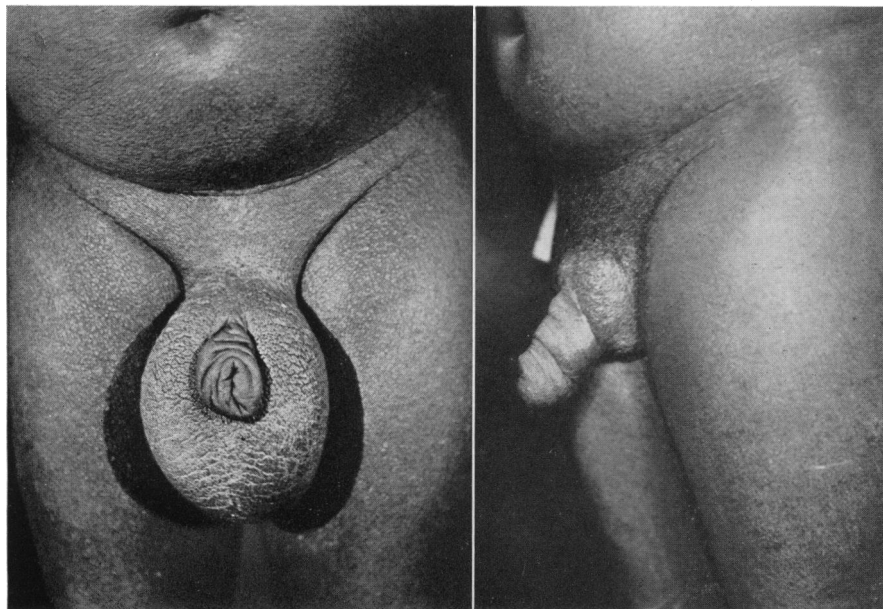


FIG. 5.—(Case III.) Elephantiasis of scrotum before operation. FIG. 6.—Elephantiasis of scrotum nine months after operation.

applied and arcase dressings. The temperature dropped the next day and the leg became progressively smaller, the redness disappeared, but the inguinal lymph-nodes remained large and tender, and he developed subsequently an abscess in his inguinal region which was incised and drained. A non-hemolytic *Staphylococcus aureus* was recovered and a vaccine was prepared. The patient was discharged again and vaccine treatment was started. His right leg has been free of attacks of swelling since then. He was seen at the follow-up clinic March 10, 1932, and he reported another acute attack of swelling and redness in his left leg, which, however, subsided in twenty-four hours. Both ankles at this time were still somewhat swollen but his legs had never reached the previous elephantoid proportions. He is still wearing an ace bandage, and he is still receiving vaccine treatment. In his case the erysipeloid attacks might prevent a permanently good result.

CASE III.—Man, colored, aged fifty-one years. First admitted May 20, 1931, to Doctor Muller's service, University Hospital. For five years the patient has been treated for lichenplanus in his inguinal and perineal region. During this time there was

a gradual swelling of his scrotum. He also had a strongly positive Wassermann. He was diagnosed as elephantiasis nostra by the dermatology department. Physical examination revealed a scrotum the size of a man's head (Fig. 5) with the penis entirely buried in the scrotum. Operation May 21, 1931, lateral elliptical incisions, removal of almost the entire scrotum. The testicles were replaced in a small pocket of skin. Convalescence normal. Culture showed a none-hemolytic staphylococcus and diphtheroids. Patient was seen in follow-up April, 1932, and his scrotum was small, while the penis was of normal proportions. (Fig. 6.) He had not had any attacks of painful swelling and he was again able to have intercourse, which had been absolutely impossible before operation.

CASE IV.—Male, white, aged forty-eight years. First admitted November, 1930, to Doctor Muller's service, University Hospital. He was sent in as an inguinal hernia. Four years before admission he was operated upon for bilateral inguinal hernia. Three months later the patient noted the swelling in the left side of the scrotum reappearing. Physical examination showed poor teeth and tonsils; there was considerable suprapubic swelling and the scrotum was as large as a coconut. The skin over the scrotum is reddened, thick, scaly and boggy; the left side of the scrotum was larger than the right side. The penis is very swollen. Because of recent flare-up of redness and swelling, the operation was postponed. The diagnosis was elephantiasis nostra, probably following his bilateral hernia operation. The patient gave a definite history of fever and vomiting associated with the scrotal swelling. February 19, 1931, operation was performed, consisting of excision of scrotum, bilateral bottle operation for hydrocele and a plastic on the penis. The hydrocele operation was not performed for hydrocele but to prevent the later formation of hydrocele. Lateral incisions were made from the antero-lateral portion of the scrotum down to the postero-lateral portion, the direction of the incision being governed by the extent of diseased tissues. Lateral flaps of normal skin were then secured and the entire scrotum was excised; the testicles themselves were freed and completely isolated; the tunica vaginalis of each testis was opened and inverted to prevent formation of a traumatic hydrocele. A diamond-shaped piece of skin and subcutaneous tissue was then removed from the posterior part of the penis. The lateral flaps of normal skin were then shaped into the new scrotum. The recovery from operation was uneventful. A culture from the scrotum showed hemolytic staphylococcus albus of auranticus type and a none-hemolytic *Streptococcus ignavus*. These are both rare organisms, and are of great interest. The pathological section showed marked oedema of the scrotum and penile tissues with localized areas of round-cell infiltration. The patient was seen in follow-up clinic March 10, 1932. His scrotum is much smaller. He still has occasional attacks of fever, chills, swelling and redness of the scrotum, but the swelling always returns to its normal size very soon and the attacks are much shorter.

CASE V.—Male, white, aged forty-seven years. Admitted November 3, 1931, to Doctor Muller's service, University Hospital, with a fracture of the right femur, lacerations of the forehead, right knee and left ankle, after being struck by a truck. He also had a possible skull fracture and an intra-abdominal or retroperitoneal hæmorrhage. He was put into the Russel traction apparatus and after a fair reduction of his fracture no further attempts were made on account of his serious condition. During the first two weeks he was very sick and several times it seemed necessary to open his abdomen. He was in apparent good health again one month after the accident, but at this time X-ray revealed non-union of his fracture. Open reduction with plating was undertaken December 8, 1931, and a body plaster case was applied. Immediately after the open reduction there was only a slight flare-up of temperature and some abdominal distension. On December 18, 1931, he developed a fever of 104°, his left leg, the right leg was fractured, became red and swollen, he was very sick with chills and vomiting. The entire case was removed and the patient was placed in the Russell's apparatus again. There was a temporary improvement following the removal of his case and then his temperature began to mount again and his leucocytes rose to 26,000. On December 27, 1931, there began

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oedema and redness of right leg at the ankle and slowly involving the entire leg up to the iliac crest. One area over the right thigh appeared almost fluctuant and was incised but only clear white lymph was obtained. X-rays at this time showed possible osteomyelitis; this, however, was proved to be wrong in subsequent films. There were several areas of redness, erysipelas-like, with very brawny induration developing around both his ankles, over his right thigh and just below the left iliac crest. During early January, or two months after the accident, there was also the development of scrotal oedema. The entire picture appeared at this time as of an acute attack of elephantiasis nostra of both legs and scrotum. Both legs and the scrotum became tremendously enlarged. On January 10, 1932, active treatment for acute elephantiasis nostra was started. He received repeated small blood transfusions and repeated injections of twenty-five cubic centimetres of polyvalent anti-streptococcus serum. Ultra-violet therapy was also started and viosterol by mouth. After six injections of anti-streptococcus serum, the oedema of his legs began to subside gradually. During the last two months he has improved steadily but slowly. His fracture seems well united and he is out of bed; of course he is not yet able to walk. There remains only one area of brawny induration over the lower right femur. He has not had any more attacks of fever and swelling. The sinus over the right thigh has stopped draining large quantities of lymph. Cultures taken from tissue did not show growth, but cultures taken from the sinus in his leg showed a hemolytic *Staphylococcus albus* and *Bacillus pyocyaneus*. Of course, the great question in this case, as it is in all other cases of elephantiasis, is, will the patient continue to have erysipeloid attacks; and if the attacks will recur he surely will develop true elephantiasis.

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