SPONTANEOUS CIRCUMRENAL HAEMATOMA

A REVIEW AND REPORT OF TWO CASES

BY

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The term "spontaneous circumrenal haematoma" is self-explanatory and excludes, by definition, cases of traumatic origin. The condition is said to be a rare one, though I have had to deal with two cases in the past nine years. These two cases and a short review of the subject are thought to be worth publishing, as no mention is made of the condition in most of the standard surgical textbooks, and no description of it had appeared in any English journal until that of Heritage in 1934. The condition is referred to by Hamilton Bailey (1944) and in American papers under the title of spontaneous perirenal haematoma. At risk of seeming pedantic, however, the Graeco-Roman bastardy of the word "perirenal" should condemn it. The condition is probably not as rare as the literature would suggest; it may possibly be the underlying lesion in such labels as "retroperitoneal sarcoma" and "retroperitoneal haematoma of obscure aetiology."

Case 1

A youth aged 18 was suddenly taken ill at sea with abdominal pain and vomiting on Nov. 14, 1945. There was no history of trauma. Next morning his condition was worse. He had a furred tongue and a weak thready pulse of 130; his temperature was 103° F. (39.4° C.). There was extreme tenderness and rigidity in the right loin and right iliac fossa. Examination of the urine, including centrifugalization and microscopy, was negative.

A diagnosis of retrocaecal appendicitis with generalized peritonitis was made; he was shipped ashore and admitted to the Calvary Hospital, Hobart, Tasmania. He was seen by two surgeons, who agreed with the diagnosis, and immediate operation was undertaken.

At operation the appendix was normal, but a large retroperitoneal haematoma, which appeared to surround the kidney, was found in the right paracolic gutter. The incision was extended and the swelling was being explored further when the anaesthetist reported that the patient was *in extremis* and the abdomen had to be quickly closed. He was given a blood transfusion of 4 pints (2.2 litres) and his condition improved considerably. Ten hours after the first operation the wound was reopened. This approach, rather than a lumbar incision, was chosen to enable the presence of a left kidney and its condition to be ascertained. The ascending colon was mobilized, and after mopping out blood clot a transperitoneal nephrectomy was carried out. After this operation he made a rapid and uninterrupted recovery.

Pathological Report.—"The kidney is lobulated, resembling the foetal type, with dilated pelvis and calices. No visible inflammatory reaction in the pelvic cavity. A haemorrhagic area over the hilum shows only a layer of tissue between the pelvis and the exterior. Sections show widespread interstitial nephritis, with areas of small round-cell infiltration scattered through the renal tissue. At the site of haemorrhage there is complete replacement of the renal tissue by inflammatory scar tissue."

Pyelograms, taken two and six months after operation, showed a large left kidney and large pelvis with good secretion. Now, three years after, the patient is perfectly fit and still serving as a regular officer in one of the armed Forces.

Comment.—The points of interest are: (1) this was a case of spontaneous circumrenal haemorrhage, probably secondary to a congenital hydronephrosis in a foetal-type kidney;

(2) it presented as an acute surgical emergency, and was diagnosed by three surgeons as acute appendicitis with generalized peritonitis; (3) the urine contained no red blood cells; and (4) uninterrupted recovery followed nephrectomy.

Case 2

A man aged 35 was admitted to Worthing Hospital on June 21, 1948, complaining of abdominal pain and backache for 24 hours. The pain seemed to come from the right loin and was maximal in the right iliac fossa. There had been no vomiting and no central pain, but his abdomen felt distended. Bowels and micturition were normal. He had had amoebic dysentery in 1942, with recurrences in March and September, 1943. There was no history of recent trauma.

On examination he was obviously ill and in pain. His temperature was 100° F. (37.8° C.), and pulse 92. The tongue was furred. Localized tenderness and guarding were present in the right iliac fossa, over McBurney's point and extending up into the loin. There was guarding of the lumbar muscles, but no abdominal rigidity. Considerable meteorism was noted. The liver was not enlarged, and neither kidney was palpable. Peristaltic sounds were normal. Rectal examination revealed no tenderness. The heart sounds were normal. The blood pressure was 150/95. An occasional leucocyte and some ammonium urate crystals were found in the urine, the pH of which was 6. No red blood cells or casts were seen. There was no growth on culture for 24 hours. A blood count showed: haemoglobin, 80%; red cells, 3,610,000; colour index, 1.11; white cells, 14,800 (90% polymorphs); blood urea, 46 mg. per 100 ml. Amoebae were not seen in one specimen of faeces. Two hours later his condition was worse and operation was advised. The pre-operative diagnosis was retrocaecal appendicitis, ? amoebic perityphilitis.

Operation.—Through a right paramedian incision I found that the caecum was ballooned and the small bowel distended with gas. The appendix was normal. A large retroperitoneal haematoma involved the posterior aspect of the caecum and ascending colon. It filled the right paracolic gutter, extended over the brim of the pelvis, and seemed to originate from the kidney, which was large but smooth. There was no evidence of amoebic colitis. The abdomen was closed. The anaesthetist noted that the blood pressure was 180/100. This hypertension persisted post-operatively.

Post-operative Course.—This was marked by progressive vomiting and abdominal distension. His blood pressure remained high. He developed a classical adynamic or neurogenic ileus, which failed to respond to continuous gastric suction and intravenous therapy. His condition slowly but inexorably deteriorated until death supervened on the eleventh post-operative day.

Necropsy.—A large unorganized clot surrounded the right kidney and stripped up the peritoneum. Both kidneys and suprarenals appeared normal. There was no evidence of active amoebic disease, and the liver was normal.

Microscopical Report (Dr. Standish).—"There is no microscopical evidence of haemorrhage in the suprarenals, and their vessels show no evidence of either endarteritis or polyarteritis nodosa. The surrounding blood clot shows a very early stage of organization, but no inflammatory change. There is no evidence of the cause of the haemorrhage either in the suprarenal or in the clot itself."

Comment.—(1) The hypertension found at operation and afterwards was an interesting phenomenon and one which has not, so far as can be ascertained, been noted before in the literature. The patient's general practitioner, Dr. Shepherd, told me that he had found the blood pressure normal at several previous examinations. The cause of this is presumably compression of the renal parenchyma by clot, and is analogous to the hypertension produced experimentally by Page (1939) and Graef and Page (1940). These workers were able to produce a sustained hypertension in dogs from perinephric scarring by silk or from compression of the kidneys by wrapping "cellophane" round them.

(2) The cause of death was adynamic ileus, due again, presumably, to irritation of the aortico-renal plexus by clot. I was satisfied at necropsy that there was no mechanical obstruction to the second part of the duodenum by the haematoma itself—an explanation which had been suggested. Some degree of meteorism is, of course, common after nephrectomy. It is my impression that a greater degree of ileus is found after a right nephrectomy than after a left; but this is only an impression, and I am not aware that the matter has been investigated.

Discussion

Aetiology

Polkey and Vynalek (1933), of Chicago, have written the fullest review of the literature up to 1933 and collected 178 cases, mostly from Continental writers. They suggested that "sudden renal congestion" might explain some of the spontaneous cases in which no cause was discovered. In experiments on dogs they ligated the renal vein with silk thread and found subcapsular haemorrhage in 33%, extracapsular haematoma in 42.8%, and some degree of parenchymal haemorrhage in 100%.

I have found a further 13 cases reported in the literature in the last 15 years. The pre-operative or clinical diagnoses of these cases, together with those of my own two, and the stated causes and results of treatment, are shown in the accompanying Table.

Spontaneous circumrenal haematoma must be regarded as a symptom-complex or complication and not as a disease entity. It follows that the lesions responsible for it are numerous. They fall into three main groups—renal, extra-renal, and the blood dyscrasias.

Group 1: Lesions of the Kidney and Suprarenal Gland.—A lesion of the kidney or suprarenal accounts for about 60% of cases. Nearly every lesion affecting these organs has been reported—renal and suprarenal cysts and tumours, chronic nephritis, hydronephrosis, aneurysm of the renal artery, lithiasis, tuberculosis, and thrombosis of the renal vein. Usually the haemorrhage is the precipitating symptom, with no previous history pointing to renal disease. Thus in one of Heritage's cases it was only with difficulty and after microscopical examination that "ghost cells" of a haemorrhagic and necrosed neoplasm were found in the clot.

Group 2: Extrarenal Retroperitoneal Lesions.—Included in this group are such differing conditions as perinephritis, arteriosclerosis, aneurysm of the abdominal aorta, and hypertension. One case was associated with periarteritis nodosa.

Group 3: The Blood Dyscrasias.—Haemophilia, polycythaemia, leukaemia, scurvy, thrombocytopenic purpura, and Hodgkin's disease have all been reported.

Clinical Course

Clinically one must distinguish between the acute and the chronic form of the disease. The acute form is by far the more common. The onset is sudden and dramatic and presents as an acute surgical abdomen. There may be no history suggesting renal disease. The classical findings

are the triad of pain, a renal tumour, and signs of internal haemorrhage (Lenk, 1909), but these are seldom all present. The muscular rigidity usually precludes the palpation of a renal tumour in the acute form of the disease. Pyrexia is due to blood absorption, and pressure on the renal parenchyma may cause albuminuria or hypertension. Later, increasing abdominal distension, vomiting, and prostration confuse diagnosis and suggest the perforation of an abdominal viscus. In fact, the haematoma may perforate the peritoneum and give a clinical picture very similar to that of a "ruptured ectopic," but in a male.

A more insidious form of the disease occurs with a sense of fullness or discomfort in the affected loin. Here there is time for investigation. A renal tumour can as a rule be felt and the diagnosis is usually hydronephrosis or hypernephroma. Careful study of pyelograms, as Ekman (1946) has pointed out, will show displacement of the kidney: the direction of displacement may indicate the location of the source of the haemorrhage.

The only reported cases in which correct pre-operative diagnoses were made are described by Cibert, Vachon, and Cavaillher (1942). In all these cases a tender tumour in the loin was present. The cases were of the insidious or chronic form and they all presented a similar physical sign, not previously described in the literature. This consisted of a lumbar ecchymosis, which in time tracked down to the scrotum. This ecchymosis, when present with a tumour in the loin and in the absence of trauma, is pathognomonic of spontaneous circumrenal haematoma.

Treatment

Where the condition is suspected emergency intravenous pyelography should be undertaken. It is essential to confirm the presence, and preferably the normality, of the opposite kidney. To quote the words of Cibert, Vachon, and Cavaillher: "La conclusion, exprimée par tous les auteurs, découle de cette constatation: l'opération s'impose." Non-operative treatment was uniformly fatal in every published case, except one. It must be accepted, however, that the diagnosis is usually made with certainty only at operation or post mortem. Nevertheless, operation should be advised as soon as the necessary resuscitation has been undertaken, for three reasons: (1) to arrest haemorrhage and save life; (2) to prevent the side-effects of a large haematoma—meteorism, ileus, hypertension, sepsis, and possible rupture into the general peritoneal cavity; and (3) to explore what may well prove to be a malignant condition of the kidney or suprarenal gland.

In theory it might occasionally be possible, in the extra-renal type of haematoma, to clear out the clot and ligate the bleeding vessel. According to the literature this never seems to have been done. If the haematoma appears to have originated from the kidney, nephrectomy should be carried out. If bilateral renal disease is suspected or proved it might be wiser to rest content with packing or drainage. If both kidneys appear normal and no cause is apparent the best results seem to follow nephrectomy.

As the Table shows, seven out of 15 cases were subjected to nephrectomy without a death.

Table showing Cases Reported in the Last 15 Years

Author	Sex and Age	Clinical Diagnosis	Cause	Treatment	Result
Polkey and Vynalek (1933)	F. 39	Ovarian cyst	Unknown	Drainage	Recovery
Heritage (1934)	M. 51	P.D.U.	Neoplasm	Packing	
"	M. 65	P.D.U.	Suprarenal neoplasm	Laparotomy	Death
"	M. 41	Acute cholecystitis	Unknown	Drainage	"
Carver (1939)	M. 43	Hydronephrosis	Hydronephrosis	Nephrectomy	Recovery
"	M. 47	"	Hypertension	Nil	Death
Elmer and Wyngarden (1939)	F. 46	Hypernephroma	Unknown	Nephrectomy	Recovery
Daro and Todd (1941)	F.	Retroperitoneal cyst	"	"	"
Cibert, Vachon, and Cavaillher (1942)	M. 51	Circumrenal haematoma	"	"	"
"	M. 61	"	Suprarenal cyst	"	"
"	M. 58	"	Chronic nephritis	Expectant	"
Ekman (1946)	M. 78	Appendicitis	Unknown	Laparotomy	"
"	M. 48	Hypernephroma	Hypernephroma	Nephrectomy	"
Martin	M. 18	General peritonitis	Hydronephrosis	"	"
"	M. 35	Appendicitis	Unknown	Laparotomy	Death

Summary

The condition of spontaneous circumrenal haematoma is described, with two personal case histories.

Though rare the condition is an important one to bear in mind, as it may enter into the differential diagnosis of the acute abdomen and if treated expectantly is nearly always fatal.

The aetiology and clinical course are discussed. An acute and a chronic form can be distinguished.

Treatment should consist in surgical intervention and, in most cases, nephrectomy.

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APLASTIC ANAEMIA FOLLOWING NEOARSPHENAMINE

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Aplastic anaemia is a well-recognized but rare complication of organic arsenical therapy. Wintrobe (1946) states that fewer than 100 cases have been reported in the literature, also that less than one-half of the cases described showed complete aplasia of the marrow. Recovery after complete aplasia has been recorded, however (Wintrobe, 1946). The case described below, which exhibited anaemia, leucopenia, thrombocytopenia, and a completely aplastic marrow on sternal puncture, seems to fall into this class and to be worthy of record.

Case Report

The patient, a man aged 52, contracted primary syphilis and gonorrhoea abroad in June, 1946. He received 37 injections of neoarsphenamine and 37 of bismuth during the next nine months.

He noticed nothing abnormal until April, 1947, when he had a severe epistaxis lasting 16 hours. The red blood cells numbered less than one million. Injections were stopped, but a month later further severe nose-bleeding occurred, with the appearance of petechiae and "papules" on the arms and legs. His haemoglobin was 30%; colour index, 1; red blood cells, 1,500,000 per c.mm.; white blood cells, 3,000 per c.mm. (L. 78%, M. 1%, P. 21%); platelets, 25,000 per c.mm.; coagulation time, 14 minutes. Wassermann reaction and Kahn test were negative. The spleen was never palpated.

He was treated with iron, a fresh blood transfusion, and 10 ml. of pentnucleotide every 12 hours. He received altogether some 6 pints (3.4 litres) of fresh blood. Four days later a blood count showed: red cells, 1,670,000 per c.mm.; haemoglobin, 30%; colour index, 0.9; bleeding time, 3½ minutes; coagulation time, 6 minutes; leucocytes, 2,000 per c.mm. (L. 73%, N. 2%, E. 3%, P. 22%).

He then returned to England, and during the voyage was given 1 ml. of "hepastab" daily, 45 gr. (3 g.) of iron and ammonium citrate, and 5 mg. of folic acid thrice daily. On his return he stated that he did not feel very ill, but became giddy

and tired on the least exertion. Up to this time he had had 11 blood transfusions in all, the last being on May 19. No further epistaxis had occurred, but he had noticed some oozing of blood from his gums and he found that he bruised easily and "came out in little red spots." Previous illnesses and family history revealed nothing relevant.

He was admitted to hospital under our care on May 31. He was a very co-operative man, very pale, and obviously grossly anaemic. His skin was sallow and sunburnt. There was pronounced foetor oris; his teeth were in a bad condition, with much dental sepsis; the gums were soft and spongy, with pus oozing from them. The tongue was furred but moist. No bismuth line was present. No glands were palpable and no petechiae or bruises were seen. Over his legs and feet the skin was scaly, pigmented, and ulcerated. The blood pressure was 120/60. A soft systolic murmur was present over all areas in the praecordium. The capillary resistance test was strongly positive in three minutes at a pressure of 90 mm. Hg. Nothing abnormal was detected in the chest apart from an occasional rhonchus at the bases. The spleen was not palpable.

The pupils reacted normally to light and to accommodation. Ophthalmoscopy showed normal optic disks. No exudates were seen, though multiple large haemorrhages were present in both retinae. The central nervous system was otherwise normal.

Chart showing haemoglobin findings

The diagnosis of aplastic anaemia due to arsenical therapy was made. The haemoglobin findings are shown in the accompanying graph. Radiographs of lungs and tibiae were negative. The B.M.R. was 4%.

The patient was kept in hospital until Sept. 16, when he was discharged home. During this time his haemoglobin rose from 20% to 104%. A total of 2 pints (1.14 litres) of whole blood and 29 pints (16.5 litres) of packed red cells was given over this period. He did not react adversely to any transfusion, and over the whole period made slow but steady improvement. Penicillin was administered, 40,000 units four-hourly, for the first month, the dosage gradually being reduced over the following three months to 100,000 units twice daily. Liver extract was given by injection (hepalon) 2 ml. daily at the onset, being reduced to twice a week after two months. He also received pyridoxin 100 mg. daily, ferrous sulphate 10 gr. (0.65 g.) thrice daily, two multivitamin tablets thrice daily, and folic acid tablets (15 mg.) daily.

On Oct. 2, 14, and 27 he was readmitted for the transfusion of 2 pints (1.14 litres) of packed cells on each occasion. The condition of the skin of the feet and legs greatly improved, and the capillary resistance test, still positive on Oct. 14, became negative after this time.

Two further pints of packed cells were given on Nov. 20 and Dec. 18. It was apparent at this time that he was making steady progress, and that he was beginning to pick up on his own. He was last seen on May 31, 1948, when he was in excellent general condition. The Wassermann reaction and Kahn test were negative. Our last report, received from abroad in October, 1948, was that he was in perfect general health and had received no further treatment.

Haematological Findings.—On June 2, 1947, these were as follows: haemoglobin, 22% (3.03 g.%); red cells, 1,210,000