
A POSSIBLE HAZARD OF SPLENECTOMY

Case Report: Torsion of the Wandering Spleen

R. ATTARD

M.D., B.Sc., F.R.C.S.

T. J. AGIUS-FERRANTE

M.D., B.Sc., F.R.C.P., D.C.H.

*St. Luke's Hospital
and Royal University of Malta*

King and Shumacker (1952) were the first to draw attention to the possibility of fulminating infection occurring after splenectomy. They described the cases of

five infants under six months who underwent splenectomy for congenital haemolytic anaemia. Four of them developed fulminating meningitis or meningococcaemia in from six weeks to three years after operation and one of them died. The fifth infant developed a rapidly fatal febrile illness. Since then several papers have been published supporting the findings of King and Shumacker.

Case Report

R.E., a four month old male infant, was admitted to St. Luke's Hospital on 11 May, 1965 for "gastroenteritis and dehydration". On the morning of admis-

sion, he had vomited bile-stained fluid three times and he had passed three loose stools over the previous twentyfour hours. The baby had been constipated for two days before then but otherwise he had been healthy since birth. He had been delivered normally at full term and his birth-weight was 9 lbs.

On examination, the infant was pale and sallow. Temp. 99.6 F., Pulse 104/m., Resp. 40/m. No cyanosis, jaundice or lymphadenopathy. Heart and lungs n.a.d. Abdomen was somewhat distended and a distinctly palpable mass could be felt occupying the right lumbar region and extending into the right iliac fossa. Investigations: Hb 72%, WBC 11200/c.mm., Neut. 57%, Eos. 1%, Lymph. 34%, Mono. 8%. A straight film of the abdomen (12 May 1965) showed small fluid levels and a soft shadow in the right iliac region. Urine analysis: traces of protein and a few granular casts. Blood urea: 56 mg./100ml.

On admission, the infant was put on intravenous fluids, the stomach was aspirated at intervals and penicillin and streptomycin were started. However, his condition continued to deteriorate and by the morning of the thirteenth May the stomach aspirate had become coffee-ground with fresh blood occurring at intervals. It was therefore decided to explore the abdomen.

Through a right paramedian incision, the mass in the right lumbar region was found to be the spleen, measuring $8.8 \times 6.6 \times 4.2$ cms. The convex "diaphragmatic" surface was lying against the anterior abdominal wall. The pedicle attached to the hilum showed three twists and was 9 cms. long, disappearing towards the left hypochondrium. Splenunculi were embedded in the pedicle, the largest of which was 0.7 cms. in diameter. All other organs were in their normal position; there was no intestinal malrotation. After transfusion and ligation of the pedicle, the spleen was removed and the abdomen was closed in layers without drainage. Microscopical examination of the spleen, which weighed 99 grams — the normal average weight in a three to six month old boy is 18.9 grams (Spector, W.S., 1956) —

showed recent haemorrhages in the pericapsular region, as well as congestion of sinusoids in the vicinity. Malpighian corpuscles were not prominent. There was autolysis of the inner pulp.

The infant received a transfusion of 30 c.c. of blood during and after the operation from which he made a good recovery. In the postoperative period, he developed an enteritis which was treated with furoxone. No pathogenic organisms were grown from the stools. The sutures were removed on the tenth day. A mild sepsis subsided after extrusion of some catgut. The infant was discharged in a healthy state on 16 June, having steadily gained two pounds in weight over the previous month. Postoperative investigations on the infant and its parents failed to reveal any evidence of congenital haemolytic anaemia.

This was then a case of torsion of a wandering spleen. It is a rare surgical emergency and up to 1952 less than 150 cases had been described (Maingot, R., 1952). This is the only such case recorded so far at St. Luke's Hospital since it was opened in 1951. A wandering spleen, a rare condition itself, may be found in any part of the abdomen and it is particularly liable to torsion (Aird, I., 1957).

On the 25th June, 1965, the infant was seen in the paediatric outpatient department and was found to be in excellent health. Late the same afternoon, he was admitted as an emergency in a state of semi-coma in a very ill condition. His temperature was 103°F., pulse rate 170/m., respiration rate 76/m. There was a generalised purpuric rash, more marked in the extremities, and the skin was dry and dehydrated. There were no cyanosis, anaemia or jaundice. There was no obvious localising infection and a diagnosis of fulminating septicaemia was made. Penicillin, streptomycin and chloromycetin were started and an intravenous drip was set up. During the next fifteen hours, the infant vomited twice and passed loose yellow stools three times. The only investigation carried out was a blood count: Hb 88%, RBC 4580000/c.mm., WBC 28000/c.m., Neut. 83%, Eos 2%, Lymph.

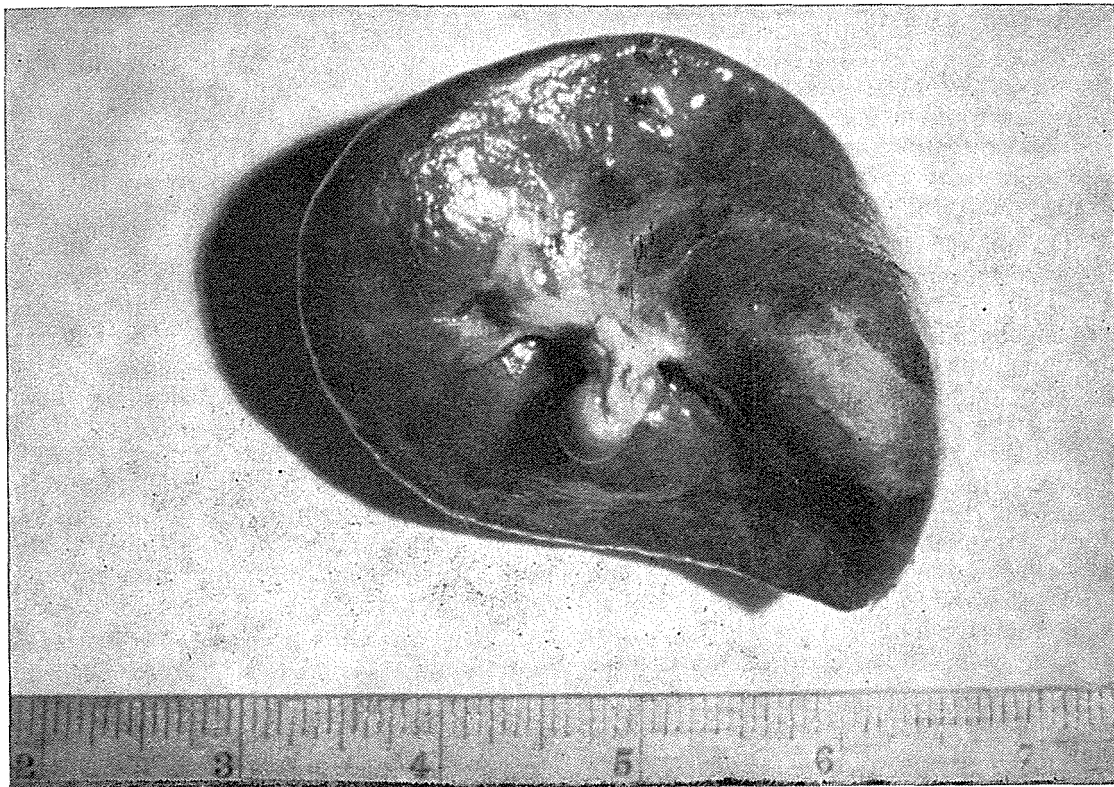


Figure: the visceral surface of the spleen distinctly shows the largest of the splenunculi found at operation as a round knob in the centre of the organ.

12%, Mono 3%, Platelets 100700/c.m. The patient was dead at 8 a.m. on 26 June, 1965. Permission for autopsy was refused.

Discussion

King and Shumacker (1952) thought that serious infection was likely to occur after splenectomy only in infants. Gofstein and Gellis (1956) concluded in their review of cases that there was some correlation between splenectomy and sepsis but did not find a relation to age at operation. Smith et al. (1956, 1957) put the incidence of severe infection after splenectomy at 28% in their series of infants and children. Huntley (1958) discussed seven patients who developed severe infection after splenectomy in a series of 43, and five of the seven were under one year. Four out of nine infants in Horan's series (1960) died. Broberger et al. (1960) calculated at 20% the risk of serious infection after

splenectomy with a 50% mortality rate if splenectomy is performed in infancy. On the other hand, Robinson and Sturgeon (1960) found the incidence of a serious infection to be 10% in their series and about 5% in other series. These last two authors introduced the phrase "life-threatening infection" to describe these cases. Lucas and Krivit (1960) saw only a slight but real increased susceptibility to infection after splenectomy. Lawdon, Walker and Walker (1962) described four cases of severe infection in 75 children who underwent splenectomy for various reasons. One of the four was under one year and two of the others died. Horan and Colebatch (1962), in a careful analysis of their own and other series, came to the conclusion that serious infection in childhood is of the order of 8% and the incidence of fatal infection is close to 5%. Lowdon, Stewart and Walker (1966), in an exhaustive study of 1167 patients

undergoing splenectomy in England and Wales in 1961, stated that "it seems clear that in about 2% of cases splenectomy does predispose to serious infection".

A number of authors have none the less denied this casual relationship. Thus, Cole, Walter and Limarzi (1949) found no cases of infection in their series; Miller and Hagedorn (1951) did not mention serious infection as occurring during the five year follow-up of some 75% of their 140 cases. Gross (1953) merely states that he found little evidence to support the view that serious infection may arise after splenectomy. Of the same opinion were Walter and Chaffin (1955) and also Laski and Macmillan (1959) whose paper though, was strongly criticised by Greenberg (1960-61). Other authors reported single cases with no resulting untoward infection.

Characteristic pattern

A study of the literature reveals a characteristic clinical pattern in a large majority of cases of serious infection after splenectomy — a pattern that is also shown by the cases above described. A septicaemia, with or without meningitis, takes a rapid fulminating course and becomes associated with marked circulatory collapse and also sometimes with the Waterhouse-Friedrichsen syndrome. The incidence of septicaemia and meningitis in such cases is much higher than in the normal population (Horan and Colebatch, 1962). Pneumococci have been the immediate cause of serious infection in some 50% of recorded cases (Lawdon et al., 1962). Other organisms involved have been the following: Meningococcus, H. influenzae, E. coli, Staphylococcus aureus, Streptococcus haemolyticus. 8% of these infections occur within two years of splenectomy (Smith et al., 1957; Horan and Colebatch, 1962). Another feature is the recurrent nature of infection — some 40% of cases had recurrent episodes of serious infection (Lawdon et al., 1962; Horan and Colebatch, 1962). A few of the recorded infections have not been septicaemias. Thus, fatal tracheobronchitis (Gofstein and Gellis, 1956), the re-activation of tuberculosis (MacPherson, 1959) and the

dissemination of lupus erythematosus (Carpenter et al. 1959), among others, have been laid at the door of splenectomy. But not all authors (Robinson and Sturgeon, 1960) will accept the casual relationship in these cases.

Factors influencing reaction to splenectomy

The age of the patient is an important factor for there is no doubt that serious infection is most common if splenectomy is carried out in the first year of life, though later ages are not exempt. It should be possible in most cases to defer the operation until the school period as conservative measures almost always achieve symptomatic control of the primary condition. Hence, splenectomy is rarely indicated in the first year of life; and, during childhood, it should not be recommended solely for haematological abnormality but for real disability.

Huntley (1958) noted that three of the seven patients in her series developing severe infection after splenectomy had a primary disease which made them more liable to infection. Robinson and Sturgeon (1960) presumed that in eleven of the thirteen children who developed serious infection after splenectomy in their series of 110 the infection may have been due to the primary condition like spherocytosis or portal hypertension. Horan and Colebatch (1962) concluded from a study of several series that the risk of serious infection occurring in splenectomised patients was as follows: 0% in cases of traumatic rupture; 2% in hereditary spherocytosis and idiopathic thrombocytopenic purpura; 10% in thalassemia and fully 21.4% in patients undergoing splenectomy for portal hypertension. Patients on continued treatment with corticosteroids are also more liable to serious infection after splenectomy. However, the primary disease factor does not appear to be related to the high incidence of fulminating infection in the first year when splenectomy is rarely performed for conditions that themselves predispose to infection. Smith et al. (1957) and Lawdon et al. (1966) recommended that antibiotic (penicillin cover should be given two years after splenectomy to those

infants and children in particular as well as to other patients who, in the light of what has already been stated, are considered to be specially at risk.

Though there can be no doubt that in individual cases splenectomy may predispose to infection, the reason is far from clear. The spleen is the largest single reservoir of lymphocytes in the body and is concerned with antibody production. However, in many cases extensively investigated after splenectomy (Smith et al, 1967; Krivit and Good, 1959; Broberger et al., 1960), no abnormality liable to increase susceptibility to infection has been discovered, nor was any alteration found in known immune mechanism. Saslow et al. (1959) found that 90% of splenectomised adults and older children were able to produce antibodies in amounts equal to those produced by controls. There were, though, a small number of splenectomised patients who showed a lesser antibody response than controls. Hence, Lucas and Krivit (1960) conclude that "a change in immunologic capacity after splenectomy may be an individual characteristic rather than a regular consequence".

The spleen is also concerned with the removal of micro-organisms from the bloodstream. Kerby (1950) showed that the reticulo-endothelial system of animals is less efficient in removing encapsulated bacteria from the bloodstream than in removing non-encapsulated bacteria. This may explain the 50% incidence of pneumococcal infection in these cases.

Recorded cases of congenital asplenia in children and adults coming to autopsy have been few. It is interesting to note that many of these died of fulminating illnesses associated with the Waterhouse-Friderichsen syndrome, or have suffered from life-threatening pneumococcal infections and recurrent attacks of meningitis. As this is the pattern of serious infection occurring after splenectomy, there is no doubt that absence of the spleen does predispose to serious infection (Horan and Colebatch, 1962).

Summary

The first case of torsion of the wandering spleen in St. Luke's Hospital in a

four month old infant, is described. The infant died from fulminating septicaemia just over six weeks after splenectomy. The question of "life-threatening infection" after splenectomy is discussed. There is no doubt that it sometimes occurs, particularly in infancy and in patients suffering from a primary disease which predisposes to infection or which has required the prolonged use of corticosteroids. It is suggested that splenectomy should not, if at all possible, be performed in the pre-school child, and that antibiotic cover might well be given for two years after operation to those patients specially at risk. It would be advisable to follow up all splenectomised patients carefully and to treat, early and vigorously, any infection no matter how minor it may appear to be.

We wish to thank the C.G.M.O. for permission to publish the above case report, Prof. G. P. Xuereb for the histology report and Mr. J. Xuereb for the photograph.

References

- AIRD, I. (1957). Companion in Surgical Studies, second ed., Livingstone.
- BROBERGER O., GYULAI, F., HIRSCHFELDT, J., (1960). Acta paediat., Stock. 49, 679.
- CARPENTER, A.F., WINTROBE, M.M., FULLER, E.A., HAUT, A., CARTWRIGHT, G.E., (1959). J. Amer. Med. Ass., 171, 1911.
- COLE, W.H., WALTER, L., and LIMARZI, L.R., (1949). Ann. Surg. 129, 702.
- GOFSTEIN, R., and GELLIS, S.S., (1956). A.M.A.J., Dis. Children, 91, 566.
- GREENBERG, M. (1960-61). In "Year Book of Paediatrics," ed. Sydney S. Gellis, p. 87. The Year Book Publishers, Chicago.
- CROSS, R.E. (1953) The Surgery of Infancy and Childhood p. 547. Saunders, Philadelphia.
- HORAN, M. (1960) Med. J. Aust., ii, 809.
- HORAN, M., COLEBATCH, J.H. (1962) Arch. Dis. Childh., 37, 398.
- HUNTLEY, C.C. (1958). A.M.A.J. Dis. Children, 95, 477.
- KERBY, G.P., (1950). J. Immunol., 64, 131.
- KING, H., and SHUMACKER, H.B., (1952). Ann. Surg., 136, 239.
- KRIVIT, W. and GOOD, R.A., (1959). A.M.A.J. Dis. Child., 97, 137.
- LASKI, B. and MACMILLAN, A., (1959). Pediatrics, 24, 523.
- LOWDON, A.G.R., STEWART, R.H.M. and WALKER, W., (1966). Brit. Med. J., I, 466.
- LOWDON, A.G.R., WALKER, J.H. and WALKER, W., (1962). Lancet, I 499.
- LUCAS, R.V. and KRIVIT, W., (1960). J. Paediat. 57, 185.
- MACPHERSON, A.I.S., (1959) J. roy. Coll. Surg. Edinb., 4, 305.
- MAINGOT, R. (1952). Lancet, I, 625.

MILLER, E.M., and HAGEDORN, A.B., (1951). *Ann. Surg.*, 134, 815.
 ROBINSON, T.W. and STURGEON, P., (1960). *Pediatrics*, 25, 941.
 SASLOW, S., BOURONDE, B., WALL, R.L. and DOAN, C.A., (1959). *New England J. Med.*, 261, 120.
 SMITH, C.H., ERLANDSON, M., SCHULMAN, I., STERN, G., (1956). *A.M.A.J.Dis. Children*, 52, 507.
 SMITH, C.H., ERLANDSON, M., SCHULMAN, I., STERN, G., (1957). *Amer.J.Med.*, 22, 390.
 SPECTOR, W.S., (1956). *Handbook of Biological Data*, p. 162., Saunders, Philadelphia.
 142, 798.
 WALTER, L.E. and CHAFFIN, L., (1955). *Ann. Surg.*

~~THE VALUE OF THE HISTORY OF MEDICINE IN MEDICAL EDUCATION~~

~~(St. Luke's Day Oration, British Medical Association, Malta Branch, 18 Oct. 1960)~~

~~V. G. GRIFFITHS~~

~~M.D., B.Sc., F.R.C.S. (ENG.)~~

~~Professor of Anatomy and Histology,
Royal University of Malta;
Surgeon, St. Luke's Hospital~~

~~Among the several reasons that have concurred towards my choice of subject for this St. Luke's Lecture are, firstly, a personal inclination to the study of the History of Medicine of several years' standing and, secondly, a potent reinforcement of this interest by way of a very recent archaeological tour of Greece and the Levant which took me to several sites that St. Luke, in the company of St. Paul, must have visited before that eventful day when a propitious Gregale blew them to our shores — a happening which, among other effects, has resulted in this celebration of St. Luke's Day by us doctors in Malta.~~

~~Of St. Luke as a Physician we know little or nothing by way of direct evidence. Even legend has nothing to say of him in this respect, unlike its proliferation as regards his prowess as a painter, whereby hundreds of churches in Europe have made attribution to him of hundreds of their Byzantine or even more recent ikons! That he was a physician cannot be denied, since he is given that specific title by St. Paul himself, and there is much internal evidence besides this in his Gospel and in the Acts of the Apostles. Nor can it be doubted that it was both as physician and as~~

~~companion that he went with St. Paul to Cos, Rhodes, Athens, Pergamum and other places that have this summer been the route of my own historical pilgrimage.~~

~~The Asklepeia of Cos, Pergamum, Epidaurus, Athens and Corinth were medical shrines of the ancient world which naturally held great interest for me and which St. Luke must surely have visited. Asklepios, if not of divine origin, certainly became in time the Greeks' God of Medicine as Imhotep was to the Egyptians. Homer's testimony would make of him a historical, and not just a legendary personage and his sons Podalirius and Machaon figure as surgeons at the siege of Troy; and who, since Schliemann's excavation of Troy and Mycenae, would doubt the factual historical basis of most of Homer's epic? Every one of the temples of Asklepios was a marvellous combination of holy precinct and medical centre, beautifully sited, offering all the amenities and healing aids that we seek in our spas. There the sick in body and mind congregated not only to be attended by the temple priest-doctors, but also to submit at the incubation to the nocturnal visit of the god in the shape of his sacred serpents; and thence went the healed, loud in their praises, leaving behind them tokens of gratitude in the shape of the "ex voto" that now lie on the shelves of the museums.~~

~~I stepped on Cos to stand in the shade of the ancient plane tree sacred to Hippocrates, Father of Medicine. He belongs to History, with records vouched for by Plato, and a fairly reliable birthdate of 460 B.C. His title he well deserves even if only because of Celsus's tribute that he "first separated medicine from philosophy", that is, made personal observations and drew inferences free of preconceptions. The famous Hippocratic Oath in the form that we know it dates only from the 3rd century A.D., but something more than pious belief must have gone to its attribution to Hippocrates. So also the great corpus of 70 to 100 treatises that form the Hippocratic collection is undoubtedly the fruit of various authors at very various dates, but much of it bears the unmistake-~~