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ANEURYSMS OF THE PATENT DUCTUS ARTERIOSUS

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Rokitansky in 1852 gave the classical definition of an aneurysm of the patent ductus arteriosus as “a spherical or ovoid tumour larger in the middle than at either end, but smallest towards the pulmonary artery, with which, as well as with the aorta, it communicates, filled with old or recent thrombus and varying in size from a cherrystone to a hazelnut or even a walnut.”

Interest was aroused in this condition when we saw two post-operative aneurysms within a year. Martin (quoted by Scheef, 1939) is credited with having first described an aneurysm of the ductus arteriosus. Examination of the literature between 1827 and 1955 has yielded some 56 cases which satisfy Rokitansky’s definition. Excellent reviews of the cases can be found in the papers of Scheef (1939), Graham (1940), and Lennox and MacCarthy (1951). Their surgical importance was stressed for the first time by Graham (1940) in an interesting paper entitled “Aneurysm of the Ductus Arteriosus with a Consideration of its Importance to the Thoracic Surgeon”—its importance in the differential diagnosis of mediastinal tumours.

CASE REPORTS

CASE 1.—V. J., a 26-year-old housewife, had a very thin-walled ductus arteriosus doubly ligated elsewhere in June, 1953. At that time a gross dilatation of the pulmonary artery was noticed. The systolic element of the murmur persisted, and three and a half months after ligation the typical ductus murmur reappeared and there was little doubt that the ductus had recanalized.

During the year after the operation she had two attacks of “pleurisy” with negative blood cultures.

On June 17, 1954, she was admitted to the City General Hospital, Sheffield, for investigation.

On admission she had central cyanosis and was in sinus rhythm, the pulse being of normal volume. The apex beat was in the fifth left interspace just outside the mid-clavicular line. The cardiac impulse was a localized thrust indicating some left ventricular hypertrophy. There was a systolic lift over the right ventricular outflow tract and a diastolic thrill palpable in the second left interspace. P₂ was palpable. Auscultation revealed an apical diastolic murmur thought to be conducted from the base. In the pulmonary area there was a loud blowing systolic murmur obscuring the first sound and maximal in inspiration. The second sound was loud and just split, and there was a loud rough diastolic murmur of maximal intensity 2½ cm. outside the parasternal line. Blood pressure was 115/50 mm. Hg.

A radiograph of the chest (Fig. 1a) showed that the transverse diameter of the heart was much increased with marked fullness of the mid-arc curve, above which there was seen to be a curvilinear opacity consistent with calcification in the wall of the aortic knuckle. The main pulmonary arteries were abnormally prominent, associated with a normal prominence of the perihilar vessels. Also the vascularity of the lung fields was generally increased.

Screening confirmed the above appearances; enlargement of the right ventricle was demonstrated, and in the L.A.O. position the posterior border of the heart was superimposed over the spine, which was probably due to an enlarged left ventricle or right ventricle displacing the left ventricle backwards. The main pulmonary artery and its right and left branches were dilated and strongly pulsating, and strong expansile pulsation was seen in the perihilar vessels on both sides.

Appearances were suggestive of recanalization of the ductus.

The E.S.R. was found to be 63 mm. in one hour (Wintrobe) and a few days after admission her temperature went up to 103°F. Blood cultures at this stage grew coagulase-negative staphylococci, fortunately sensitive to practically all antibiotics. She was placed on a million units penicillin six-hourly: full control of the infection did not seem possible, and it soon became apparent that further operation would be necessary.

On July 23 she was operated on under general anaesthesia (Dr. J. Johnston). A left posterolateral thoracotomy (J. T. C.) with excision of the previous scar and resection of the third and fifth ribs revealed dense adhesions between the left lung and the mediastinum. These were gradually freed, and an aneurysm of the ductus arteriosus, the size of an unshelled walnut, with a band of calcification across its middle was discovered. The aorta opposite to the ductus arteriosus, as well as the proximal pulmonary artery, were grossly dilated. The left vagus and recurrent
laryngeal nerves were incorporated in the wall of the aneurysm and had to be sacrificed.

The pericardial sac was intentionally opened and control of the left pulmonary artery obtained. The aorta was mobilized with division of the two uppermost sets of intercostals. Then it was realized that none of the Potts-Smith clamps available was big enough to go around the dilated aorta, and it was obvious that cross-clamping of the aorta was the only way out. The left pulmonary artery was clamped proximal to the ductus, and Crafoord clamps were applied across the aorta above and below the ductus. Part of the aneurysm was excised as a cuff across its equator. The resultant opening in the aorta was about 2 cm. across, and it was repaired with interrupted silk sutures. Owing to the restricted field deep down in the wound this procedure proved to be both tedious and time-consuming. At intervals of 10 minutes the cross-clamps across the aorta were released to ensure an adequate blood supply through the descending and abdominal aorta, while haemostasis was obtained by finger pressure over the aortic opening. Closing of the aortic opening adequately alone took 50 minutes, and the Crafoord clamps were then removed. Attention was next turned to the pulmonary opening, and this was fairly easily closed and a small cuff of aneurysmal wall itself was sutured over it as a reinforcement. The pericardium and the thoracotomy wound were closed, as is routine, with an underwater drain. The whole operation took five hours 10 minutes, during which 12 pints of blood were transfused.

The patient’s immediate post-operative condition was stormy. Her urinary excretion was adequate and post-operative chest radiographs satisfactory. Blood cultures at this stage were negative. She was maintained on penicillin. An electrocardiogram showed an auricular tachycardia, and digitalization was begun. Her further progress was entirely satisfactory, and she was discharged home on September 4. At this time a harsh systolic murmur could be heard in the left second interspace. When we last heard from her (Christmas, 1955) she was in very good health for the first time in her life (Fig. 1b).

CASE 2.—In C. M., a girl aged 5, a patent ductus arteriosus was detected at the infant welfare clinic when she was a year old. All her life the child had been susceptible to colds and respiratory infections.

She was admitted to the City General Hospital, Sheffield, on April 27, 1955. The heart was in sinus rhythm and blood pressure was 130/65 mm. Hg (Fig. 2). There was a continuous thrill over the pulmonary area, where a typical Gibson murmur could be heard. There was also a loud first sound and diastolic murmur at the apex. A provisional diagnosis of patent ductus arteriosus was made.

On May 3 a left postero-lateral thoracotomy was done under general anaesthesia (Dr. J. Johnston). A thick-walled ductus about 1 cm. long and 0.4 cm. across was doubly ligated with No. 6 silk (J. T. C.). Her blood pressure changed immediately from 110/60 to 110/90 mm. Hg.

The child’s immediate post-operative period was uneventful. Sixteen days after operation a pre-discharge E.S.R. was reported on as 55 mm. in one hour (micro method). A blood culture at this stage was negative. Twenty-four days after the ligation a chest radiograph showed a large round opacity pro-
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FIG. 2.—Case 2: Radiograph on admission to hospital for ligation of the ductus arteriosus.

jecting from the left hilum into the left upper zone, and it was queried whether this could be a localized collection of fluid (Fig. 3). Fluoroscopy revealed a circular mass just behind the pulmonary artery on the left side and expanding soon after cardiac contraction. The total and differential white cell counts were within normal limits. On auscultation a faint systolic murmur could be heard over the pulmonary area, with the child sitting up. During the 10 days over which the child was being observed the expansile mass was observed to increase rapidly (Fig. 4).

As we had already seen a similar case nearly a year before, a provisional diagnosis of aneurysm of the ductus arteriosus was made and an immediate left thoracotomy was done (J. T. C.) under general anaesthesia (Dr. B. H. Egerton and Dr. J. R. Munro) exactly a month after the original ligation. This revealed a large, thin-walled, pulsating mass 5 cm. across in the region of the ductus, infiltrating into the left upper lobe (Figs. 5, 6, and 7). The aortic arch and beginning of the descending aorta were mobilized and a Potts-Smith clamp applied across the aorta, occluding the aortic end of the ductus aneurysm. The pericardium was then opened and an attempt at control of the left pulmonary artery, as in the previous case, proved unsuccessful. At this stage the aneurysm tore off from the left pulmonary artery, leaving a hole 3–4 mm. across; it was also noticed that the Potts-Smith clamp across the aorta was not very effective at haemostasis. A decision to sacrifice the left lung was made, and, with finger pressure controlling the haemorrhage, a two-tourniquet pneumonectomy was done: it was, indeed, the quickest pneumonectomy we had ever done. The aneurysm came away with the lung, and now all we had to do was to close the two bleeding holes, but the field of view without the lung was excellent and the openings in the aorta and pulmonary artery were quickly repaired with interrupted arterial silk. Much of the pericardium was excised, as suture was found impossible. The chest was closed without drainage. The

FIG. 3.—Case 2: Radiographs 24 days after ligation of the ductus.
Fig. 4a.—Case 2: Radiographs 10 days after Fig. 3.

Fig. 5.—Case 2: Left lung with aneurysm of the ductus arteriosus. The pointer is in the upper lobe branch of the left pulmonary artery.

Fig. 6.—Case 2: View of the apex of the left lung, showing the extensive consolidation (due to the false aneurysm) in the apical segment.

Fig. 7.—Case 2: Macroscopic section of the outer wall of the false aneurysm of ductus, demonstrating the lamination in lung parenchyma.
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left phrenic and left vagus nerves with the recurrent laryngeal nerve were sacrificed during the operation, as they were all intimately incorporated in the wall of the aneurysm.

At the conclusion of the operation the patient's condition was excellent, with a pulse of 100 and blood pressure 125/80 mm. Hg. The patient had a transfusion of 9 pints of blood during the operation.

The post-operative period was very satisfactory. She was rather hoarse, but this improved gradually. On the sixth post-operative day 500 ml. of blood-stained fluid was aspirated from the left pneumonectomy space. During the first post-operative week she had penicillin and streptomycin and during the second week terramycin, 250 mg. six-hourly. She was discharged home a month after the second operation and is now back at school (March, 1956) leading a normal life (Fig. 8).

The left lung showed a circular laminated mass 5 cm. across, infiltrating the lung just above the hilum for a depth of 3 cm. Microscopy showed a dissecting aneurysm invading the lung, which over this area showed inflammatory changes. Fragments of elastic tissue were present in the inner layers.

DISCUSSION

Taussig, writing in 1947, mentions only four cases of aneurysms of the ductus arteriosus in the literature, and goes on to say that “an aneurysmal dilatation of the ductus arteriosus occurs only when the obliteration of the ductus arteriosus is complete at the pulmonary end but incomplete at its point of entrance into the aorta.” Yet the 56 cases studied had a true patency of the ductus unassociated with any anomalies of the aorta. Thoma (1890) and Simmonds (1906) drew attention to the ease with which aneurysms of the aorta at the site of insertion of the ductus could be confused with aneurysms of the ductus arteriosus itself.

ANATOMY.—In the ductus, in marked contrast to the aorta and the pulmonary artery, there are almost no elastic fibres in the media (Boyd, 1941). Hence the ductus is a purely “muscular” artery, while the aorta and pulmonary artery are “elastic” vessels. Jager and Wollenman (1942) demonstrated the condensation of the elastic fibres of the great vessels into a coarse elastic band at either orifice of the ductus, while a small portion coursed along the ductus as the internal elastic lamina: the external elastic lamina is absent and the adventitia itself is poorly delimitated from the outer media and contains collagen, elastic fibres, smooth muscle, and blood vessels.

INCIDENCE.—Ten of the 56 cases occurred as a result of operation on the ductus itself. Out of the remaining 46, only eight were seen in older children and adults, the oldest being 51 years old and the youngest 4 years old, the rest having been incidental findings in routine necropsies on babies a few days to several weeks old.

Forty-two of these aneurysms had been necropsy findings, where death had been due to other causes; unfortunately the details of the earlier cases are meagre. We feel that these aneurysms are often missed at routine necropsies. In a series of 1,400 consecutive necropsies on the newborn, one of our pathologists in the paediatric service, Dr. J. L. Emery (personal communication, 1956), was able to discover three aneurysms. Definite evidence of an infected ductus is available in eight cases, and the probability of infection in another five is high.

AETIOLOGY.—These aneurysms fall into the three groups congenital, infective, and traumatic.

Congenital.—Though the surgery of the patent ductus has been enthusiastically practised since 1938, we are aware of only two instances of a truly congenital aneurysm seen at surgical clinics: Potts (1955), while operating for a ductus on an 8-year-old girl, discovered one and divided it between clamps successfully; the other one was seen in a 22-year-old boxer by Dvořák and Schmittová (1953).

Infective.—The purely muscular ductus arteriosus lends itself to rapid aneurysmal dilatation as
a result of infective endarteritis from as diverse causes as umbilical sepsis (Kaufmann, 1929), infective endarteritis of the hypogastric arteries (Lennox and MacCarthy, 1951), and even infected haemothorax (Scheef, 1939). d'Abreu (1955) treated a girl who was diagnosed as “patent ductus arteriosus” at the age of 2 years but did not come into hospital until she was 12 years old, when she had a subacute bacterial endocarditis with a positive blood culture for *Strep. viridans*. Treatment with antibiotics for three weeks did little to control the fever. At operation a large mycotic aneurysm “exceeding the dimensions of the aorta itself” was found. Study of these cases raises the possibility that infection not only may cause an aneurysm of the ductus but can also help to close the ductus itself if the patient survives the infection. Hutchison (1922) reports the necropsy findings on a little girl, who died of infection where the aneurysm contained organized clot and recent obstruction of the pulmonary end of the ductus. Birrell (1954) reports the case of a 9-week-old infant who had a thrombosed aneurysm already containing much fibrous tissue. It seems possible that fibrosis and regression of the aneurysm can take place, leaving a cord which resembles the normal ligamentum arteriosum.

**Traumatic.**—The third causative factor is trauma and is the important one from the surgeon’s point of view. We found 10 such cases, including two of our own: eight of these were false aneurysms and only two were true aneurysms (Case 1; and Holman, Gerbode, and Purdy, 1953). Nine cases occurred after ligation (Jones, Dolley, and Bullock, 1940; Humphreys, 1942; Gebauer, 1943; Tubbs, 1944; Barrett, 1955; Lindskog and Liebow, 1953; Holman et al., 1953; Cases 1 and 2), and one after division (Crafoord, reported Ekström, 1952); three of these were infected from the onset (Crafoord, Tubbs, and Case 1), though five more became infected later.

Halsted and Reid (1916) have shown that the occlusion of a large artery by ligation in continuity results in an anaemic necrosis of the coarcted segment, and the ultimate result of the ligation is directly dependent upon the rapidity of necrosis of the segment ligated and the rapidity of substitution of this area by scar tissue. The wider the coarcted segment (and hence the thicker the ligation) and the lesser the mechanical damage or fracture of the arterial wall, the slower will be the necrosis and the chances of complete scar substitution will be the greatest. The real difficulty in the dissection of a ductus is encountered on its deeper aspect where dissection is done blindly and only too often with a blunt instrument. This is the level where the fibrous pericardium, after blending with the adventitia of the great vessels, is prolonged upwards and backwards to become continuous with the pretracheal fascia. Isolation of the ductus by sharp dissection is easy and safe (Potts, 1953), and it is much simplified if the perivascular sheath over the aorta is entered first and dissection continued towards the ductus, which in most cases can then be almost lifted off the fibrous pericardial bed; then the adventitia of the ductus itself is not injured. In at least three cases there had been considerable difficulty during this dissection at the first operation. When the adventitia is injured during a difficult dissection the main buttress of the ductus wall is lost. And when the insult of too tight an occluding ligature is added to this injury, unless rupture and catastrophic haemorrhage occur on the spot, the necrosis of the arterial segment is rapid, the ligature cuts through with ultimate restoration of the lumen, and a haematoma with a certain amount of healing on the outside is formed, sufficient to prevent any massive bleeding. This, we believe, is the sequence of events in the formation of a traumatic false aneurysm of the ductus, and is corroborated by the finding of ligatures still tied in the anterior wall or even in the interior of the aneurysm in three of these cases (Jones, Tubbs, and Barrett).

In Case 2 histology of the resected aneurysm showed fracture of the arterial wall and the haematoma dissecting out through the media.

Case 1 was a true aneurysm of the ductus; so also was Holman’s case (1953). Here the genesis was different. These were most probably instances of post-stenotic dilatation of the ductus, a condition well known to Halsted (1920), and much more recently worked out in detail by Holman himself (1954, 1955) in cases of partial occlusion of a vessel reducing the lumen to one-third or one-quarter of its original diameter. The spraying action of the jet of blood at each systole through the narrowed lumen converts the high kinetic energy of the aortic stream into potential energy or lateral pressure on the partially necrosed post-stenotic segment of the ductus; further, the eddy currents in this relatively stagnant area also contribute towards the bulge. Thus the relentless alternation of high and low pressure waves on the already weakened wall of the ductus leads to the formation of an aneurysm.

**Diagnosis.**—It is impossible for us to generalize as regards diagnosis when our personal experience extends to only two cases. But it is worth while remembering this possibility when one comes
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across a case of recanalization. In the case of traumatic aneurysms the signs are those of a re-established aortico-pulmonary fistula. There may be superadded infection also; eight of the 10 cases had an infected recanalized ductus. The only symptoms that may be attributed directly to an aneurysm are haemoptysis and hoarseness. Haemoptysis occurs when the aneurysm infiltrates the lung, but has been seen also in cases of aortico-bronchial fistulae (Jones, 1947). Hoarseness as a fresh symptom occurs when the left recurrent laryngeal nerve becomes involved in the tumour mass.

As in most cases of patent ductus, the evidence is mainly auscultatory. Gross and Longino (1951) in an analysis of 412 ductuses submitted to operation noticed complete disappearance of the murmur in 90% of cases. In the other 10%, or a total of 56 cases, the systolic element of the murmur persisted and was thought to be purely functional in 28 cases, while in the other 28 it was believed to arise from a second cardiovascular defect like septal defect, pulmonic stenosis, rheumatic mitral stenosis, and bicuspid aortic values. To this list we would like to add a grossly dilated pulmonary artery and acute rheumatic carditis. Recently we ligated an infected ductus in a girl of 10 years, whose pulmonary artery was four times the size of the aorta. Her infection was cured for the time being, but the systolic murmur in the pulmonic area persisted and at times a faint diastolic murmur could also be heard. We felt that the murmurs were due to the dilated pulmonary artery. She was submitted to surgery elsewhere, as the infective episodes recurred. It was found that the ductus had not recanalized and the pyrexia was due to an abscess around an aspirated tooth. A right lower lobectomy was done, and she has remained well since. Acute rheumatic carditis can occur at any time in a child who has had the ductus ligated; it is probable that cases with congenital cardiac disease are more susceptible. In the first attack of rheumatic carditis a systolic murmur and a diastolic murmur may be heard over the praecordium, a basal diastolic murmur due to relative incompetence at the aortic orifice, better heard over the second or third left interspace (Levine, 1951), and an apical systolic murmur, the main distinguishing feature here being the differential splitting as to maximal definition of each murmur over the different areas. We had a similar case where the fever, thought to be due to infection in a recanalized ductus, was controlled by salicylates alone, and cardiac catheterization studies proved that the ductus had not recanalized.

Radiology helps when an expanding and expansile mass is seen in the region of the mid-arc. In the presence of much laminated clot no expansile pulsations will be visible. Infiltration of the left lung field is positive evidence of aneurysm, and is seen mostly in cases of traumatic false aneurysm. On the other hand, perihilar pulsations alone may be present, and suggest a recanalized ductus only. It is possible that cardiac catheterization studies and left-sided cardio-angiography may be of great help.

In cases of congenital and infective aneurysms the clinical diagnosis will be one of patent ductus arteriosus, while the radiograph demonstrates an arched shadow in the region of the left hilum, projecting into the left lung field. This diagnostic feature was postulated by Hutchison and demonstrated by Scheef.

TREATMENT.—In the surgical treatment of these aneurysms the greatest operative hazard, as was amply proved in both our cases, is haemorrhage. At the very beginning we prefer to mobilize the aorta beyond the left subclavian artery with sacrifice of the uppermost set of intercostals, so that a side-occlusion clamp of the Potts type can be slipped across the aortic end of the ductus, or the aorta can be cross-clamped above and below the ductus level. Cross-clamping of the thoracic aorta under normothermic anaesthesia for any prolonged period is dangerous because of the risks of shock, renal failure, and spinal ischaemia and paraplegia (Pontius, Brockman, Hardy, Cooley, and De Bakey, 1954). Then we proceed to clamp the left pulmonary artery intrapericardially: this may not always prove possible, as the ductus may be attached at or close to the saddle of the main pulmonary trunk: then one has to face a steady bleeding from a fortunately low-pressure system before closure of the pulmonary opening. When the left lung is infiltrated, as it invariably is in cases of false aneurysm where dissection of the actual aneurysm will end in catastrophic haemorrhage, a simultaneous pneumonectomy, though drastic, may be necessary: a left upper lobectomy may be sufficient but may not be possible. A dissection-pneumonectomy is too time-consuming to be safe, whereas the two-tourniquet method is swift and safe, in no way enhancing the dangers of an already long and tedious operation.

CONCLUSION.—We are glad we had only two of these cases to treat, and it is impossible for us to assess their importance in the field of surgery of the ductus arteriosus. In conclusion, we can only quote from Maude Abbott's (1936) introduction to her atlas: "It is, however, a well-
recognized fact that in clinical medicine the intimate personal knowledge of a relatively small number of individual cases is likely to yield a richer harvest in the understanding of diseased conditions than wider generalizations covering a more vast material."

SUMMARY

Two cases of aneurysm of the ductus arteriosus, arising as a complication of the ligation-occlusion operation for patent ductus arteriosus, and successfully treated by resection of the aneurysm, are presented. The mode of causation, and the diagnostic features, the operative technique, and the hazards are briefly discussed.

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