MASSIVE ADRENAL HAEMORRHAGE IN THE NEWBORN

BY

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Intra-abdominal haemorrhage in the newborn is a diagnostic and therapeutic challenge; the site of the haemorrhage is commonly the liver or adrenal, rarely the other organs. (Lundquist, 1930; Potter, 1940; Henderson, 1941). If the haemorrhage is profuse it may lead to rupture of the capsule of the organ and intraperitoneal haemorrhage. In the case of adrenal haemorrhage in the newborn most authors have concentrated on post-mortem findings, many of the infants being stillborn. The first reported case to be successfully treated was that of Corcoran and Strauss (1924). Since then eight other surviving infants in whom adrenal haemorrhage was diagnosed have been described and these cases are tabulated (Table 1).

The following case report describes an infant suffering from haemorrhage into the adrenal with rupture of the capsule and intraperitoneal extension of the haemorrhage. From a review of the literature this would appear to be the first newborn infant to survive this complication. Twenty-five similar cases which ended fatally are tabulated for comparison (Table 2).

Case Report

The mother, weighing 18 st. 10 lb., was admitted to the Birmingham Maternity Hospital at the twenty-seventh week of her eleventh pregnancy because of hypertension. Her blood was Group O Rhesus negative and contained blocking antibodies. Two previous infants had haemolytic anaemia due to Rhesus incompatibility. The father was heterozygous for the Rhesus factor. The mother was in hospital a month, her blood pressure being 160/90, she was then discharged and re-admitted at the thirty-fourth week. Labour was induced surgically by rupture of the membranes at the thirty-seventh week, the indications being hypertension, oedema and the previous history of Rhesus incompatibility. The liquor was clear.

TABLE 1

SUMMARY OF RECORDED RECOVERIES FROM INTRAPERITONEAL HAEMORRHAGE

Author	Delivery	Sex	Clinical Features and Day of Onset	Treatment
Corcoran and Strauss, 1924	Precipitate	M.	4 day. Fever, vomiting, pallor. Palpable mass left side obstructing colon	Laparotomy and removal of blood clot from left adrenal
Goldzieher and Greenwald, 1928	Prolonged labour. Forceps. Primipara	F.	2 d. Fever, pallor, cyanosis, convulsions, temperature 103° F. Palpable mass right side	Transfused and given adrenal extract 4 and 12 d. Mass slowly disappeared
Arnold, 1930	Normal	M.	4 d. Pallor, listlessness, palpable right-sided mass	Transfused 120 ml. blood
	Podalic version Breech extraction	F.	3 d. Pallor, anorexia, pyrexia, abdominal distension 7 d. Convulsions, palpable right-sided mass	Transfused 95 ml. blood
Go dzieher and Gordon, 1932	Normal		7 hr. rapid respirations, shrill cry, cyanosis. Small abdominal mass. Blood sugar normal. Haemoglobin 70%	Intramuscular blood
	Normal		2 d. cyanosis, rapid respirations, abdominal distension. 3 d. fever. Right-sided abdominal mass.	Transfused 100 ml. blood
	Asphyxiated		3 d. rapid respirations Temperature 105° F. Palpable left-sided mass	Intramuscular blood
Emery and Zachary, 1952	Forceps	M.	2 d. fever, pallor, listlessness. Palpable mass right side	Laparotomy and removal of blood clot from right adrenal capsule
Marin *et al.*, 1955	Difficult breech		3 d. pallor, cyanosis. Palpable mass left side	Laparotomy. Nephrectomy and adrenalectomy, both organs surrounded by blood

TABLE 2

SUMMARY OF RECORDED FATAL CASES WITH INTRAPERITONEAL HAEMORRHAGE

Author	Delivery	Sex	Clinical Features and Day of Onset	Death (day)	Site of Haemorrhage
Riesman: quoted Hamill, 1901		F.		9	L. adrenal. Intraperitoneal rupture
Norris, 1900			Congenital heart disease	10	R. adrenal and intraperitoneal
Mattei, 1863				4	L. adrenal and intraperitoneal
Spencer, 1891	Stillborn	F.			R. and L. adrenal and intraperitoneal haemorrhage from left
	Breech. Stillborn. Hydrocephalic	F.			R. adrenal and intraperitoneal Rupture R. lobe of liver
	Mild hydrocephalic. Forceps delivery failed, Breech extraction	M.	Child only just alive when born	1	R. adrenal and intraperitoneal
Prudden, 1899	Normal		Breathed and fed badly	4	R. adrenal and intraperitoneal
Hodenpyl, 1890	Normal		3 d. Sudden collapse	3	R. adrenal and intraperitoneal. Subcapsular hepatic haemorrhage
Hervey, 1870			10 d. Sudden collapse	10	R. adrenal and intraperitoneal
Fiedler, 1870	Normal		4 d. Sudden abdominal distension, dyspnoea	4	R. adrenal 4-5 oz. intraperitoneal blood
Milroy, 1884	Rapid	M.	Pale, cold. Dyspnoea	12 hr.	R. adrenal and intraperitoneal
Tuley, 1892	Very short	M.	3 d. pyrexia. 4 d. respiration 72 laboured. Weak pulse 200. Painful expression. Tense abdomen	4	R. adrenal and 8½ oz. in peritoneal cavity
Hamill, 1901	Normal primipara	M.	2 d. temperature 103° F. Pallor. Restless, cyanosis, rapid respiration	3	R. adrenal and 6 oz. in peritoneal cavity
Lundquist, 1930	Breech extraction	M.	Shocked at birth. 5 d. convulsions. 6 d. mass left side	7	L. adrenal and intraperitoneal
	Extraction. Difficulty with shoulders	M.	Schultze resuscitation. Convulsions 2 d.	2	L. adrenal and intraperitoneal
	Normal primipara	M.	Asphyxiated. 6 d. pallor off feed. Resistance left side abdomen	6	R. adrenal and intraperitoneal
	Normal primipara	M.	4 d. pallor. Abdominal distension	4	R. adrenal and intraperitoneal
	Normal primipara	M.	5 d. pallor. Refused feed	5	R. adrenal and intraperitoneal
	Uterine inertia. Forceps extraction	M.	5 d. Sudden collapse	5	R. adrenal and intraperitoneal
	Transverse lie. Breech extraction	M.	Found dead	2	L. adrenal and intraperitoneal
Arnold, 1930	Normal primipara	M.	4 d. Pallor, air hunger. Convulsions abdominal distension	5	R. adrenal and intraperitoneal
	Podalic version. Breech extraction		3 d. Sudden collapse with abdominal distension. Petechiae	3	R. adrenal and intraperitoneal 200 ml. of blood
Goldzieher and Gordon, 1932	Normal		3 d. Cyanosis. Pallor. Rapid respirations. Fever 101·8° F. Purpura. Blood sugar 10 mg. at 88 hr. Bilateral abdominal masses	4	Bilateral, with 3 oz. of blood in peritoneal cavity
Kohlbry and Wells, 1945	Forceps		Pallor. Cyanosis. Air hunger. Vomiting. Abdominal distension	4½ hr. (Transfused)	R. adrenal and intraperitoneal
Emery and Zachary, 1952	Normal		Pallor, rapid collapse	2	R. adrenal and intraperitoneal

Three days later as labour had not started pitocin was given and labour commenced. After four hours 20 minutes a male infant weighing 7 lb. 14 oz. was delivered through a cervix which was not fully dilated. The multiparity and the obesity of the mother made controlled delivery difficult. The infant was a little pale and shocked at birth but was quickly resuscitated following clearing of the air passages. He was not jaundiced, the liver and spleen were not enlarged, the cord blood was Group A Rhesus negative, Coombs test negative, with a bilirubin of 0·8 mg./100 ml. Unfortunately clotting of the blood prevented estimation of the haemoglobin level.

The infant's condition gave rise to no concern until the morning of the third day when he was noticed to be pale. The haemoglobin was 12·1 g./100 ml. An hour later he became fretful and resented examination. His respirations became shallow and rapid, and cyanosis developed. The rectal temperature was 96·6° F. The differential diagnosis at this stage lay between an overwhelming infection, or an intracranial or intra-abdominal haemorrhage. Penicillin and streptomycin

were given. The child's condition further deteriorated, the pallor increased, the heart sounds became soft and a definite resistance to palpation was present on the right side of the abdomen. Within 15 minutes the infant's breathing ceased but the heart continued to beat. Meanwhile, a rapid transfusion of 80 ml. of blood was given through an umbilical catheter and cortisone 5 mg. and vitamin K 2·5 mg. were given intramuscularly; respirations recommenced and the colour improved. Two hours later breathing again became rapid and a further 5 mg. of cortisone was given intramuscularly. The child improved sufficiently to be transferred to the Children's Hospital. He was still pale, with peripheral cyanosis, though the haemoglobin was now 16·8 g./100 ml. The abdomen was tense, and again resistance was felt more over the right side. The respirations were rapid and shallow and during examination twitching of the face and arms was observed. An accurate diagnosis seemed essential for the carrying out of further treatment. Hepatic haemorrhage with rupture of the capsule was considered the most likely, but the convulsions pointed rather to an adrenal haemorrhage. It was therefore decided to do a laparotomy if abdominal paracentesis confirmed the presence of intraperitoneal blood. Blood was cross matched and an intravenous drip set up.

Abdominal paracentesis, one inch below the right costal margin in the anterior axillary line, produced 4 ml. of unclotted blood. The child was therefore anaesthetized and the peritoneal cavity opened by an incision parallel to the right costal margin. A large quantity of free blood and clots were removed from the peritoneal cavity. Inspection of the liver showed it to be intact. The right suprarenal capsule, however, was grossly distended by a blood clot and blood was present round the right kidney. The appearance of the kidney and adrenal closely resembled a tumour. The left suprarenal felt normal. After evacuation of the clot the adrenal capsule was packed with fibrin foam. No bleeding points were located and the abdomen was closed. The blood clot was later reported by Dr. A. H. Cameron to contain adrenal cortical tissue (Fig. 1).

The immediate post-operative condition was satisfactory. For two days the child remained on intravenous fluids to which hydrocortisone, 12·5 mg., was added each 24 hours. Thereafter oral feeds were tolerated. Cortisone in decreasing doses was given over a further five days. At no time did the child's condition or the serum electrolytes indicate adrenal insufficiency. The wound healed rather slowly but the infant's general condition remained excellent.

Discussion

Various factors seem to be concerned in the production of haemorrhage into the adrenal gland.

Trauma. Throughout the literature, trauma, at or soon after birth, is considered to be the main cause of intra-abdominal haemorrhage. In Lundquist's (1930) series of 18 adrenal haemorrhages only five deliveries were normal. Three of the infants had been delivered rapidly because of prolapse of the cord. Podalic version, forceps extraction and breech deliveries are all mentioned. Rogers (1934) in his paper on rupture of the liver describes three cases in which labour was induced medically with pitocin and it is interesting that this drug was used in our case. In 10 of the 25 fatal cases, labour was recorded as abnormal. In the reports of the infants who recovered details are fuller and seven of the 10 deliveries were abnormal. Birth weights are often above average (Henderson, 1941) and corresponding with this there is a preponderance of male infants.

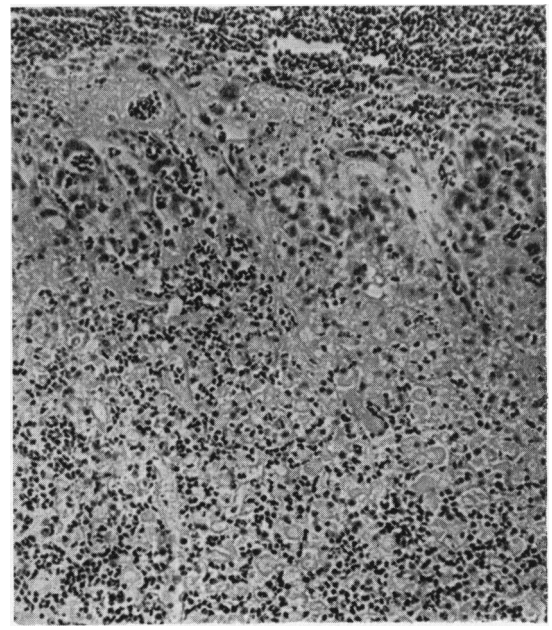

FIG. 1.—Adrenal haemorrhage showing 'permanent' cortical tissue between the external haematoma and the internal haemorrhagic 'foetal cortex'.

Asphyxia. Asphyxia probably plays a minor part in the causation of large haemorrhages though small bilateral adrenal haemorrhages, in themselves insufficient to cause death, are a common finding in asphyxiated infants. In such cases haemorrhages are usually found at other sites and these asphyxial haemorrhages are not a feature in the cases under discussion (Arnold, 1930; Henderson, 1941).

Coagulation Defect. A lowered prothrombin level would appear to be of considerable importance, for the time of onset of the symptoms of acute haemorrhage into the adrenals is the second to seventh day

which coincides with the age at which haemorrhagic disease of the newborn occurs (Arnold, 1930; Marin, Graham and Kickham, 1955). In the earlier writings, before hypoprothrombinaemia was known to occur, haemophilia is mentioned as an aetiological factor (Townsend, 1894; Morison, 1908).

Infection. Infection, which is the commonest cause of adrenal haemorrhage in the older child, is not often found in this group of infants.

Adrenal Vascularity and Involution. At birth the adrenals are relatively large having an average combined weight of 6·5 g. which is reduced by the age of 2 to 3 months to 3·51 g. (Tähkä, 1951). Direct trauma, unlikely to cause damage to the relatively much smaller gland of the older child, may well affect the larger gland of the newborn. The change in size is entirely due to the involution of the foetal cortex (Kern, 1911; Baar, 1954). During this process extreme hyperaemia with congestion and dilatation of capillaries occurs chiefly in the inner zone which comprises 80% of the foetal cortex. There is also a breaking down of the structure of this zone which may lessen any support given to the capillaries and make them more liable to rupture from increased congestion or external pressure. Areas of the outer, more compact layer of cortex may, as in our case, be found in the clot of blood suggesting that haemorrhage does indeed start in the inner hyperaemic layer (Hamill, 1901; Marin *et al.*, 1955).

Anatomy. In 18 of the 25 fatal cases the right gland alone was affected; in two the haemorrhage was bilateral and in five left sided only. In the surviving infants haemorrhage was considered, clinically or proved at laparotomy, to be on the right side in six out of the 10 cases. The explanation of this right-sided preponderance is considered to be the different venous drainage of the adrenals. The vein from the right gland drains directly into the inferior vena cava and is therefore more liable to be affected by pressure on the inferior vena cava during delivery or during resuscitation. The vein from the left adrenal drains into the smaller renal vein (Mattei, 1865). Magnus (1911) believes that the right adrenal gland, having the liver anteriorly and ribs posteriorly, is also more liable to direct compression.

Clinical Findings. The clinical findings are discussed fully by Goldzieher and Gordon (1932) in their paper on suprarenal haemorrhage. They divide the signs into those due to acute adrenal insufficiency on the one hand and acute haemorrhage on the other. The former consist of rapid respirations, pyrexia, purpura, metabolic upsets, convulsions and cyanosis, abdominal pain, vomiting and diarrhoea. The general effects of the latter are shock, collapse, weak pulse, cold extremities, pallor and air hunger, whilst the local effects are a distended abdomen and a palpable mass. It is not known whether there is any change in serum electrolytes in the acute phase suggesting adrenal insufficiency. A low blood sugar of 10 mg./100 ml. was recorded by Goldzieher and Gordon (1932) in one infant six hours before death from bilateral adrenal haemorrhage with intraperitoneal rupture. In practice awareness of the condition and recognition of the clinical picture has far more bearing on the immediate prognosis. Pyrexia and convulsions may be present but the important signs are, as in the case described, the sudden onset of a pneumonia-like illness with the addition of abdominal distension or a palpable abdominal mass.

Minor haemorrhages into the adrenals insufficient to cause clinical signs in the neonatal period may later present as calcification of the adrenals with or without signs of adrenal insufficiency. The finding of bilateral adrenal calcification following adrenal haemorrhage at birth has been reported by Snelling and Erb (1935). Gardner (1957) describes an infant with bilateral calcification presumably following birth trauma due to a footling presentation, who showed signs of adrenal insufficiency. Classical Addison's disease in children may be the result of haemorrhage into the adrenals in the newborn (Williams and Robinson, 1956). Larger haemorrhages especially when bilateral would certainly be expected to cause death from adrenal insufficiency and no surviving case has been found though many fatal cases with large bilateral suprarenal haemorrhages are recorded.

Where the haemorrhage is large but unilateral the clinical picture may be far less dramatic without definite signs of adrenal insufficiency or acute blood loss. In this instance the abdominal mass is the main finding. In the case reported by Corcoran and Strauss (1924) the distended adrenal had, by pressure on the descending colon, caused signs of intestinal obstruction. Marin *et al.* (1955) report the case of an infant who had a normal kidney and a haemorrhagic adrenal removed in the belief that it was a Wilms' tumour. The adrenal capsule was distended with old blood clots in which calcium was found. Only the cortex of the adrenal was intact. We have found an exactly similar case to this in the records of the Children's Hospital.

Where haemorrhage is extensive blood loss may be so acute that even when the diagnosis is correctly

made the infant dies before treatment can be instituted (Arnold, 1930).

Treatment. In infants with symptoms and signs suggestive of acute intra-abdominal haemorrhage replacement of the blood loss is the most urgent need. The umbilical vein is a convenient route for transfusion in the newborn. This is one of the few neonatal emergencies which call for rapid transfusion and amounts up to 100 or even 200 ml. of blood may be required. As much as $8\frac{1}{2}$ oz. of blood have been recovered from the peritoneal cavity (Tuley, 1892). Vitamin K should be given.

Whether the child should be subjected to laparotomy is debatable. It should not be necessary when the infant's condition improves rapidly after transfusion and remains satisfactory. In the surviving infants not operated on, the palpable abdominal masses have been noted to disappear. In suspected intraperitoneal haemorrhage the method of abdominal paracentesis strongly advocated by Rogers (1934) can be used to confirm the diagnosis. In cases where haemorrhage appears to be continuing, especially in those, the majority, where the exact site of haemorrhage is in doubt, laparotomy is advisable and may be life saving.

Adrenal extract was first used by Goldzeiher and Greenwald (1928) in the treatment of adrenal haemorrhage in the newborn. They believed that it was an important factor in the infant's survival. However, Arnold (1930) did not consider that any of the infants he describes died from adrenal insufficiency. We have no proof that our case was helped by steroid therapy but in the initial collapsed state, when the infant's condition was critical, and certainly after the confirmation of adrenal haemorrhage, its use seemed advisable. Now that steroid therapy is so readily available it is hoped, that with the recording of further survivals, its use can be better assessed.

Summary

A case surviving extensive haemorrhage into the right adrenal with rupture into the peritoneal cavity is described. Twenty-five similar but fatal cases are tabulated with nine other surviving cases who did not show intra-peritoneal haemorrhage.

The aetiology, clinical picture and treatment is discussed.

The need for urgent diagnosis and treatment if the child is to survive is stressed.

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