BILATERAL URETERAL OBSTRUCTION DUE TO ENVELOPMENT AND COMPRESSION BY AN INFLAMMATORY RETROPERITONEAL PROCESS

JOHN K. ORMOND

From the Henry Ford Hospital, Detroit, Michigan

The following 2 cases I deem worthy of presentation to you because not only are they unique in my experience but I have so far been unable to find any similar cases in the literature.

The first case is that of a Greek male whose age was 45 when first seen by us in June 1942. His complaints were substernal pains radiating down the inner surfaces of both arms; low backache of 4 months duration, radiating down both thighs but especially the left; and loss of 10 pounds in weight in the previous 5 months.

There was nothing of special interest in his family and past histories except that his father had died of Bright's disease and his mother was hypertensive.

General physical examination showed as follows: Weight 129½ pounds. Ocular reflexes normal. Tonsils small. Teeth in good repair. No general lymph gland enlargement. Lungs and heart clear to auscultation and percussion. Blood pressure 138/100. No hernia. No abdominal masses or tenderness. No costovertebral tenderness. No abnormalities of bones and joints. Deep reflexes normally obtained. Prostate not unusual. There was a small right hydrocele.

Urine: specific gravity 1.033, no albumin, sugar, or red blood cells, but a few pus cells. Hemoglobin 10.8 gm. per 100 cc. Red blood count 4,320,000, leukocytes 5,800. Wassermann negative.

A complete gastro-intestinal examination disclosed no organic lesion.

An orthopedic consultation was obtained because of the backache radiating to the thighs; x-rays showed slight lipping of the tenth and eleventh dorsal vertebrae; the spinal motions were normal; the right leg was a bit shorter than the left and a lift was placed under the right heel. He was put on treatment for angina pectoris.

A few days after the completion of this series of examinations he was admitted to the hospital because of sudden, unheralded and complete anuria of 4 days duration. He was vomiting and his nonprotein nitrogen of the blood was 110 mg. per cent. The next day cystoscopy was done. The bladder was normal and the catheters passed easily up the ureters, meeting no appreciable obstruction and resulting in immediate hydronephrotic drip of clear urine from both pelvises. The catheters were left in place and in the first 24 hours the right drained 2000 cc and the left 1000 cc. Pyelograms showed an almost normal right kidney pelvis, the left showing some irregularities suggestive of pyonephrosis. When the catheters were withdrawn there was again complete anuria.

1 Read at annual meeting, American Association of Genito-Urinary Surgeons, Absecon, N. J., June 6, 1947.
and they had to be reinserted. Transfusions were attempted, but caused cardiac pain and it was deemed best not to repeat the attempt in spite of marked secondary anemia. A few days later a right nephrostomy was done. No obstruction of the upper ureter was noted and the cortex seemed only a little thinner than normal. The nonprotein nitrogen became normal. On one occasion urine appeared in the bladder but thereafter none appeared, so left nephrostomy was performed at the end of 2 weeks, and the cortex of the left kidney found to be fairly well preserved. At this time he was very anemic; the hemoglobin reading being 7.0 g. per 100 cc.

The renal function rapidly improved, but not until the ureters were dilated with bulbs 8 on right and 10 on left on two occasions was the ureteral flow re-established.

The patient was sent home voiding normally, and with both nephrostomy wounds closed. On October 23, 1942, three months after the onset of anuria, his urine contained pus and marked secondary anemia persisted. During the next 3 years he reported for observation and treatment at irregular intervals. At first the nephrostomy wounds reopened occasionally, but they remained closed after March 1943, five months after his discharge from the hospital. His urine remained infected in spite of the use of sulfa drugs; his anemia improved at first, but after a few months relapsed. He continued treatment for angina pectoris; and almost every time he reported to the clinic his ureters were dilated with bougies with bulbs 8 or 10, which passed with ease. His course was gradually downhill, the anemia persisting, the urinary infection persisting, and the renal function diminishing.

In January 1945 he was admitted to the hospital for penicillin treatment. The urine was cloudy with pus; the nonprotein nitrogen was 82 mg. per cent; his blood pressure was 175/90. Cultures at various times during the course of his illness had showed staphylococcus, hemolytic streptococcus, and B. aerogenes; and the infection had not yielded to the sulfa drugs. During this admission he was given 100,000 units of penicillin per day for 3 days, but without effect on the urinary infection.

Six months later his hemoglobin showed 7.5 g. per 100 cc blood, so he was given transfusions, this time with no reaction and his hemoglobin rose to 10.3. The phenolsulphonphthalein output registered zero in 2 hours. In October 1945 catheters were easily run up to the kidney pelvis; the urine contained pus and the indigo carmine did not appear in 14 minutes. He was again admitted for transfusions since the hemoglobin had gone back to 7 gm.

In February 1946 he was admitted in uremia and with advanced anemia and died. Autopsy showed a fibrous retroperitoneal mass covering the promontory of the sacrum and extending up to the kidney. The ureters, which were structurally normal and showed no strictures, ran through the lateral margins of this mass, and were enveloped and compressed by it, but not invaded. The kidneys were completely destroyed by infection and back pressure. I quote from the autopsy report:

"The retroperitoneal tissues surrounding the aorta and extending from the
kidneys to below the sacral promonitory show almost complete replacement with a dense, greyish-white fibrous connective tissue mat. This tissue is 2 cm. in thickness at its thickest point and extends out to and including the ureters.

![Image of retroperitoneal tissue](image)

**Fig. 1.** Case 1. Sections from autopsy specimen of retroperitoneal tissue

The ureters, aorta and vena cava all traverse this dense mass of fibrous tissue which is sufficient to nearly occlude the lumina of the ureters.

"Sections of this dense fibrous mass (figs. 1 and 2) reveal that it is composed
entirely of extremely dense bundles of collagen. This tissue is poorly nucleated, but the cells, where seen, are of typical fibrous connective tissue type. This fibrous connective tissue surrounds without invading the large vessels, lymph nodes, nerves and ureters.

"Sections at the edge of this area of fibrosis reveal a rather sharp demarcation, but absolutely no evidence of encapsulation. The cells composing this area of fibrosis are all extremely small and show no evidence of malignancy. In one portion the fibrous tissue is invading the surrounding skeletal muscle, which shows, as previously surrounded muscle is examined, a gradual atrophy and disappearance. This fibrous tissue has no sign of malignant characteristics and has rather the appearance of a keloid."

The second case is that of a man whose age was 43 when he first came under urological observation in January 1946. Previous to this he had been under observation by various divisions of the Henry Ford Hospital over a period of about 22 years, so his past history was well known to us. He was never robust, was always under par, and showed a mild secondary anemia over several years.

In 1924 cholecystectomy and appendectomy were performed. Following this he had a wound infection that cleared up in 3 months.

Between 1924 and 1943 he received attention at various times for upper respiratory infections, furuncles in various regions, hypothyroidism, and in 1941 an attack of bacillary dysentery. As this period is reviewed the impression is gained that he was a man rather unusually subject to infections. At no time in this period did the urine show any abnormality and the blood Wassermann test was negative.

In 1943 he first complained of lumbo-sacral backache and late in 1945 the
back pain became severe. X-rays showed calcification of the intervertebral
disc between the eleventh and twelfth thoracic vertebrae and a number of calci-
fied glands in the right lower abdominal quadrant. There was one large dental
abscess.

In January 1946 the back pain increased. Neurological examination gave
normal findings, so his back was strapped, but with no relief. At this time he
was seen in urological consultation because of the backache associated with
ache in the testicles. The urine was clear, of normal specific gravity, and routine
examination of the genito-urinary tract gave no indications for more extensive
examinations. In February the back pains became so severe as to require
Demerol for its relief. At this time the blood showed hemoglobin 11 grams per
100 cc, red cells 3,800,000, leukocytes 8,250; urine, specific gravity 1.032, no
albumin or sugar, and no red blood cells or pus cells. An x-ray of the urinary
tract showed no abnormality. His pain increased, being worse at night. Bar-
ium enema and chest x-rays showed nothing abnormal.

A pantopaque myelogram showed some suggestion of a pressure defect in the
region of the third lumbar disc and also in the region of the lumbo-sacral
articulation. So on February 15, 1946 a bilateral laminectomy between the
third, fourth, and fifth lumbar vertebrae was done. Some extradural adhesions to
the lumbar nerves were freed and the presence of some varicosities noted.

It was noted on March 19 that there were no urinary or gastro-intestinal symp-
toms; that the back symptoms were largely relieved, but that there was pallor,
asthenia and no gain in weight. The prostate was again examined but it showed nothing suspicious. On March 29 there was sudden complete urinary
suppression, without any pain.

Physical examination showed no general or local lymph gland enlargement;
the thyroid was not enlarged; heart and lungs were clear to auscultation and
percussion; blood pressure 138/100; no abdominal tenderness or masses; no
abnormality noted so far as bones or joints were concerned except the recent
operation on the back; deep reflexes were normally obtained. The Wassermann
test had always been negative. Catheterization showed no urine in the bladder.
Cystoscopy was done and catheters passed up both ureters, finding urine in
both pelves. Pyelograms were unsatisfactory, but some dilatation of the pelvis
on the left was noted. The catheters were left in place and dripped continu-
ously for some hours. Next day they became plugged and were withdrawn.
The one on the right was re-introduced all the way to the pelvis, but that on the
left would pass only part way, although urine dropped from it freely. With
these catheters in place the nonprotein nitrogen fell in 3 days from 73 mg. per
cent to 24 mg. per cent. When the catheters were withdrawn no urine reached
the bladder and it was necessary to re-introduce them.

Because of his anemia and of the puzzling character of his course in the past,
it was suspected that the ureters might be involved in some malignant process,
and the decision was made to explore the abdomen.

On April 4, 1946 a right para-median lower abdominal incision was made.
The peritoneal surfaces were smooth, the kidneys felt normal, the liver was not
enlarged and no nodules were felt on its surface. The gallbladder had been previously removed and there were numerous adhesions in this region. A retroperitoneal flat mass with indistinct lateral margins was felt covering the promonitory of the sacrum. A bit of this was removed for biopsy and frozen section showed it to be inflammatory in nature. I quote from the pathological report:

"Sections of the retroperitoneal tissue reveal alternating areas of dense, fibrous, connective tissue and fat (fig. 3). Throughout the entire sections examined in a diffuse infiltration of small, round and wandering cells, occasional eosinophils and polymorphonuclear leukocytes. In some instances the lymphatic cells are collected into small follicles. Considerable fibroblastic activity is seen but no abscesses are present and there is no evidence of a specific etiology. There is no microscopic evidence of a neoplastic process."

The ureters were located below this mass, traced up, and found to pass through this tissue near its lateral margins. Both ureters were dissected entirely free from this enveloping tissue and seemed entirely normal. They were left free in the abdominal cavity, no attempt being made to cover them with peritoneum. The abdominal wound was closed with drainage because of oozing. The ureteral catheters were removed in the next 2 days. Wound healing and convalescence were uneventful and there has been free and normal urination ever since. He was given deep x-ray therapy directed at the inflammatory mass described above, and his health has improved remarkably. He is no longer anemic, has put on weight, is working regularly and feels the best he has felt in several years. We have followed his kidney condition by means of the phenolsulphonphthalein test and excretory urography. The phenolsulphon-
phthalein output has been normal and his last urogram showed no evidence of ureteral obstruction.

On May 13, 1937 a cystoscopic examination was done. The bladder seemed everywhere normal. Indigo carmine appeared from the left ureteral orifice in 3 minutes and from the right a few seconds later. A catheter was easily passed up the left ureter, but it could not be engaged in the right ureteral orifice.

On May 19, 1937 a phenolsulphonphthalein test showed 60 per cent in the first hour and urinalysis showed as follows: specific gravity 1.024, no albumin or sugar, no pus cells, occasional red blood cell.

**Discussion**

Here are reported two instances of ureteral compression due to envelopment by a retroperitoneal, non-malignant mass. In both instances there was sudden, complete anuria. In both there was a preceding period of reduced health, unexplained anemia, and backache. In neither had there been any preceding urinary disturbance. The microscopic appearance of the retroperitoneal tissue in the 2 cases shows some similarity or relationship between them. In the second case the tissue is obviously inflammatory in origin and fairly recent or acute. In the other it could well be in the nature of scar, or the result of past inflammations. In other words, it is conceivable that the tissue from the first case represents a later stage of a process of which the tissue of the second case represents an earlier stage and this would be in accord with the different periods during which each was observed.

We were much puzzled by the course of events in the first case. We could not detect any ureteral strictures. Ureteral catheterization was always easy, yet there was pelvic retention of urine, and persistent and increasing pyelonephritis, and renal deterioration.

As I look back on it, it seems that we were rather stupid not to recognize that there was ureteral obstruction of extra-ureteral origin and that we should have explored the abdomen. It seems likely that had we done so and freed the ureters as we did in the second case, the result would have been very different.

It seems highly probable that the backache in both instances was due to the inflammatory retroperitoneal activity, and it seems possible that the reference of that ache, in one case to the thighs and in the other to the testes, was due to irritation of the ureters as they became involved in this enveloping and compressing tissue.

We have no theories to offer as to the genesis or development of these inflammatory masses or processes. It is possible that they have something to do with the lymph glands of this region, but there is in neither case any history of preceding infection of the genitalia, legs, urinary tract, or of any evidence of diverticulitis of the bowel. The possibility of brucellosis has been suggested. This could not be proved now and was not suspected at the time. I do not know whether this is a new observation or not. It is certainly new to me and to our pathological department but it is entirely beyond the bounds of possibility that there have been only two instances of this condition.
Since the process is not invasive it would probably make its presence known as it developed and spread only by the result of compression of the neighboring structures as they became enveloped by it. Of these structures the ureters would be most apt to give unmistakable evidence of their compression.

Both these patients had severe backache, which may be attributable to this inflammatory process, but obscure backaches are common and rarely demand radical or emergency treatment, while complete obstruction of the ureters urgently demands attention.

In a thorough review of the urological literature I have not found similar cases reported. If I have overlooked any reports or if any of you have seen such cases, I shall be much interested to learn of them.