During the 1960s I was particularly interested in the aetiology of congenital anomalies. I was able to show that they fell into two main groups: First, the malformations which arose during the period of organogenesis and were therefore teratological embryopathies and secondly, the deformations which arose during fetal life and were therefore alterations in a previously normally formed part of the body.

The majority of deformations arise in late pregnancy as the fetus becomes increasingly constrained within the uterus. Because of fetal plasticity and rapid growth any part of the fetal body may become moulded and deformed. Among these congenital postural deformities are plagiocephaly, congenital sternomastoid torticollis, congenital postural scoliosis, congenital dislocation of the hips (CDH), genu recurvatum and various forms of talipes. These postural deformities are particularly prone to arise when there is a lack of amniotic fluid. They are also more common in first pregnancies because of the unstretched state of the mother’s abdominal wall. Another factor is inability of the fetus to kick and change its position in utero either because of paralysis of the fetal legs or because they become trapped. This is likely to happen with breech presentation when the fetal buttocks lie deep within the mother’s pelvis and the legs are acutely flexed at the hips and extended at the knees (wrestlers call this immobilising position ‘the folding body press’). This position, known as the frank breech presentation, is particularly common in first pregnancies.

Observation of this characteristic scaphocephalic shaped skull warned the paediatric examiner at birth that the baby has been presenting by the breech and also that he should most carefully examine for congenital dislocation of the hips. This deformity was present in 14% of breech deliveries in a series I studied in the 1970s (Figures 2a and 2b).

**Sagittal craniostenosis following breech presentation: A congenital postural deformity**

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**Fig 1a**
Frank breech presentations near term showing scaphocephalic moulding of the skull in the fundi of the uteri.

**Fig 1b**
Figures 1a, 1b and 1c.

**Fig 1c**
Stillborn infant at post-mortem; bilateral renal agenesis. Note severe scaphocephaly; the infant also had bilateral congenital dislocation of the hips.

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**Fig 2(a)**
Frank breech presentation at term. Note oligohydramnios and acute flexion of the hips and extension of the knees.

**Fig 2(b)**
Stillborn infant at post-mortem; bilateral renal agenesis. Note severe scaphocephaly; the infant also had bilateral congenital dislocation of the hips.
CASE REPORTS

In April 1974 I encountered the following infant (Case 1). The mother, aged 36, was pregnant for the first time. The fetus presented as a frank breech at 39 weeks gestation and was delivered by Caesarean section. The baby girl weighed 2.96kg with a head circumference of 35cm and a length of 50cm. Apart from marked scaphocephaly no other abnormalities were detected at birth (Fig 3a & b).

However, at the age of 10 weeks, sagittal craniostenosis was suspected on clinical examination and confirmed radiologically (Fig 3c & d).

This was the first case of simple sagittal craniostenosis that I had personally diagnosed among the many thousands of infants that I had examined in the newborn period during the previous fifteen years. With hindsight, I appreciated that I had probably overlooked many such cases in the past and determined to be more careful in future. Indeed, in the next three years I encountered three cases similar to the one just described. All three had presented by the breech at term to mothers in their first pregnancies (Fig 4, 5 and 6).
In 1978 I gave a paper entitled: ‘Anomalous congenital anomalies’ at the 50th Anniversary of the founding of the British Paediatric Association held in York (7). Among the cases presented at that time were the four cases of craniostenosis described above. My suggestion was that steady compression in utero across a suture line might lead to fusion of the growing cranial plates and to craniostenosis. While my observations had at that time (1978) only applied to the sagittal sinus, it seemed probable that it might also apply to other cranial sutures.

Previously I had always regarded craniostenosis as a malformation. Certainly in a number of rare syndromes such as Crouzon’s or Apert’s, there is a heavy genetic component. Now, for the first time, it was being suggested that perhaps the majority of cases of craniosynostosis might be due to deforming forces in late pregnancy. In other words many cases might be considered as congenital postural deformities rather than malformations. Strong support for this suggestion came three years later from the distinguished dysmorphologist, David Smith of Seattle (8,9).

CONCLUSION

In summary, four cases of sagittal craniosynostosis were diagnosed soon after birth, within a four year period. All four babies had presented by the breech to mothers in their first pregnancies. It was suggested that constant pressure across the sagittal sinus in the fundus of the uterus had led to fusion and craniosynostosis. As a result, sagittal craniosynostosis should in future be classified as a congenital postural deformity.

REFERENCES