

because of relative hyperkalaemia and reduced cellular potassium content.⁴ Potassium loss from the atria has been suggested as an aetiological factor in atrial arrhythmias⁵ and may explain the development of atrial flutter in this patient. We suggest that the assessment of all infants born to mothers on lithium treatment during pregnancy should include an electrocardiogram.

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Received 21 January 1983

Lithium toxicity in a neonate

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SUMMARY Severe transplacental lithium toxicity in a neonate is described. There were gross functional lesions of the cardiovascular, renal, and neuromuscular systems with no structural abnormalities. At 1 year of age cardiovascular and renal function is normal, but there is developmental delay.

Case report

The baby's mother suffered from manic depression and had been treated with lithium for 7 years. Throughout this pregnancy (her fourth) the mother was maintained on lithium carbonate 1200 mg/day, chlorpromazine 50 mg 3 times per day and orphenadrine 50 mg 3 times per day. At 35 weeks' gestation she developed polyhydramnios. Two weeks later she developed signs of acute lithium toxicity—serum lithium 2.6 mmol (mEq)/l, sodium 133 mmol (mEq)/l, urea 11 mmol/l (66 mg/100 ml), creatinine 159 mmol/l (1.8 mg/100 ml)—and all drugs were stopped. The following day she went into labour and delivered a girl, weighing 2.78 kg.

The baby's Apgar score was 2 at 1 minute. Resuscitation resulted in spontaneous respiration by 15 minutes. At 3 hours of age a severe apnoeic episode occurred, necessitating continuous assisted ventilation and at this stage the infant was transferred to the regional cardiothoracic unit. On arrival she was shocked with an aortic blood pressure of 35/25 mm Hg and central venous pressure of 10 mm Hg.

Heart sounds were normal with a soft parasternal mid-systolic murmur. Hepatomegaly was present and generalised hypotonia was pronounced.

Radiography confirmed gross cardiomegaly with normal lung vascularity. Electrocardiography showed sinus bradycardia (105/minute), right atrial hypertrophy, and a prolonged Q-T interval and T wave inversion in the left chest leads. Cross sectional echocardiography showed a structurally normal heart with marked left and right atrial enlargement, a poorly functioning left ventricle, and increased left and right ventricular wall thickness. These echocardiographic features suggested a primary cardiac muscle dysfunction.

The results of biochemical investigations (Table 1) showed a high serum lithium value, hyponatraemia, uraemia, a high serum creatinine value, and hypocalcaemia. Acid base balance and cardiac enzymes were normal. There was thrombocytopenia (platelets $50 \times 10^5/l$) and prolonged clotting times (prothrombin time 34 s, kaolin cephalin time 78 s, thrombin time 31 s).

A cardiac inotrope—*isoprenaline*—was infused (6 $\mu g/hour$) and produced instant improvement of left ventricular function monitored by 2-D echocardiography. The cardiac output remained stable during *isoprenaline* treatment, which was continued for 54 hours. With improving cardiac output, murmurs suggestive of tricuspid and mitral regurgitation became apparent. Echocardiography showed persistent poor left ventricular function at

Table 1 Biochemical values during the first week of life

Time (days)	Sodium (mmol (mEq)/l)	Potassium (mmol (mEq)/l)	Chloride (mmol (mEq)/l)	Urea (mmol/l)	Creatinine (mmol/l)	Calcium (mmol/l)
1	128	4.6	97	15.4	231	1.45
2	123	4.6	87	13.0	222	1.47
3	125	3.5	82	8.8	158	1.30
4	124	3.4	90	4.8	102	1.86
5	113	3.9	81	3.5	75	1.41
6	132	3.6	104	1.7	63	1.78
7	114	4.7	85	2.1	65	1.62

Conversion: SI to traditional units—urea: 1 mmol/l \approx 0.17 mg/100 ml; creatinine: 1 mmol/l \approx 0.1 mg/100 ml; calcium: 1 mmol/l \approx 4 mg/100 ml.

Table 2 Relation between serum and urine osmolality, urine output per 24 hours and serum lithium concentration during the first 7 days of life

Time (days)	Lithium (mmol(MEq)/l)	Plasma osmolality (mmol/kg)	Urine osmolality (mmol/kg)	Urine output (per day)	Anti-diuretic hormone given (μ g)
1	2.8	—	—	250	—
2	2.6	266	170	225	—
3	1.5	274	181	345	10
4	1.0	263	123	111	1
5	0.6	232	308	720	10
6	0.3	267	305	366	2.5
7	0.2	239	83	747	—

day 13. On examination at 1 year of age cardiac function was normal.

A high urine output was apparent on day 1 and continued throughout the baby's stay in hospital (Table 2). A double volume exchange transfusion was performed on day 2 and during this the serum lithium value fell from 1.8 mmol (mEq)/l to 1.6 mmol (mEq)/l. (Total amount of lithium removed was 380 mmol.) Her urine output remained high, however, and despite apparently adequate fluid replacement she became dehydrated. Antidiuretic hormone (Desmopressin, DDAVP, Fleming) was given by nasal insufflation and urine output was controlled using a dose of 2.5 μ g every 4 hours. Plasma osmolality remained low until day 8. At 1 year of age renal function was normal.

Her serum calcium value was low on hospital admission and remained low despite intravenous supplements (Table 1). Serum magnesium concentration was normal (range 0.70–1.09 mmol/l (1.7–4.6 mg/100 ml)). Serum urea and creatinine values were high at first but rapidly became normal. Blood sugar remained normal. The serum bilirubin value rose to 230 mmol/l (1.3 mg/100 ml) on day 1 but fell rapidly with phototherapy. Abnormal clotting times at first were corrected with fresh frozen plasma and were normal thereafter. The early low platelet count rose spontaneously.

Profound hypotonia with depressed tendon reflexes was present until day 6. A paralytic ileus was present until day 12. At age 1 year there was evidence of delayed motor development and a concomitant squint.

Discussion

Most of this baby's problems can be directly attributed to lithium toxicity. Cardiac dysfunction was caused by poor myocardial contractility without structural abnormalities. The tricuspid and mitral valve regurgitation was secondary to papillary muscle involvement associated with uncoordinated ventricular contraction. Echocardiographic evidence of impaired ventricular function persisted for 13 days but had resolved almost entirely by day 19 despite the persistence of left ventricular hypertrophy. It has been suggested that lithium reduces cardiac contractility by interfering with the activation of adenylyl cyclase by hormones that normally increase myocardial contractility.¹ This was in an infant with severe myocardial dysfunction related to lithium toxicity. Our experience suggests that this may be at least partially reversible if adequate supportive treatment is given.

It is known that lithium may produce nephrogenic diabetes insipidus and also cranial diabetes insipidus.¹ The prompt response to treatment with anti-diuretic hormone suggests poor endogenous secretion despite the low plasma osmolality. It is likely that the high lithium values affected renal tubular function causing polyuria and renal sodium loss.

The association of neonatal lithium toxicity and hypotonia is well documented,^{2,3} and was severe in our patient. The neuromuscular effects are thought to be caused by alteration of neuronal ion transport, or inhibition of adenylyl cyclase mediated neurotransmitters, or direct interference with neuronal

carbohydrate metabolism.¹ The hypotonia persisted until lithium was undetectable in the serum. The apparently poor development at 1 year is disappointing, but it is not clear whether this can be attributed to the very high lithium values at birth or cerebral hypoxia associated with a poor cardiac output in the immediate postnatal period.

The management of neonatal lithium toxicity of this degree has not been described before. Exchange transfusion did little to reduce serum lithium values. Haemodialysis or peritoneal dialysis are the recommended treatments in adult lithium poisoning⁴ but, with all their attendant risks it is doubtful whether these are the best treatments in neonates. Our case shows that recovery in the acute phase of lithium toxicity can be expected provided adequate supportive treatment is given.

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Received 16 March 1983

'Windswept deformity'

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SUMMARY We describe 8 children with 'windswept deformity'—a valgus deformity of 1 knee in association with a varus deformity of the other. The disease is a physeal osteochondrosis and conservative treatment with serial corrective plaster casts is as effective as corrective osteotomies.

'Windswept deformity' is the abrupt development of angulation of 1 knee into varus deformity and the other knee into valgus (Fig. 1). Although the disease has received little attention in published reports, it is common in Nigeria and other parts of Africa.¹ We describe 8 children with this condition whom we have treated and discuss the aetiology, natural history, and management.

Clinical features

Relevant clinical, radiological, and laboratory data were obtained while treating 8 children who presented with 'windswept deformity' at this hospital in the first 3 months of 1982. In all cases early developmental milestones were reached and the deformity arose spontaneously shortly after the previously healthy child had begun to walk. Most patients were girls. The deformity was not preceded

by any local or systemic illness and there was no clinical or radiological evidence of rickets or of other generalised bone disorder. Serum calcium, phosphorus, and phosphatases were within normal limits. General metabolic disease, malnutrition, and haematological disorders were not found.

Radiographic appearances

The radiological features were characteristically osteochondritic and the lesions occurred in the distal end of the femur or the proximal end of the tibia, or both. The medial portion of the epiphysis in genu varum or the lateral in genu valgum became wedge shaped and more dense, while the adjoining metaphysis developed a beak like projection. Radiological examination showed 3 types: Blount's tibia vara² associated with physiological valgus deformity³ in the other knee; tibia valga with a physiological varus⁴ component (Fig. 2); and Blount's disease coexisting with tibia valga (Fig. 3).

Treatment

Our first 2 patients were treated by corrective osteotomies of the proximal tibia and the rest by serial corrective plaster of Paris knee cylinder casts.