

TRANSACTIONS
OF
The Society of Tropical Medicine and Hygiene.

A CASE OF ELEPHANTIASIS, TREATED BY
LYMPHANGIOPLASTY.

SHOWN BY W. SAMPSON HANDLEY, M.S.LOND., F.R.C.S.

Friday, November 20, 1908.

J. B., a clerk, aged 46, was sent to me in April, 1908, by Dr. J. J. Udale, and I cannot do better than quote in abstract his account of the case :—

“Your communication to the *Lancet* re the restoration of lymphatic circulation in an cedematous limb has induced me to write to you respecting a patient who is suffering from sporadic elephantiasis of the left leg. He is an Englishman and has never been abroad. I have seen the case almost from the first onset in 1895. He first complained of swelling of the testicle and of pain in the leg. The limb was at that time only moderately swollen, and presented patches of erythema. The left testicle was much enlarged, hard and nodular, and the case wore the aspect of a sarcomatous growth of the testicle, with secondary glands causing the œdema. Since that time he has been to various hospitals, St. Bartholomew's, West London, Hampstead and Poplar.”

In 1905 he was under the care of Sir Patrick Manson at the Albert Dock Hospital. The leg was incised, and portions of tissue removed, with some benefit. At the same time the swollen testicle was also apparently

incised. Subsequent removal of the testicle was necessary, owing to an attack of acute sepsis following the incisions. His subsequent history confirms the view that the sepsis was due, not to the introduction of bacteria, but to their presence in his tissues at the time of operation.

He still has recurrent pyrexial attacks, ushered in by a rigor and accompanied by headache. The leg during the attacks shows patchy erythema, and is the seat of pain, increasing in severity at night. While in bed the leg becomes comparatively flabby, being less tense than when he is getting about.

The patient states that the swelling began in his foot, and that he had to cut his boot. For three years after 1895 the swelling of the leg remained comparatively slight. He can walk about with difficulty.

Condition on Admission.—The left testicle is absent. The left lower limb is enormously enlarged, especially below the knee. On the posterior aspect of the calf are two huge masses separated by a deep cleft or crease. Another large mass occupies the dorsum of the foot. The patella cannot be felt. The thigh is much enlarged, but presents no pendulous folds. The skin is much thickened, coarse and rugose. Mr. R. Paget made nocturnal examinations of the blood for filaria without result.

On April 18, 1908, I performed lymphangioplasty,¹ the first occasion, so far as I know, that my operation has been applied to elephantiasis. Eight stout silk threads were led upwards from the foot to a point in the iliac

¹“Lymphangioplasty: A New Method for the Relief of the Brawny Arm of Breast Cancer, and for Similar Conditions of Lymphatic (Edema,” by W. Sampson Handley. The *Lancet*, March 14, 1908. See also *Archives of the Middlesex Hospital*, 1908, vol. xii., p. 28.

fossa just above Poupart's ligament. These were spaced out round the limb at intervals. Above they united to form a leash. The iliacus muscle was exposed by a short incision like that for ligature of the external iliac artery. Half the threads were passed through a portion of the iliacus muscle, and were knotted in pairs to the other four threads, thus fixing them securely to the tissues of the iliac fossa. The multiple small incisions through which the threads were introduced were closed by horse-hair sutures, leaving the silk completely buried in the subcutaneous tissues. The leg was raised on pillows.

TABLE OF MEASUREMENTS.

Date	CIRCUMFERENCE IN INCHES						
	Foot	2 in. above ankle	6 in. above ankle	Calf (maximum diameter)	Knee	11 in. above knee	Groin
April 17 (before operation)	15	21	21	24½	19	23	23
April 23 (five days after operation)	12	14½	17½	20	18	20½	23
April 27	11½	14	15	17	17½	19½	21
May 2	12	15	16	17	17	21	21
June 14	13½	17½ (silk removed on May 2)	17½	18½	16½	19½	20
Normal right limb..	9	7½	9	10	12½	14	15½

At first all went well, and the success of the operation seemed almost assured. The drainage of lymph from the limb was so free that a large cushion of œdema appeared in the loin on the day following operation. This swelling extended upwards nearly to the axilla, and pitted freely on pressure. Concurrently the swelling of the leg subsided rapidly, the bandages became loose, and the pendulous folds of subcutaneous tissue shrank and took on the consistence of a sucked orange. The preceding table of measurements gives an idea of the effect of

the operation. Within five days, it will be noted, the ankle diminished in circumference from 21 inches to $14\frac{1}{2}$ inches, and in nine days the calf shrank from $24\frac{1}{2}$ inches to 17 inches. By April 24 the lumbar swelling had commenced to subside and was less tense.

About April 27 trouble began. The temperature, which even before the operation had not been quite steady, reaching over 100°F . on one occasion, had not subsequently exceeded 99.4° until the stitches were removed on the ninth day. All the wounds had healed except one on the outer side of the calf, from which pus could be expressed, and the one above the groin, from which clear serum exuded. On the 28th another wound on the inner side reopened and discharged pus; on the 29th another on the outer side. On the 30th there was redness along the track of the silk on the outer side of the leg. The constitutional disturbance was extraordinarily slight, and the patient only felt slight malaise, but the temperature gradually rose to 101.8° on May 2, and other wounds reopened. Retreat, if possible, was clearly imperative, and I removed all the silk, under an anæsthetic, on May 2 without much trouble, drawing it out from above after the strands had been cut across subcutaneously in the region of the foot. The tracks were syringed through with zinc chloride, 20 grains to the ounce.

At this time the case caused me not only keen disappointment but great anxiety, for pyæmia seemed a likely sequel. The suppurating tracks left by the removal of the silk were regularly syringed with antiseptics, and hot fomentations were applied. Three days after the removal of the silk, the swelling of the leg had begun to recur, although its posture remained unchanged, a fact which demonstrates that the capillary

action of the silk is the essential factor in lymphangioplasty. On May 3 the temperature had fallen to 99·2°. Subsequently the discharge of pus rapidly diminished and many of the silk-tracks appeared to close at once. On May 21 a large abscess which had formed in the thigh was evacuated. It was reported to contain *Staphylococcus aureus* in pure culture. On May 26 secondary hæmorrhage took place into the abscess cavity. The house surgeon, Mr. P. J. Chissell, very skilfully tied the external circumflex artery and stopped the bleeding. To his care and skill at this trying period the patient and I owe much. The incisions ceased to suppurate soon afterwards, and on June 11 only one incision in the leg continued to discharge. On June 19 the patient was sent to a convalescent home with the wounds entirely healed.

Up to this point the case had been a disastrous failure, but it seemed to me that hope need not be abandoned, a view which the patient fortunately shared. It was remarkable that suppuration had not set in until the ninth day after operation. If it had been due to infection of the silk from the skin during the operation, it appeared to me that it would have begun earlier. I felt very strongly that the suppuration was of endogenous origin, that it indicated a prior chronic infection of the tissues of the leg, or of the blood, by some bacterium. On June 12 I accordingly asked Dr. A. G. R. Foulerton to cultivate the blood. The result on this occasion was negative.

On August 1 the patient returned from Clacton in good health, and was re-admitted for further bacterial investigation and treatment. On August 4 a specimen of blood was drawn from the left arm, and streptococci were reported to be present in it, a startling finding,

since the pulse and temperature were normal and the patient was apparently in excellent health. Three days later further specimens of blood were taken from the left arm and from the left leg.

On August 12, with a Southey's tube, a specimen of lymph was withdrawn from the left leg and submitted to Dr. Foulerton. His report was as follows :—

"A number of diplococci were found in films made from the lymph withdrawn from the leg, but the cultures obtained were heavily contaminated, and it was not possible to isolate the organism."

A similar diplococcus was seen in lymph drawn on August 18, and was obtained in pure culture.

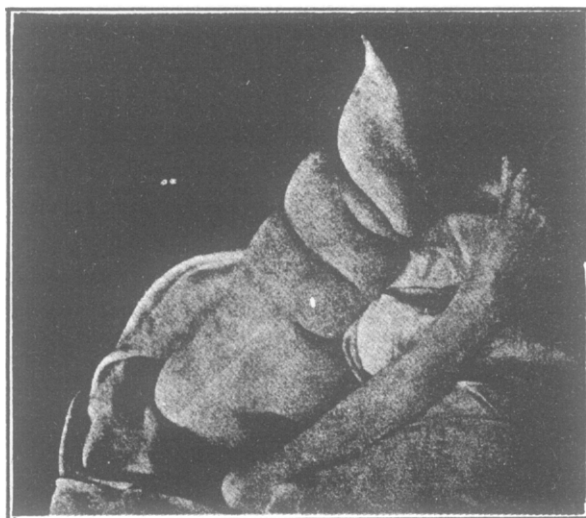
The organism thus detected by Dr. Foulerton presented a remarkable similarity to that recently described by Dufougère¹ under the name of the lymphococcus, as one of the factors in the causation of elephantiasis, but I shall not enter into the bacterial side of the case, which will be described in a separate paper by Dr. Foulerton and his assistants, Mr. W. P. Hillier and Miss H. K. Whittingham.

Obviously the first thing to be done was to exterminate the infective cocci as a preliminary to further operative treatment. Dr. Foulerton prepared for me a vaccine, of which the first dose was given on August 26 (400 million cocci). Subsequent doses were given on August 31, September 8, 15, and 23, October 1 and 12. On September 11 lymph from the leg was examined, and cocci were still found in it, though they were absent from the systemic blood. On September 5 both the blood and the lymph were found sterile. The vaccine treatment had not affected the swelling, which continued slowly to increase. On September 12 a further negative examination of the lymph confirmed the result of the

¹ *L'Éléphantiasis*, Paris, 1907.



Eighteen days after lymphangioplasty, before treatment by massage and bandage-pressure.



Before operation, but after vaccine treatment and rest in bed.

previous examination. There was no diminution in the swelling of the leg.

The sterility of the tissues being thus secured, lymph-angioplasty was again performed on October 17 after most careful and thorough preparation of the skin. On this occasion the silk threads, so as to avoid the scars of the previous operation, were carried up to the gluteal region, where they terminated in the subcutaneous fat. All again went well until October 26, when the temperature rose to 99.4° . It was on this very day, the ninth after operation, that trouble had begun on the first occasion, so that the situation was alarming, although no local irritation could be seen about any of the incisions. A dose of vaccine (200 million cocci) was immediately injected, and the supports, which had raised the foot of the bed 18 inches to promote drainage, were transferred to the head of the bed. The effect of this was of course to produce a passive oedema of the left leg, the tissues of which rapidly began to refill. The limb was thus practically treated by Bier's method. Following the application of these measures, although the temperature rose to 100° on the day following, it then rapidly subsided, without the development of any local trouble. The stitches were removed on October 27 and all the incisions were found to be healed except one on the foot, where the skin was extremely thick and indolent. Any possible risk from this was guarded against by the use for a few days of weak carbolic baths for the foot. On November 1 the dressings were left off and the limb re-elevated. The remarkable effect of the operation in reducing the swelling of the limb is shown by the measurements on the next page.

Unfortunately some of these measurements are not taken at the same precise levels as those of the previous

table. It will be seen (a) that the limb had slowly filled again after the removal of the silk, until on October 14 its diameters were greater than before the first operation; (b) that within thirteen days the second operation had reduced the diameters of the leg below the minimum after the unsuccessful first operation; (c) on the other hand; the second operation was not so effective as the first in reducing the diameter of the thigh.

	October 14	October 27	November 29
Circumference at mid-tarsal joint ..	14 $\frac{1}{2}$	11 $\frac{1}{2}$	10 $\frac{1}{8}$
„ above ankle	21 $\frac{3}{4}$	19 $\frac{1}{4}$	11 $\frac{1}{2}$
„ of leg below knee.. ..	25 $\frac{3}{8}$	17	14 $\frac{3}{8}$
„ at knee-joint	20	16 $\frac{3}{4}$	15 $\frac{1}{8}$
„ of thigh 9 in. above knee	24 $\frac{3}{4}$	21 $\frac{1}{4}$	19 $\frac{1}{4}$

(Second operation of lymphangioplasty on October 17, 1908.)

The last point is thus accounted for. At the first operation the silks were evenly distributed round the thigh. At the second operation attention was concentrated on the leg below the knee. Above the knee all the threads were led upwards in a single leash at the back of the thigh, leaving that part of the thigh to be dealt with subsequently if necessary. The argument, that the results are due to posture, rather than to the capillary action of the silk, is thus effectively met.

Sir HAVELOCK CHARLES thought the case exhibited was an exceedingly interesting one, but he did not look upon it as a cure. The limb had not yet returned to its ordinary size, and there was very great thickening of the tissues. The patient was much better than he was originally, but he did not consider that in India it would be looked upon as a cure. The method Mr. Handley had described was an excellent one, which was particularly

applicable in cases of mammary cancer and thickening of the arm. He thought that the absence of filaria was an interesting point, because, as they all knew, in cases of elephantiasis of the scrotum it was a very rare thing indeed to get filaria, but the present case was an instance where micrococci were present in the blood. He suggested that the cause of elephantiasis of the scrotum was a peripheral lymphangitis due to certain organisms. In the particular case under discussion, there were micrococci in the blood, and so long as they were present the patient did not get better, but when the vaccine was applied and the micrococci were destroyed an improvement followed. Was it a feasible explanation to suggest that the improvement in the limb was at present due to the destruction of the micrococci? In the treatment of the limb he personally should prefer a more radical method which would give a thorough result, *i.e.*, a circular incision above the knee, above the level of the diseased tissues; a vertical incision down the median line in front to the termination of the interdigital folds; a similar one on the back of the limb right down to the heel; one passing around the foot at the upper margin of the sole, leaving absolutely intact the sole of the foot and the pulps of the toes, removing the whole of the tissues right down to the deep fascia, and grafting with large grafts. The operation was quite feasible; all that was necessary was absolute asepsis. He suggested that operation to Mr. Handley in his next case. Nevertheless he thought Mr. Handley was to be highly congratulated on the result, but what struck him most was the destruction of the micrococci.

The PRESIDENT thought that, whatever the result might be in regard to the treatment of elephantiasis, Mr. Handley was to be congratulated on introducing a

new idea into surgery, and he hoped, whether it succeeded in elephantiasis or not, the mechanical idea of drainage between two different areas of connective tissue might have its application in other departments of surgery. Tropical medicine had supplied general surgery with many new ideas; it was refreshing to find that general surgery was now repaying tropical medicine.

Mr. HANDLEY, in reply, after thanking the President and Sir HAVELOCK CHARLES for their kind remarks, desired to point out that it was only a month since the case was operated upon, and doubtless there was much thickening of the tissues still left. He therefore thought it was not fair to say it was not a cure, because one did not know how far the process of involution in the tissues of the limb might go.

Sir HAVELOCK CHARLES said he understood the case was presented to the Society as a cure.

Mr. HANDLEY replied that the case was not presented as a cure accomplished, but as a cure in process. That was his view of the case; and he hoped he was not wrong.

Sir HAVELOCK CHARLES said he desired to withdraw any remarks he had made with reference to that particular point, but when the patient began to walk there would perhaps be an increase of the present swelling.

DESCRIPTION OF BILHARZIASIS SPECIMENS.

Dr. F. M. SANDWITH described a number of specimens which had been sent to him from Egypt. The first was a very good sample of kidneys, ureters and bladder, presented to the Museum of the London School of Tropical Medicine by Dr. Kartulis, of Alexandria, a Fellow of the

Society. The Fellows knew perfectly well that the kidney was not primarily affected by the eggs of the bilharzia worm. The ureters and the bladder, on the contrary, showed marked lesions. The kidney was often affected in a secondary way, because when there was any obstruction in the bladder or in the ureters, what used to be called surgical nephritis followed, hydronephrosis, pyonephrosis, and sometimes at a *post-mortem* examination the whole kidney was found converted into a thin sac of pus. He thought the Fellows would agree that these ureters were as perfect specimens as could be hoped for. The reason they were so good was because the whole of the ureter was affected. There was hardly a part of the ureter, except right up in the kidney, which was not full of little new growths of bilharzial disease, every one of which was full of eggs. The bladder itself was also a good specimen because of its thickness and the very large number of papillomata that were to be seen. It would be remembered that disease of the bladder usually began in the trigone. The kidney itself was not so interesting, although there was some dilatation of the pelvis.

The second bladder was exhibited at the Exeter meeting of the British Medical Association by Dr. A. R. Ferguson, of Cairo. He (Dr. Sandwith) brought it on purpose to contrast it with the other bladder, because it presented the most advanced overgrowth of the bilharzial disease in that organ that it was possible to have. It would be observed that there was hardly any place left for the urine to collect in. The whole viscus was distorted and put out of shape. There were some small papillomata such as were to be seen in the ureter, and some huge lumps. Not only was the bladder extremely thickened, but there was an enormous overgrowth, which, both clinically and histologically, was locally malignant.

The exact relationship between it and cancer had not been worked out, but he hoped Dr. Ferguson would do it some day.

He next exhibited a liver showing well-marked bilharzial cirrhosis, a perfectly distinct disease. He had seen such a thing half a dozen times, and at the Tropical School there were specimens of two different livers, both presented by the Egyptian Government School of Medicine. It was a real cirrhosis, though there was no prominence of the liver lobules nor retraction of the capsule as seen in other forms of liver cirrhosis. The section of liver presented an increase of new white fibrous tissue, so that its cut surface looked as if some white clay-pipe stems had been thrust into it, and it was full of eggs. The newly-formed connective tissue seemed to be confined to the neighbourhood of the portal veins, and a large branch of the hepatic vein was unaffected.

Dr. Kartulis had also sent a good specimen of the rectum showing large papillomata. It was easy to understand from its appearance how it was quite possible for the uninitiated to confuse the condition with hæmorrhoids and even with dysentery, because it produced much the same symptoms, *i.e.*, blood, mucus, tenesmus and many motions. The specimen was a typical case, although not nearly so advanced as the cases shown in the pictures exhibited, where there was a good instance of papillomata of the descending colon.

Under one microscope there was a slide showing a mass of eggs in the bladder. Those who had not worked at the subject would be interested in seeing how they were grouped together; there were literally hundreds in the field

Under the next microscope, with a higher power, there were some similar eggs from a ureter showing the miracidium within the shell.

Under the third microscope there were some bilharzia eggs from the lung. There were very few of them, there being only one or two in a field, and perhaps only twelve in the whole slide. The only interest in showing the slide was because of the rarity of bilharzial disease in the lung. Evidently the female that laid the eggs had lost her way and had not gone to the bladder and the rectum as most of the females did. Dr. T. S. Kerr had also very kindly sent a specimen of lateral-spined ovum from Porto Rico, which was to be seen under the fourth microscope, while under the fifth were eggs on a warm stage, from which living miracidia could be seen to emerge. These eggs were obtained from urine from Haslar, kindly supplied by Fleet-Surgeon Bassett-Smith.

There was also on exhibition a series of photographs of bilharzial disease, including some illustrations of how the disease caused prolapse of the rectum, and there were also some instances of huge cauliflower excrescences of the vulva in cases where the urethra and the bladder were apparently normal in the female. Then, lastly, there was shown a model of a vesical calculus from a case described by Mr. Owen Richards in the *Lancet* of July 25, 1908, now on its way to the Museum of the London School of Tropical Medicine. By its side was a model of the largest stone ever successfully removed, a feat accomplished by Mr. Milton in 1893, the stone weighing $34\frac{1}{2}$ oz. Mr. Richards's case was the second largest, weighing 31.4 oz. Mr. Milton's stone was removed by abdominal section, because it was impossible to remove it by lithotrity or by suprapubic operation; while Mr. Richards's stone was extracted piece-meal by osteotome and mallet above the pubes, aided by perineal section. He tried to perform lithotrity, but failed to do so because Milton's 5-inch lithotrite was not large enough

for it, and the stone was therefore broken up and extracted in pieces. Subsequently the broken pieces were put together in order that a cast might be made. He thought the Fellows would all agree that the originals of the two stones were in their proper place, namely, in the best pathological museum in the world, in Lincoln's Inn Fields.

DISCUSSION.

Dr. HARFORD enquired whether Dr. Sandwith could state in what proportion of cases evidence of bilharziasis disappeared. He was very much interested in the subject, because he had seen that day the wife of one of the missionaries of the Church Missionary Society who came home from East Africa with the repute of having had bilharzia in the bladder. She had passed urine and also a calculus which contained bilharzial eggs, and afterwards she had had mucous colitis. When she came home no ova could be discovered at all, so that it could not be said that the mucous colitis was associated with the bilharzial disease. Since she came home, about eighteen months ago, she had not had any sign of diarrhoea or any other bad symptom; and he therefore would be very much obliged if Dr. Sandwith would state his general view of the case, and whether the trouble was likely to appear again.

The PRESIDENT asked Dr. Sandwith whether so-called bilharzial cancer of the bladder was ever associated with secondary growths; and also whether the ova in the liver were terminal-spined or lateral-spined. The Fellows might remember that some time ago a discussion occurred at the Society on the subject of the specific differences between the lateral-spined ovum and the terminal-spined ovum, and some of the Fellows then maintained that the two were distinct species. Quite recently a paper had

been received at the School of Tropical Medicine from a Brazilian observer, who described in detail some cases of rectal bilharzia, in all of which the ova were lateral-spined; and although he had investigated the urine of hundreds, if not thousands of Brazilians, including the cases of rectal bilharzia, in no instance did he find bilharzia in the urine, and in no instance had he ever found a terminal-spined ovum, bearing out the contention that the parent of the lateral-spined ovum and the parent of the terminal-spined ovum probably belonged to different species. He would be glad to know if Dr. Sandwith had any recent information bearing on that subject.

Dr. SANDWITH, in reply, said he was very much obliged to Dr. Harford for putting his question in the way he did. The general way of asking such a question was "How soon will the patient recover?" But Dr. Harford had been bold enough to say that the patient had recovered, and asked what guarantee was there that the patient would not have a relapse? He imagined from what Dr. Harford said to him before the meeting that he referred to the case of people who had lived in a country where bilharzial disease was endemic, and came home to a country like England, where there was no bilharzial disease, and therefore no fear of fresh reinfection. In those cases he maintained that they all got well if they lived a sufficiently long time in a country where there was no bilharzial disease. That meant in point of fact that the parent worm was not eternal, that it would not live for ever, although apparently it could live for a great number of years. He began by thinking, judging from the cases that he had seen, that the patients lost the eggs of the bilharzial disease within two years. He confessed, however, that he had now had to extend the time to four

years, and cases had been published where people, even after ten or fourteen years, had been known to pass bilharzial eggs, especially after exertion such as riding. He advised Dr. Harford's patient not to have her urine examined under the microscope immediately after some prolonged exertion, such as bicycle riding, horse riding, a long walk, or something of that kind. He knew from experience that several young Englishmen contracted the disease in Egypt and lost it within a couple of years. He hoped Dr. Harford's patient had lost the disease, and it would remain absent unless she subjected herself to some great fatigue. The President had enquired whether the condition which was seen in the bladder given by Dr. Ferguson was cancer followed by secondary manifestations. Speaking generally, that was not so, the case had no secondary manifestations. That was one of the interesting points, because one would expect so much irritation in the bladder to give rise to the occurrence of cancer. When cancer did exist in a bilharzial patient (and one native out of three in Lower Egypt had bilharzia), there was no very great probability that the cancer had been produced by the bilharzial irritation. The President asked a question about the liver; and he (Dr. Sandwith) asked to be allowed to read a note by Professor Symmers of the microscopical examination of such a liver: "There is a great increase of the periportal connective tissue of the sublobular and larger portal canals, the fibres of which are mostly wavy and parallel, but here and there arranged concentrically, surrounding ova. Eggs found in the liver consist usually only of the shell with lateral spine." That was the answer to the President's question, that they had a lateral spine, *i.e.*, that they belonged to the rectal form rather than to the bladder form. They were generally shells; he could not

show a perfect specimen under the microscope because the shells were destroyed in cutting sections. No eggs are seen lying free among the hepatic cells, nor usually in the larger blood-vessels which permeate the new cirrhotic tissue. The whitish nodules on the surface of the liver consist of fibrous tissue containing eggs. The President had also told them of the Brazilian cases, which confirmed previous knowledge, that in the rectum the lateral-spined egg was obtained and not the terminal-spined egg. Many hundreds of cases had been examined by Bilharz and Griesinger and ever since their day and that seemed to be an invariably true rule. Wishing to avoid a debatable point, he had desired to confine his remarks that evening to the terminal-spined eggs until Dr. Kerr offered to bring forward a specimen of lateral-spined ovum. The whole question had been extremely well summed up by Professor Looss in the July number of the *Annals of the Liverpool School of Tropical Medicine*, to which he (Dr. Sandwith) was unable at present to add anything.