

of the patients the reaction was mild, in the second instance the reaction was more severe, with urticaria, fall in blood pressure and some respiratory distress. The examination had to be postponed and was later carried out successfully without the use of dye.

### Summary

Studies were made in 205 consecutive lymphographies, 101 without and 104 with the aid of dyes. The failure rate and operation time were the same in both groups. Two patients had allergic reaction to the dye.

### References

- 1 *Kinmonth, J.B.*: Lymphography in Man: A Method of outlining Lymphatic Trunks at Surgery. *Clin. Sci.* 11 (1952) 13-20
- 2 *Kinmonth, J.B., G.W. Tylor, R.K. Harper*: Lymphography; a Technique for its Clinical Use in the Lower Limb. *Brit. Med. J.* 1 (1955) 940-942
- 3 *Mortazavi, S.H., B.D. Burrows*: Allergic Reactions to Patent Blue Dye in Lymphangiography. *Clin. Radiol.* 22 (1971) 389-390
- 4 *Elke M., I. Hug*: Erprobte Modifikation der Lymphographischen Untersuchungstechnik. *Röntgenpraxis* 24 (1971) 37-42
- 5 *Kenyon, N.M., M. Soto, M. Viamonte Jr., R. E. Parks, J.J. Farrell*: Improved Techniques and Results of Lymphography. *Surg. Gynecol. Obstetr.* 114 (1962) 677-682
- 6 *Schaffer, B., P.R. Koehler, C.R. Daniel, G.T. Wohl, E. Rivera, W.A. Meyers, J.F. Skelley*: A critical Evaluation of Lymphangiography. *Radiology* 80 (1963) 917-930
- 7 *Koehler, P.R.*: Complications of Lymphography. *Lymphology* 1 (1968) 117-120

*Proofs should be sent to: Kristjan Sigurjonsson, M.B. Kjartangsgata 10, Reykjavik, Iceland (Island)*

Lymphology 3 (1974) 123-130  
© Georg Thieme Verlag, Stuttgart

## The Renogram in Lymphatic Dysplasia\*

J. Beninson, R.L. Rian, W.R. Eyler, L.A. DuSault

Henry Ford Hospital, Detroit, Michigan 48202, Department of Dermatology

### Summary

1. Forty-two patients with lymphatic dysplasia were examined by hippuran iodine 131 renograms. The patients all had clinical and lymphangiographic proof of lymphatic dysplasia. None of the patients were hypertensive.
2. Of the 84 kidneys studied, 17 handled the hippuran in normal fashion with only 7 patients showing bilaterally normal renograms.
3. Abnormal tracings were categorized as mild in 44 kidneys, moderate in 16 kidneys, and severe in 7 kidneys, many of which could have been confused with nephrosclerosis or renal artery stenosis.

\* Presented: March, 1973; Fourth International Congress of Lymphology, Tucson, Arizona

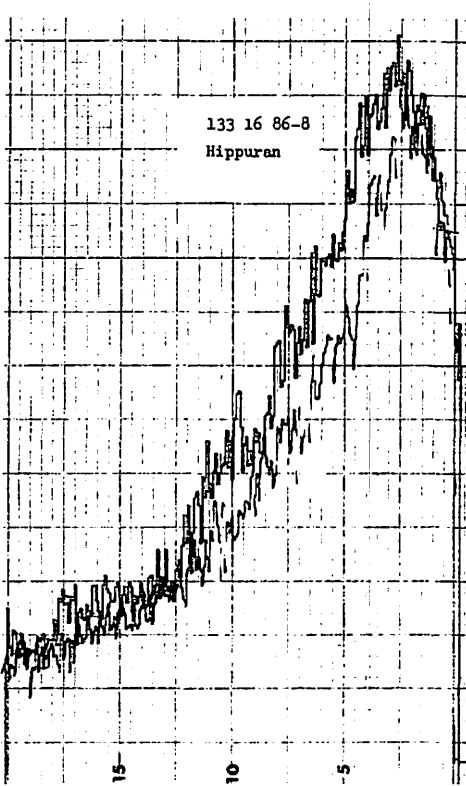


Fig. 1 J.H. a 15 year old black female (case No. 39) presented with lymphedema of the lower extremities. The blood pressure was 112/66. The renogram tracing reads from the reader's right to the reader's left. The lower tracing is the right side; it is considered to be within normal limits. Note the peak at 2 1/2 minutes and the downward convexity of the descending limb in the subsequent 5 minutes. The upper tracing of the left side shows a peak at a time between 2 1/2 minutes and 3 minutes. The descending limb, however, shows less downward convexity than the opposite side. Note that the upper tracing (left side) is straight from 4 to 9 minutes. This is the least degree of abnormality which can be detected in the renogram and is classified as mild abnormality.

the examination is performed between 8:00 a.m. and 11:00 a.m. the following morning. The patients have been examined during an interval when they are not on medication and usually before any pumping procedure has been done for lymphedema of the involved extremity. A three probe instrument employing 2" x 2" scintillation crystals with straight bore collimator is used. One probe is placed over the marker for each kidney, the third over the urinary bladder area or shoulder area. Ten to 15  $\mu$ c of hippuran I 131 are injected intravenously and recordings made for the following 20 to 30 minutes.

### *The Renogram in Lymphatic Dysplasia*

Study of patients with lymphatic dysplasia has given support to the concept that this condition is a widespread systemic disorder involving many organ systems. Little has been done to investigate functions other than extremities in these patients. The senior author suggested that the renovascular system be investigated and this report is based on 42 patients with lymphatic dysplasia who have been evaluated by the renogram according to the previously described techniques (3, 4, 5). In our institution the hippuran iodine 131 renogram is an accepted technique for the evaluation of function in patients with a variety of urinary tract and renovascular disorders.

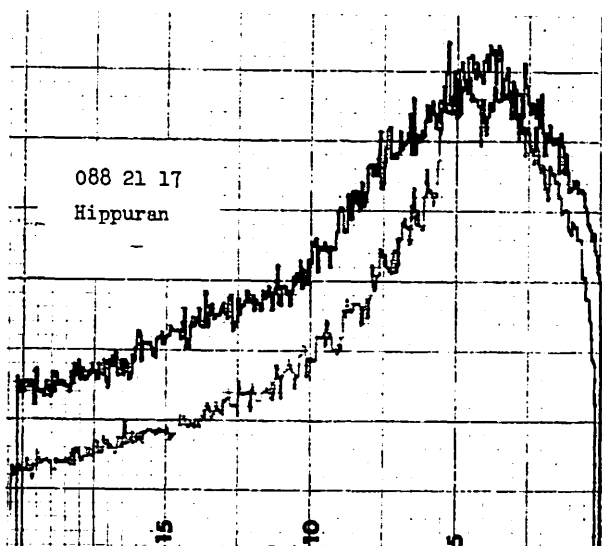
### *Materials and Methods*

The renal function of 42 patients with classical clinical lymphangiographic and laboratory manifestations of lymphedema (1, 2) was studied by hippuran I 131 renography using previously described techniques (1, 2). These studies done 8 to 12 years ago involved 37 females and 5 males ranging in ages from 7 to 59 years.

No patients show clinical or historical findings of dysuria, hematuria, or pyuria. Most patients had BUN determinations and all urinalyses were within normal limits. One or more days prior to the renogram a regular or abbreviated excretory urogram has been done. The examination has included at least one film centered directly over each kidney with the patient prone on a table tilted 20° from the horizontal with the patient's head higher than his feet.

The patient is given nothing by mouth from midnight prior to the renogram and

Fig. 2 R.M. a 16 year old white female (case No. 23) presented with lymphedema of the lower extremities. Blood pressure was 110/70. On each side the peaks are seen at 3 1/2 to 4 minutes. The tracing of the right side is the lower one. It shows a minimal straightening of the descending limb in the 5 minutes after the peak and is classed as mild abnormality. The upper tracing from the left side shows somewhat more loss of downward convexity and is classified as moderate abnormality. Note that the slopes of ascending and descending limbs of the tracing from this side are nearly symmetrical. The tracing of the right side shows a more rapid fall of the descending limb.



The following descriptive terms are used to categorize the renogram and are defined as follows:

*Normal* The renogram shows a peak at 2.5 to 4.5 minutes with a distinct downward convexity of the tracing in the 5 minutes following the peak.

*Mild* The renogram shows a peak at 4.0 to 5.5 minutes with a loss of some of the curve, but still convex downwards.

*Moderate* The renogram shows a peak at 4.5 to 7.0 minutes with an essentially straight line downward after the peak.

*Severe* The renogram shows a peak at 7 to 10 minutes with an upward convexity of the tracing which can be prolonged for 30 minutes or more.

### Results

Under these conditions a normal renogram shows a peak at 2 1/2 to 4 1/2 minutes with a distinct downward convexity of the tracing in the 5 minutes following the peak (Fig. 1). Of the 42 patients 7 showed normal renograms from both kidneys. Of the 35 remaining abnormal tracings, 22 were symmetrical while 13 were asymmetrical. Three patients showed abnormality only on one side while 24 showed bilateral symmetrical mild to moderate abnormalities. The remaining patients showed an even distribution of various combinations of asymmetrical severity of renal involvement. The age and severity of renogram abnormalities are shown in the table. Thirteen patients from this series showed abnormal renograms with one side worse than the other. Nine of the 14 patients had normal BUN values (4 had no BUN study recorded), and none of them were hypertensive; the tracings were indistinguishable from those seen in patients hypertensive from renal artery stenosis and renal ischemia (Figs. 2, 3, 4), or in some cases of urinary tract obstruction, (Fig. 5).

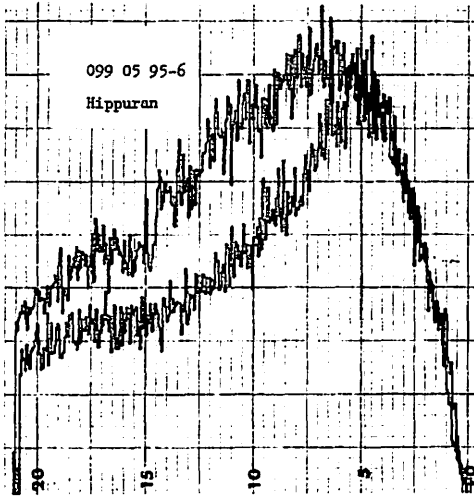


Fig. 3 M.D. a 55 year old white female (case 39) with lymphedema of the lower extremities had an episode of pseudotumor cerebri and was suspected of lymphatic abnormalities of the intestinal tract. The higher tracing is from the right side. It shows a peak at 7 minutes and a descending limb which is not only straightened but slightly convex upwards. This is classed as a severe abnormality. The lower tracing is from the left side. It shows a peak at 5 1/2 minutes with moderate straightening of the descending limb, and is classified as a mild abnormality

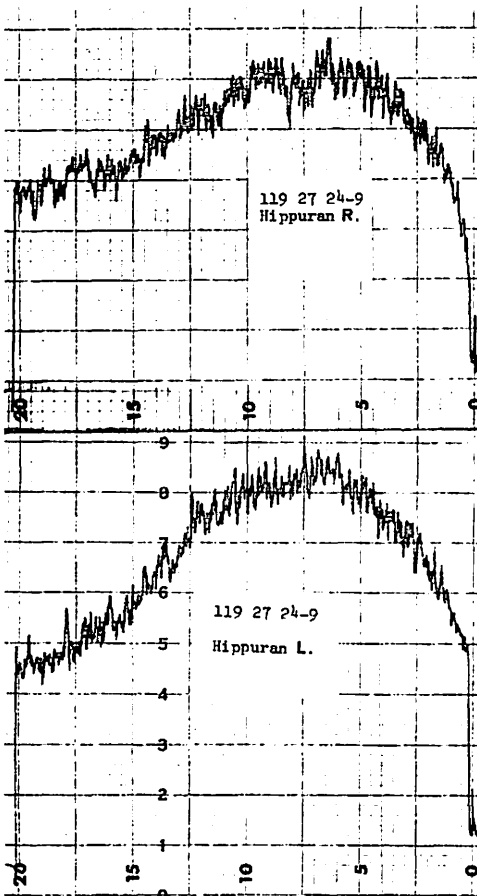


Fig. 4 P.P. is a 14 year old white female (case 36) who presented with lymphedema of the lower extremities. Blood pressure was 122/66. The tracing from the right side shows a peak at 7 minutes. There is upward convexity of the descending limb with very slow descent, classed as severe abnormality. On the left side the peak is found at 7 1/2 minutes and there is upward convexity of the descending limb but the curve falls more rapidly than the right side. Each side shows a severe abnormality and this is most marked on the right.

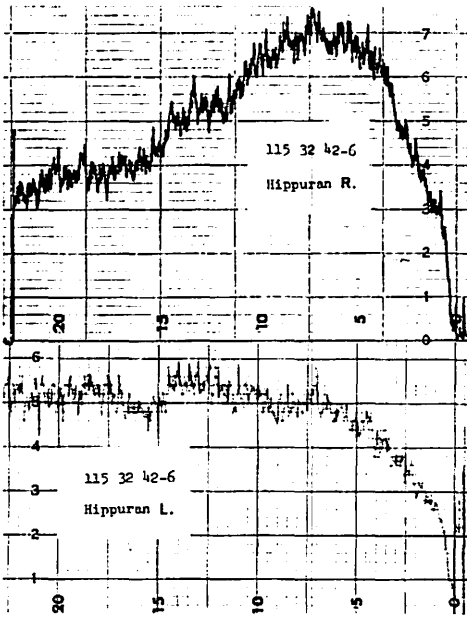


Fig. 5 M.S. a 37 year old white female (case 18) complained of lower extremity lymphedema. The upper tracing is of the right side; it shows a peak at 7 1/2 minutes with slight upward convexity of the descending limb and marked delay in the descent. The lower tracing is from the left side; a poorly defined peak is estimated at 10 minutes. There is prolonged curve over the following 20 minutes. These tracings are both classified as severe abnormalities with the left much more marked than the right.

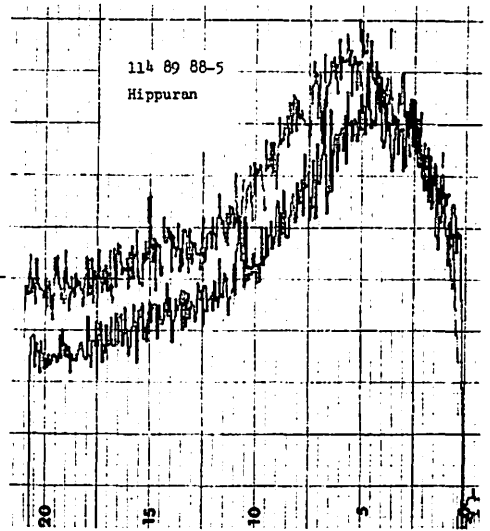


Fig. 6 A.C. a 13 year old black female (case 12) who complained of swelling of both lower extremities. The upper tracing is that of the right side. It shows a peak at 5 to 5 1/2 minutes. The descending limb shows moderate straightening. The slope of the ascending and descending limb is almost identical. The lower tracing is that of the left side.

It shows a peak at 4 1/2 minutes. On this side also slopes of ascending and descending limbs are very similar. Note that the two descending limbs are parallel. Although this tracing is that of a young woman with normal blood pressure and normal NPN, the renogram shows a tracing which otherwise would be suggestive of nephrosclerosis.

60 kidneys showed a straightening of the descending limb of the tracing with loss of the downward convexity in the 5 minutes following the peak.

This frequently results in the tracing showing a similar slope of the ascending and descending limbs to each of the peak with some delay peak time; 29 kidneys distributed among 18 patients showed that appearance.

In hypertensive patients with diffuse small vessel disease the tracings are similar to those just described and are bilaterally symmetrical.

Twenty-two patients showed symmetry between the right and left renograms; these tracings are indistinguishable from those found in patients hypertensive due to nephrosclerosis with involvement of the renal arterioles. Eleven of this 22 also showed similar slopes of the ascending limb before the peak and the descending limb after the peak (Fig. 6). Nine of the 42 patients had complete excretory urograms.

Table 1 Summary of 42 patients with correlation of their clinical and laboratory findings. Creatinine levels similar slopes of ascending and descending limbs of the renogram

Case No.	Sex	Age	Area of	Mg %		Renograms			
				BUN	und IVP	Sec. Peak	Impairment	Sec. Peak	Impairment
1	M	7	both feet	7	-	3	normal	3	normal
2	F	13	both lower	11	normal	3	normal	3	normal
3	F	18	both lower	11	-	4	normal	4	normal
4	M	21	Rt lower	-	-	4	normal	4	normal
5	F	22	Lt lower	12	Rt mild Hydro	3.5	normal	3.5	normal
6	F	32	both feet	9	-	3	normal	3	normal
7	F	41	both lower	11	-	4	normal	4	normal
8	M	13	both lower	19	normal	4	mild	3	normal
9	F	15	both lower	13	-	2.5	normal	3	mild
10	F	40	bil. lower	14	-	4	mild	4	normal
11	F	59	Rt Lower	-	-	6	moderate	5	mild
12	M	12	Both lower pudendal	-	-	3	mild	4	mild
13	F	12	both lower	-	-	5	mild	4.5	mild
14	F	13	both lower	-	-	5	mild	4.5	mild
15	M	14	both lower	11	-	4	mild	4	mild
16	F	14	bil. lower Rt lower	12	-	5	mild	5	mild
17	F	16	Rt arm - Rt face	10	-	5	mild	5	mild
18	F	16	both lower	-	-	6	mild	6	mild
19	F	18	both lower	-	-	4	mild	4	mild
20	F	19	Rt lower	10	-	5	mild	5	mild
21	F	24	both lower	21	-	4	mild	4	mild
22	F	26	Rt lower	15	-	5	mild	7	mild
23	F	33	both lower	12	-	3	mild	3	mild
24	F	33	Rt lower	9	-	6	mild	5	mild
25	F	34	left lower	11	-	5	mild	6	mild
26	F	37	both lower	12	-	5	mild	4	mild
27	F	52	both lower	13	-	4	mild	4	mild
28	F	53	Rt lower	-	-	4	mild	4	mild
29	F	57	both lower	1.0	Neg Symm Pelvis	6	mild	5	mild
30	F	16	left lower	-	-	4	mild	4	moderate
31	F	53	lower	14	-	7	moderate	5	mild
32	F	24	both legs	10	-	4	moderate	7	moderate
33	F	27	left lower	0.8	-	3	moderate	2	moderate
34	F	41			normal				
34	F	41	Rt lower	10	Rt pelvis bigger	4	moderate	3.5	moderate
35	F	41	both lower Rt lower	13	-	5	moderate	5	moderate
36	F	48	Rt face	16	normal symm pelvis	7	moderate	5	moderate
37	F	49	both lower	17	-	5	moderate	5	moderate
38	F	19	both lower	12	normal symm pelvis	8	severe	5	mild
39	F	55	Rt lower	-	small Rt kidney	7	severe	5.5	mild
40	F	35	Lt lower	10	Rt > Lt Hydro	9	severe	6.5	moderate
41	F	14	both lower	-	-	7	severe	7.5	severe
42	F	37	both lower	-	-	7.5	severe	10	severe

Of six excretory urograms done in the asymmetrical group, three were normal, one showed ureteral reflux on voiding, and two showed mild caliectasis with an appearance no different from that which would be seen in a patient who had pyelonephritis, of which there was no evidence in these patients. Indeed, the mild caliectasis could also represent a congenital variation. Two excretory urograms done in the symmetrical group were also normal. An additional one of this group showed mild asymmetrical caliectasis.

### Discussion

Involvement of upper extremities, right side of the face, abdominal and thoracic wall, intestinal lymphangiectasia with associated hypoalbuminemia and pseudotumor cerebri are manifestations which have been seen in our patients with lymphatic dysplasia of the lower extremities. Abnormal renograms in these patients constitute evidence of another major physiological abnormality which may be localized in the kidney or to which the kidneys may be responding. Consideration must be given briefly to the possibility of abnormal peripheral fluid collections influencing the renogram. Because of the sequence of procedures employed in the study of these patients, their fluid retention in extremities was minimal at the time of the renogram. This is due to the fact that the first examinations after admission were the previously reported (1, 2) radionuclide studies of the extremities which required 24 hours in bed. In addition, if extremity fluid collections were important to the renogram any delay in return of isotope to the kidney from the periphery should affect the two sides symmetrically, but the considerable number of asymmetrical renograms, thirteen, could not be explained by this factor.

The results of this study suggest, that renal physiological abnormalities should be more carefully evaluated in patients with lymphedema. Even though mild abnormalities appear frequent, they tend to be clinically silent in many instances.

### References

- 1 Beninson, J., H.S. Jacobson, W.R. Eyer, L.A. DuSault: Preliminary report on genetic lymphedema as studied by venography, lymphangiography, and radioisotopic tracers, <sup>22</sup>Na and RISA. *Progres in angiology, proceedings of the "Internationale Gespräche über Angiologie*, 1962. Darmstadt, Germany. Dr. D. Steinkopff Verlag, 1963 114-112
- 2 Beninson, J., H.S. Jacobson, W.R. Eyer, L.A. DuSault: Additional observations on Genetic lymphedema as studied by venography, lymphangiography, and radioisotopic tracers, <sup>22</sup>Na and RISA. *Vasc. Surg.* 1:1 (1967) 43-52

- 
1. Urines were normal in all 42 patients.
  2. Venograms – 17 patients had bilateral venograms.all were normal.
  3. Renograms – 7 were normal bilaterally, 29 were symmetrically abnormal, and 13 were asymmetrically abnormal.
  4. Lymphangiograms – No lymphatics were found bilaterally in 10 cases, 3 lymphatics were found unilaterally, in 13 they were hypoplastic bilaterally, in 5 they were hypoplastic unilaterally, and in 1 case they were tortuous.

- 3 Clark, M.D., W.R. Eyler, L.A. DuSault, E.J. Kochkodan, J.R. Caldwell: The renogram in hypertension. *Radiology* 89 (1967) 667-675
- 4 Tauxe, W.N.: In "Nuclear Medicine" ed. by Blahd, 2nd Edition, McGraw-Hill, St. Louis (1971) 382-415
- 5 Tauxe, W.N., J.C. Hunter, M.K. Burbank: The Radiosotope Renogram (Ortho-iodohip purate<sup>131</sup>). *Am. J. Clin. Path.* 37 (1962) 567

*J. Beninson, M.D., Henry Ford Hospital; Dept. of Dermatology, 2799 W. Grand Blvd. Detroit, Michigan 48202.*

Lymphology 3 (1974) 130-136  
© Georg Thieme Verlag, Stuttgart

### Lymphogenic Enteropathy from Thoracic Duct Obstruction

Z. Molnár,\* G. Karoliny,\*\* Gy. Mózsik\*\*\* and A. Németh\*\*\*\*

Department of Radiology\*, Department of Padiatry\*\*, First Department of Medicine\*\*\* and Department of Pathology\*\*\*\*, University Medical School, Pécs, Hungary

#### Summary

A twelve year-old girl was studied with protein-losing enteropathy, confirmed by <sup>51</sup>Cr-labeled albumin test. Pedal lymphangiography demonstrated an obstructed thoracic duct at the level of the cisterna chyli with mesenteric reflux and lymphatic collateralization through the liver.

A large proportion of patients with protein-losing enteropathy have insufficiency of the intestinal lymph circulation. Földi (1) calls this form of protein-losing enteropathy lymphogenic enteropathy, thus distinguishing it from exudative enteropathy where pathologic changes of the mucosa are primary and lymphatics have only a secondary role.

Enteric loss of protein is caused by a variety of abnormalities of the intestinal lymph circulation, but in some instances, the primary cause must be looked for outside the lymphatic system (e.g. increased central venous pressure or nephrotic syndrome). Intestinal lymphatic insufficiency may be congenital or acquired. Congenital disturbances are usually manifest at infancy or early childhood (2-4), and frequently the peripheral lymphatic system is also affected (5-7). In some patients, a strong hereditary trait is demonstrable (8, 9). Acquired disease often results from tumor invasion of lymphatics (10-12), but inflammation too may impair flow of intestinal lymph (13). In rare instances, fistulae may develop between the lymphatic system and the bowel lumen producing considerable protein loss (14-16), occasionally in conjunction with chylous ascites (17). In this report, a young girl developed obstruction to the lower part of the thoracic duct with protein-losing enteropathy.

An unusual compensatory network of lymphatic drainage developed through the liver and then towards the anterior mediastinum thereby circumventing the obstructed thoracic duct.