

Congenital anomalies

## Tongue tie

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### Common problem or old wives' tale?

The resurgence of interest in breast feeding has been accompanied by a lively debate about the significance of "tongue tie" or ankyloglossia. Symptoms attributed to tongue tie include nipple pain and trauma, difficulty in the baby attaching to the breast, frequent feeding, and uncoordinated sucking. These problems may result in the mother deciding to terminate breast feeding prematurely, slow weight gain for the baby, and even hypernatraemic dehydration. Speech defects have also been attributed to tongue tie. Strong views have been expressed by many eminent authors on the subject (box 1).

This paper reviews what is known about tongue movements and the significance and treatment of tongue tie. It is based on two literature reviews, one conducted on behalf of NICE<sup>1,2</sup> by one of us (MR) and updated by further searches of published and grey literature and conference abstracts. The publications reviewed for this paper are summarised in table 1.

As our review found little high quality objective evidence, we begin by making explicit the personal experience and bias with which we commenced the review. One of the authors (MR) felt that

tongue tie is an important issue—she experienced pain for many weeks while breast feeding her first child, who exhibited features said to be typical of tongue tie, and has since discussed this issue widely with lactation specialists and women having similar problems. The other (DH) accepted that ankyloglossia occurs in dysmorphic infants<sup>10</sup> and occasionally in otherwise normal babies,<sup>11</sup> but was sceptical about the high prevalence of the condition now being described by several authors.

#### ANATOMY AND PHYSIOLOGY

The tongue is a highly mobile organ made up of longitudinal, horizontal, vertical, and transverse intrinsic muscle bundles. The extrinsic muscles are the fan-like genioglossus which is inserted into the medial part of the tongue and the styloglossus and hyoglossus into the lateral portions. The sub-lingual frenulum is a fold of mucosa connecting the midline of the inferior surface of the tongue to the floor of the mouth. Tongue tie is the name given to the condition arising when the frenulum is unusually thick, tight, or short. There are many variations and differing degrees of severity (fig 1).

The movements of the tongue during infant feeding have been studied by cine-radiography and more recently by ultrasound.<sup>12,13</sup> Ultrasound reveals some similarities between the movements made by the baby when either breast or bottle feeding,<sup>14</sup> but also some important differences.<sup>15</sup> The tongue is projected further forward in breast feeding<sup>16</sup> and the human nipple elongates during each suck in a way that an artificial teat cannot do.<sup>14</sup> During feeding, the artificial teat, or the nipple together with some breast tissue, is held fully in the mouth with the tongue covering the lower gum ridge. The nipple is protected from damage and pain at the back of the baby's mouth.<sup>16</sup> The baby's lower jaw is then elevated, compressing the artificial teat, or the breast immediately behind the nipple, while the front of the tongue moves up to aid the expression of milk. In breast feeding, this is by compression of the milk ducts under the areola. A wave of upward movement of the medial part of the tongue progresses backwards, and the expression of the milk is further facilitated by negative pressure generated by downward movement of the back of the tongue and the lower jaw and, in breast feeding, by the active expulsion of milk once the let down occurs.

In coordinated feeding, the sucking, swallowing and breathing movements follow in a 1:1:1 sequence. This can take several days to become established in healthy full term infants. In pre-term infants and in some term infants a variety of poorly coordinated feeding movement patterns are observed and sometimes persist.<sup>17</sup> Antenatal

#### Box 1: Quotes from the past

"In observing a very large series of newborn babies, we have never seen a tongue that had to be clipped" (McEnery and Gaines, Chicago, 1940)

"While tongue tie is not nearly as common as members of the public believe, nevertheless a genuine case is occasionally seen and the condition is not entirely mythical although surrounded by an aura of superstition and old wives' tales" (Cullum, UK, 1959)

"Tongue tie...has been described as a myth of hoary antiquity...but it is probably wrong to suggest that it never causes symptoms. A case is reported in which a tight fraenum ruptured spontaneously during feeding...this baby remained a slow feeder and...[had not been] disabled by his tongue tie" (Smithells, London, 1959)

"Tongue tie is a rare but definite congenital deformity" (Browne, London, 1959)

"Tongue tie is a rare cause of dysarthria, though it is often blamed for slow speech development...most patients who have real limitation of movement as a result of tongue tie have a history of difficult milk feeding" (Ingram, Edinburgh, 1968)

"I have never seen feeding difficulties in the first year resulting from tongue tie and I doubt whether it is ever necessary to carry out an operation on it till the age of two or three...There are still doctors who cut the frenulum in the newborn period. This is always wrong" (Illingworth, Sheffield, 1982)

"Tongue tie where the tongue is forked can, very rarely, add to the baby's difficulties in taking the breast with poor protractility" (Gunther, UK, 1970)

"To some extent tongue tie is normal in every newborn baby and it should rarely interfere with either sucking or later speech development" (Davies *et al*, UK, 1972)

"True tongue tie is a very rare condition. This condition has been over-diagnosed in the past because of the failure to recognise that the frenum passing from the tongue to the floor of the mouth is normally short in the newborn...Only in infants with severe limitation of the tongue movement and inability to suck is division of the frenum indicated" (Turner, Douglas, and Cockburn, UK, 1988)

**Table 1** Tongue tie; review of literature

Author(s)	Number and age group studied	How cases were identified	Type of study and intervention	Results
Messner <i>et al</i> <sup>f</sup>	Examined 1041 newborns. Identified 50 TT cases (4.8%). M:F ratio 2.6:1 36 cases of TT enrolled and 36 controls without TT	Screened by one doctor, confirmed by one colleague	Observational follow up study but no intervention	30/36 TT cases and 33/36 controls breast fed to 2 months (p=0.29). 9 cases and 1 control experienced breast feeding difficulties
Hogan <i>et al</i> <sup>h</sup>	Examined 1866 babies. Identified 201 TT cases (10.7%). M:F ratio 1.6:1. 44% TT cases had problems feeding. 57 TT babies entered study (40 breast fed and 17 bottle fed)	Photos to assist staff in postnatal checks	Randomised to immediate frenulotomy or support by lactation counsellor, at mean age 20 days (3–70), median age 14 days	TT cases treated by frenulotomy; 27/28 marked improvement. Counsellor cases managed conservatively; 1/29 improved
Ballard <i>et al</i> <sup>f</sup>	Examined 2763 breast fed in-patient babies and 273 attenders at lactation clinic. M:F ratio 1.5:1. Identified TT in 3.2% in-patients and 12.8% clinic attenders	One observer examining all babies. ATLFF used	123 cases underwent frenulotomy at age 1–2 days	Latch improved in all, pain scores fell significantly
Ricke <i>et al</i> <sup>f</sup>	Examined 3490 babies, identified 148 TT cases (4.24%). M:F ratio 2.3:1. Enrolled 49 TT babies for study with 2 matched non-TT breast fed babies as controls	Nurses assisted by photos, ATLFF by team	Observational study, no intervention	Mothers of TT babies three times more likely to give up breast feeding by one week; however, 80% TT breast feeding well at one week. TT and non-TT breast feeding in equal numbers at 1 month. Mothers with TT babies reporting more pain at one month but not statistically significant. Small numbers so type II errors possible
Ramsay <sup>7</sup>	Case series	Referrals to paediatric surgeon	Measured nipple tip to hard soft palate junction by ultrasound, pre- and post-frenulotomy	Distance changed from 7.99 mm to 6.49 mm. Milk transfer increased from 3.3 to 7.2 ml/min. At least 7 day interval between frenulotomy and 2nd measurement
Messner and Lalakea <sup>g</sup>	Case series of speech problems: 30 children age 1–12	Measured tongue protrusion and inter-incisal distance	Frenulotomy	Speech improved. 25 mothers had tried to breast feed; 21 said no problems
Fernando <sup>o</sup>	Case series, n ≥200	Various; majority presenting with speech disorders	Frenulotomy	Improved to varying degree. 20% had history of BF problems: 80% did not

TT, tongue tie; ATLFF, Assessment Tool for Lingual Frenulum Function.

ultrasound studies show that mouth and tongue movements are already well developed in association with intra-uterine yawning and crying.<sup>18 19</sup>

## HYPOTHESES

Review of the literature and expert opinion gave rise to the following hypotheses:

- Tongue tie is a definable condition
- Tongue tie affects 3–4% of infants
- The tight frenulum prevents the infant from getting the tongue over the lower lip and gum ridge and therefore can cause feeding problems, particularly affecting breast feeding, leading to pain for the mother and poor infant weight gain; it can also affect bottle feeding
- The impact of a tight frenulum varies between mother–baby dyads
- A tight frenulum can also cause problems in older children and adults, involving speech, dental hygiene, licking ice cream, and French kissing

- Division of the frenulum (frenulotomy) is a low risk effective treatment
- The condition is genetic

## CASE DEFINITION

Can tongue tie be defined—and to what extent do individual observers agree on the diagnosis? The length of attachment of the frenulum varies widely. In some babies it extends to the tip of the tongue. There may be an indentation of the anterior edge, referred to as a heart shaped tongue. The appearance of the tongue is not sufficient on its own to make a diagnosis, as the thickness and elasticity of the frenulum also vary widely and affect the extent to which normal tongue movements are inhibited (see fig 1).

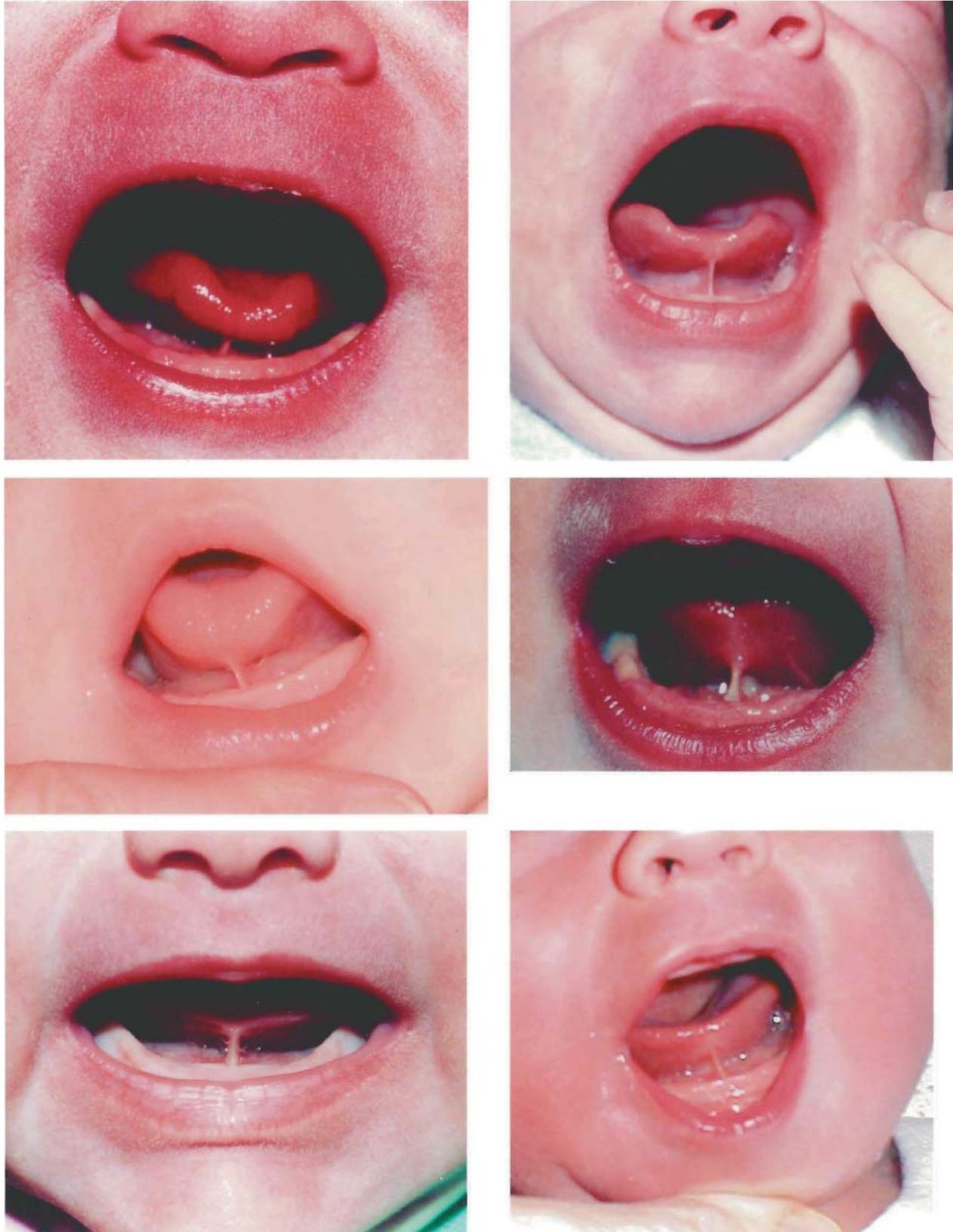
In four published studies of tongue tie in babies, the initial selection of possible cases from the whole newborn population was based only on appearance and was done by one individual (Ballard and colleagues<sup>5</sup>), or by one individual with confirmation of positive diagnoses by one other observer (Messner and colleagues<sup>3</sup>), or accomplished by providing staff with a

selection of photos (Hogan and colleagues,<sup>4</sup> Ricke and colleagues<sup>6</sup>). We have not found any formal data on observer agreement or variation in this process. The method of selection in the study by Masaiti and Kaempf<sup>20</sup> is unclear but cases were probably selected on the basis of breast feeding problems. Two authors (Messner,<sup>3</sup> Ricke<sup>6</sup>) aimed to reduce bias by trying to avoid specific mention of tongue tie to mothers but acknowledged that this was difficult.

All authors agreed that function is more important than appearance and Hazelbaker designed an Assessment Tool for Lingual Frenulum Function (ATLFF).<sup>21</sup> Ballard used this tool but did not examine inter-rater reliability. Ricke *et al* found that the inter-rater agreement using the ATLFF was only moderate and that many infants did not fit in any of the categories defined by Hazelbaker.

## BIRTH PREVALENCE OF TONGUE TIE

There is agreement among authors that tongue tie is found in around 3–4% of babies, with the exception of Hogan *et al*



**Figure 1** Six examples of babies diagnosed as having tongue tie, showing the variation in the thickness and insertion of the frenulum (reproduced with kind permission from Carolyn Westcott, Princess Anne Hospital, Southampton).

who report 10.7%. All report a male preponderance with ratios varying from 1.5:1 to 2.6:1.

### THE IMPACT OF TONGUE TIE

Maternal pain during feeding, sometimes accompanied by trauma, and difficulty in the baby taking the breast, are the main breast feeding problems attributed to tongue tie.<sup>3-6</sup> Attributing pain during breast feeding to tongue tie is not straightforward, however, since pain is a common problem that can result from several other causes, including attachment problems unconnected with tongue tie, and infection.<sup>16 22-24</sup> Ricke and colleagues<sup>6</sup> reported that more tongue tied infants than controls were bottle fed at one week but there was no significant difference at one month, though attrition meant that numbers were small. Messner *et al*<sup>3</sup> found no difference in the rate of breast feeding between tongue tied infants and controls at 2 months but a significant difference in the numbers of mothers reporting problems with breast feeding.

Ramsay<sup>7</sup> measured the distance from nipple tip to the junction of the hard and soft palate by sub-mental ultrasound. The distance decreased from 7.99 mm ( $\pm 2.80$ ) to 6.49 mm ( $\pm 1.87$ ) seven days after frenulotomy. This change, though statistically significant, is small and its practical significance is unknown. The tongue movements were said to become "more normal".

### VARIATION BETWEEN DYADS

Hogan and colleagues<sup>4</sup> found that more than half of babies with tongue tie had no problems breast feeding but could not show any correlation between severity of tongue tie and feeding difficulty. This is perhaps surprising, but it may be that only a small shift in positioning on the breast is sufficient to eliminate pain and improve feeding. Ricke and colleagues<sup>6</sup> reported that 80% of tongue tied infants were breast feeding successfully at one week. It is of interest that in two case series of older children presenting with speech difficulties and other problems attributed by the authors to tongue tie, 21/25 mothers (Messner and Lalakea<sup>8</sup>) and 80% of an unspecified number (Fernando<sup>9</sup>) who were asked about breast feeding reported no significant difficulties.

### OTHER PROBLEMS

Several case series report a range of other problems in older children associated with ankyloglossia—speech defects, difficulty in licking the lips or in kissing, dribbling, etc. These are difficult to evaluate as the authors do not give details of the catchment population, referral patterns, or detailed criteria for inclusion in the series.

### INTERVENTION

All authors agree that frenulotomy in the newborn is a low risk minor procedure, performed without anaesthetic. The presence of the deep lingual vein just lateral to the midline means that significant venous bleeding could occur if technique is not meticulous but we found no reports of serious adverse events. In older children the procedure needs an anaesthetic and sometimes a frenuloplasty, which carries some risk of scarring.

Ballard and colleagues<sup>5</sup> reported a marked fall in maternal pain scores after the procedure. Hogan and colleagues<sup>4</sup> randomised their cases to immediate or delayed intervention and found that frenulotomy was much more effective than advice from a lactation counsellor. They reported dramatic and rapid, often immediate, improvement after the procedure in most of their cases; improvement was noted in 95% of babies. The measurement of outcomes was not blinded. The precise criteria for improvement were not specified. Improvement was not always immediate, but this could be due to the need for sore nipples to heal or for the baby to re-learn optimal patterns of suckling.

In case series of older children and adults, some striking improvements were noted after surgery, but many of the children showed only gradual or modest improvement, particularly where the articulation of speech was concerned; these children often needed continuing speech therapy and this was attributed to the need to un-learn established patterns of articulation.<sup>9</sup> The absence of any comparison or control cases makes these reports impossible to evaluate and we found no comparative studies or randomised trials addressing the role of tongue tie or frenulotomy in older children.

### IS ANKYLOGLOSSIA INHERITED?

To define the inheritance of a condition, a robust case definition is needed, but tongue tie varies markedly in severity and is not an all-or-none condition. When an anomaly is identified in a newborn infant, the family searches its collective memory for other similar cases but, in the case of tongue tie, it would be impossible to assess the validity of that diagnosis in retrospect. None of the studies we reviewed considered these issues and none had gathered systematic family data across a number of families with and without the condition. Notwithstanding the comments made in several papers, no conclusions can currently be drawn about family history.

### AN OVERVIEW

Tongue tie is at first glance a minor issue, but from the results of the only randomised controlled trial yet conducted, Hogan and colleagues<sup>4</sup> suggest that at least 3% of newborns (57/1866) would benefit from frenulotomy and that this would increase the rate of continuing breast feeding. Most of the literature on tongue tie has been in connection with breast feeding; however, Hogan *et al* report that of the 57 babies in their study who benefited from frenulotomy, 40 were breast fed but 17 were artificially fed. If they are correct, this is a very common congenital anomaly that affects both breast fed and bottle fed babies, and up to 18 000 such procedures should be performed each year in the UK. It is therefore important to ask whether the evidence supports that rate of intervention and to scrutinise the evidence with a particularly critical eye.

There were a number of methodological problems with most of the studies we reviewed. These included:

- Inadequate assessment of inter-observer reliability of the initial diagnosis, the dynamic assessment of feeding and the maternal symptoms
- Ethical and practical difficulties in concealing the suspected diagnosis from the mother, thus potentially introducing a bias by raising the expectation of breast feeding problems and of improvement from intervention—this is, however, a common limitation in most studies of breast feeding problems
- Poorly defined outcome measures; it is particularly difficult to establish an objective assessment of improvement, when the primary outcome measure is reduction in maternal pain during breast feeding
- The dilemma of when to assess and intervene for tongue tie; if done very early, before breast feeding is established, as in the Ballard *et al* study, improvements may be wrongly attributed to the procedure (because suckling efficiency improves over the first few days and weeks<sup>13</sup>), but if done later (as in Hogan *et al*), many mothers may already have sore nipples or have given up breast feeding.

### CONCLUSIONS

We began this review by stating our personal bias. While DH confesses to still being somewhat more sceptical than MR, we are in complete agreement on the following conclusions:

- Individual case histories suggest that some babies do have a tight frenulum (tongue tie) which can inhibit breast

feeding and, in some cases, bottle feeding as well

- Although frenulotomy is a simple low risk procedure, it should be carried out only by those who have been trained in the procedure<sup>25</sup>
- It can be justified only if it is likely to lead to significant improvement in the comfort and the continuation of breast feeding, or of other longer term problems for the child
- We do not know the true prevalence of significant tongue tie
- There is no evidence one way or the other about inheritance
- On current evidence, there is no justification for actively searching for tongue tie during routine examination, but when mothers are having difficulty in breast feeding this should be considered as one of several possible causes
- The diagnosis should rest primarily on observation and analysis of feeding difficulties rather than the static appearance of the tongue
- It may be wise to be particularly cautious in making this diagnosis in the first two or three days before lactation is established
- The problem is of sufficient interest and importance to merit further studies of both breast and bottle fed babies, in which more precise case definition, measures of inter-observer reliability of pre- and post-intervention assessment, and ultrasound imaging are likely to play a key role
- Given the evidence that breast feeding has many advantages for both mother and baby, funding should be sought for carefully planned definitive studies on the issue.

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Parental consent was obtained for publication of the babies in figure 1

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Endocrinology

## Children with diabetes benefit from exercise

J I Wolfsdorf

Commentary on the paper by Massin *et al* (see page 1223)

How much physical activity do children require to obtain beneficial health and behavioural effects? The recent report concerning the effects of regular physical activity on health and behavioural outcomes in 6–18 year old youth recommends that

school age youth should participate daily in at least 60 minutes of moderate to vigorous physical activity that is developmentally appropriate, enjoyable, and involves a variety of activities.<sup>1</sup> There is strong evidence for beneficial effects of physical activity on: musculoskeletal and

cardiovascular health, adiposity in overweight youth, and blood pressure in mildly hypertensive adolescents. Physical activity also has a beneficial effect on anxiety, depression, and self-concept. The 60 minutes or more of physical activity can be achieved in a cumulative manner in school during physical education, recess, intramural sports, and before and after school programmes.

Exercise requires considerable alterations in fuel metabolism and presents unique challenges for the person with type 1 diabetes mellitus (T1D).<sup>2</sup> During the first 5–10 minutes of moderate intensity exercise, skeletal muscle glycogen is the major fuel for working muscle. With increasing duration of exercise, plasma glucose and non-esterified fatty acids (NEFA) predominate, and to meet the increased demand for

fuel, a complex hormonal and autonomic response increases hepatic glucose production and mobilisation of NEFA from adipose tissue. Plasma insulin concentration decreases and levels of the counter-regulatory hormones (adrenaline, noradrenaline, glucagon, cortisol, and growth hormone) increase, resulting in enhanced hepatic production of new glucose from gluconeogenic substrates such as lactate and glycerol. Large quantities of the glucose transporter protein GLUT4 are recruited to the membrane of contracting muscle, independently of insulin, increasing glucose transport into muscle.<sup>3</sup> These changes result in the increased fuel supply required to match glucose utilisation by exercising muscle and prevent hypoglycaemia. After prolonged exercise, liver and muscle glycogen stores are low and hepatic glucose production is accelerated. Resynthesis of muscle glycogen is, initially, largely a result of increased GLUT4 transporter activity and insulin sensitivity.

Glucose homeostasis, which depends on the balance between tissue glucose uptake and hepatic glucose release, is influenced by the plasma levels of insulin and counter-regulatory hormones. The normal regulation of insulin secretion is lost in T1D, and current methods of replacing insulin do not permit patients to mimic precisely the exquisite complexity of the normal physiological adaptations to exercise. Consequently, the child with T1D frequently experiences periods of either excessive or insufficient insulinaemia during exercise. When plasma insulin levels are relatively high, exercise causes blood glucose to decrease, whereas when insulin levels are low, and especially if diabetes is poorly controlled, vigorous exercise can aggravate hyperglycaemia and stimulate ketoacid production.<sup>4</sup> The child whose diabetes is out of control (marked hyperglycaemia with ketonuria) should not exercise until satisfactory glycaemic control has been restored.

Exercise acutely lowers the blood glucose concentration to an extent that depends on its intensity and duration and the concurrent level of insulinaemia.<sup>2</sup> In part, this results from accelerated insulin absorption from the injection site owing to increased regional blood flow and the massaging effect of contracting limb musculature.<sup>5</sup> If exercise is planned, the preceding insulin dose should be reduced by 10–20% and the injection given in a site least likely to be affected by exercise; for example, the anterior abdominal wall in the morning preceding a sports event. Because young children's physical activities tend to be spontaneous, this advice

is often difficult to implement consistently. Extra snacks (for example, 10–15 g carbohydrate per 30 minutes of vigorous physical activity depending on the child's age) before and, if the exercise is prolonged, during the activity are used to compensate for unplanned bursts of increased energy expenditure.

Exercise may be more predictable in older children and adolescents, and hypoglycaemia can usually be prevented by a combination of anticipatory reduction in the pre-exercise insulin dose or a temporary interruption of basal insulin infusion in patients who use continuous subcutaneous insulin infusion (CSII) together with supplemental carbohydrate before, during, and after physical activity. The optimal strategy in the individual child depends on the intensity and duration of the physical activity and its timing relative to the child's usual dietary and insulin regimen. After prolonged or strenuous exercise in the afternoon or evening, the pre-supper or bedtime dose of intermediate acting insulin should be reduced by 10–30% (or an equivalent temporary reduction in overnight basal insulin delivery in patients using CSII). To further reduce the risk of nocturnal or early morning hypoglycaemia caused by the lag effect of exercise,<sup>6,7</sup> the bedtime snack should be larger than usual and contain carbohydrate, protein, and fat. Frequent overnight blood glucose monitoring is essential until sufficient experience has been obtained to appropriately modify the evening dose of insulin after exercise.

In this issue, Massin *et al* report the results of an observational study of the amount and intensity of physical exercise, measured by 24 hour monitoring of heart rate, in preschool, school age children, and adolescents with T1D.<sup>8</sup> The structured diabetes education programme at the authors' centre includes the recommendation to obtain regular physical activity. The message is reinforced by encouraging regular physical exercise at clinic visits and by attendance at diabetes camp where children learn about the effects of different types of physical activity on glycaemia. The study involved a "snapshot" of the lives of these children on a single weekday, and it is possible that subjects knowing that their physical activity was being measured affected the results. The majority of children with diabetes receiving care at this centre met the paediatric recommendations for physical activity and compared favourably to their non-diabetic peers. The authors also observed a significant inverse association between mean annual glycated haemoglobin and the amount of time spent in light and moderate physical activity in school age children.

Despite its limitations, the study suggests that light and moderate physical activity may be associated with better glycaemic control in school age children, but not in teenagers. What might explain the difference between children and teenagers? Diabetes management is even more challenging during puberty, and glycaemia is typically less well controlled than before puberty and in adulthood. This is attributable to a combination of the endocrinological changes characteristic of puberty<sup>9</sup> and less meticulous adherence to diet and insulin administration.

Although physical exercise is complicated for the child with T1D by the need to prevent hypoglycaemia, with proper guidance and preparation, participation in exercise can and should be a safe and enjoyable experience. Despite the lack of compelling evidence that physical training and exercise per se improve glycaemic control in children and adolescents with T1D,<sup>10–16</sup> exercise clearly offers many health and psychological benefits for people with and without diabetes. At least 60 minutes of moderate to vigorous physical exercise daily should be a component of a comprehensive programme of diabetes management in children. With the increased prevalence of overweight and obesity in the population, children and adolescents with T1D may also be overweight or obese.<sup>17</sup> For these children, exercise is a critical component of a weight management strategy. Exercise ameliorates risk factors (obesity, hypertension, and hyperlipidaemia) for cardiovascular disease,<sup>18</sup> but equally important, children with diabetes are likely to benefit from the enjoyment and enhanced feeling of self-worth derived from participation in physical activity with their peers.

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## Management

# Inter-hospital transport for children and their parent(s)

R C Tasker

Commentary on the paper by Davies *et al* (see page 1270)

Each year, out of a child population of 10.5 million in England and Wales, approximately 10 000 need treatment in paediatric intensive care units (PICU).<sup>1</sup> Almost half of these children are transported between the referring hospital and their regional PICU by a specialist team; currently, the Department of Health recommends that parents should not routinely travel with their sick child in the ambulance.<sup>2</sup> So, should we be allowing parents to accompany their critically ill child during inter-hospital transport—or should they make their own way? In this issue, the PICU team from Guy's Hospital report their experience of having the child's parent accompany them during inter-hospital transport.<sup>3</sup> An emphatic "yes" comes from the South Thames Acute Retrieval Service (STARS) that covers the south of England: they still "continue to provide the service" and hope that their "results may inform other services that are considering adopting a similar policy".

In many respects it has been an error to have not considered, before now, the question of parents accompanying their critically ill child. Over 10 years ago the American Academy of Pediatrics stated: "it is sometimes beneficial when transporting the anxious and sick child to have a parent accompany him or her in the transport vehicle".<sup>4</sup> In our defence, we could cite certain hurdles to progress—concerns about accident insurance for passengers, shortage of space in the ambulance, and staff anxiety

because of the added burden of supporting relatives during transport.<sup>5</sup> The reality, however, is that the culture has evolved to exclude parents—we have streamlined the transport process and it avoids potential parental complications, by not having them there. The report by Davies and colleagues<sup>3</sup> reminds us that, like other areas in acute paediatric care, it is time to hear what parents feel and want, and now do something about it.

If we trace the pathway of care from acute presentation to later transfer to the PICU, we already know much about parents. First, in accident and emergency practice there has been growing interest in letting them stay by their child when procedures are performed, or at least giving them the choice about it. For example in the 1980s, Bauchner *et al* surveyed 253 parents and found that 78% would want to be present should their child need a blood test or insertion of an intravenous catheter.<sup>6</sup> In follow up studies, the same authors found, first, that parents chose to be present in 31 of 50 (62%) such procedures,<sup>7</sup> and second, as a consequence, they were less anxious and more satisfied with their child's care.<sup>8</sup> More recently, in a survey of 400 parents presented with five emergency department scenarios, Boie *et al* found that parents exhibited a hierarchy or order in their preference.<sup>9</sup> They were less inclined to be present with more invasive procedures, which, in decreasing order, were: venepuncture, 97.5%; suturing a laceration, 94%; lumbar puncture, 86.5%; resuscitation

of a conscious child, 80.7%; and resuscitation of an unconscious child, 71.4%. However, irrespective of these preferences, Boie *et al* found that if a child was likely to die, most parents would want to be present. Second, we know that there is likely to be a conceptual gap between what physicians think is appropriate for parents to see and what parents consider is their choice to decide. In the survey by Boie and colleagues,<sup>9</sup> only 6.5% of parents wanted the attending physician to determine their presence by their child. In a similar emergency department study, but this time surveying 645 emergency staff (306 physicians and 339 nurses) on views about six scenarios, Beckman *et al* found that almost half of the physicians believed that they alone (44%) should decide whether parents should be present.<sup>10</sup> This difference in viewpoint—between parents and physicians—is not altogether unexpected given the cultural history of our specialty: there was a time when parents were excluded from many aspects of hospital paediatric care (for example, bedside visiting for inpatients, peri-operative transfer between the ward and the operating theatre, induction of anaesthesia, etc). Now, child and parent centred care is essential to what we practice—that is, good medicine in the context of listening to patients' and parents' voices, openness, good communication, and developing a relationship based on trust. In essence, what we should learn from the studies reported by Boie and colleagues<sup>9</sup> and Beckman and colleagues<sup>10</sup> are the reasons underlying the gap between 6.5% and 44%, in parents and physicians respectively.

Third, in children who are critically ill, transport to a regional PICU is often the next step after presentation to the emergency room.<sup>11,12</sup> Patients may well have undergone resuscitation and there could be significant risk of adverse outcome. In 1995, Woodward and Fleegler (from the Transport Services of the Children's Hospital of Philadelphia) had a unique opportunity to survey two groups of parents: a group

who had been allowed to accompany their child in an ambulance because they had made a request to do so even though "this option was not actively encouraged" ( $n = 46$ ), and a group who did not accompany their child ( $n = 40$ ).<sup>13</sup> Eighty six per cent of these parents felt accompanying their child was important. Prior to transfer, 74% were either worried or very worried about their child's stability. Only 5% of parents accompanying their child found that doing so made them anxious or very anxious, whereas 56% of those who did not accompany their child felt anxious or very anxious ( $p < 0.05$ ). Finally, 94% of parents would choose to travel with their child in a future similar circumstance. These findings are not dissimilar to those found in the south of England in 2003. Colville *et al* surveyed 233 parents, of whom only 13 had been allowed to travel with their child in the ambulance: in total, 70% of these parents commented on the journey they made to the PICU, often travelling alone using their own transport.<sup>14</sup> A recurring theme in what the parents had to say was their sense of separation and the distress this journey caused them. Three quotations, relayed by the authors, capture the very essence of what these parents are feeling, and we should not forget this: "the worst journey of our lives", "the worst part was seeing the ambulance disappearing in the distance", and "for all I knew she was dying and I wasn't allowed to be with her". In this issue, the report by Davies and colleagues<sup>3</sup> focuses on logistics, staff perceptions, and adverse events—and that is entirely right given the newness of this service development in the south of England. However, another—albeit less emphasised—key message emerges from this work. The authors' interpretation of parental feedback (presumably compared with previous experience in the south of England<sup>13</sup>) was that these parents were less stressed as a consequence of accompanying their child. These audit data need to be followed up with further clinical studies. For example, do parents who travel with their children experience less acute stress disorder? Are they at less risk of post-traumatic stress disorder?

Davies *et al* tell us that their pilot study—permitting parents to accompany their critically ill child during inter-hospital transfer—was conducted in response to the distress expressed by parents, and now, because of their experience, it has become the established practice of the STARS team.<sup>3</sup> There are at least 14 other regional PICU transport services in England and Wales—how should we respond and move on? Should we determine whether there is national consensus regarding involving parents in inter-hospital transport—and only act if there is? The present standards of the United Kingdom Paediatric Intensive Care Society (1996 and 2001)<sup>12</sup> do not cover this issue. Rather, they state that it is the referring hospital that is "obligated to provide transport to the Lead Centre (that is, regional PICU) for parents". Even in the United States, there did not appear to be consensus among paediatric critical care transport team managers in 2001.<sup>15</sup> Only 63% of teams allowed parents to accompany their child in the ambulance, but the authors from Philadelphia (see also Woodward and Fleegler<sup>13</sup>) took the view that "awareness of this issue may help to establish discussion and guidelines regarding the role of parents". In other words, lack of unanimity should not detract from this potential development—it merely indicates the extent of change in practice that is needed if we are to respond to what parents need. There is now a literature showing that the most helpful coping strategy for parents of PICU patients is to allow them to stay with their child and to empower them.<sup>16–18</sup> Inter-hospital transport should be no different. However, if we cannot extrapolate the conclusion of the STARS service to our own regional practices, then we should at least look at alternative ways of better supporting parents during the time it takes for them to travel from the accident and emergency department to the regional PICU.

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## LETTER

### Adverse effects of rapid isotonic saline infusion

Neville *et al* reported on a randomised controlled trial of hypotonic versus isotonic saline for rehydration of children with gastroenteritis. They found that isotonic saline was superior with regards to correction of hyponatraemia.<sup>1</sup> The majority of patients in the study received a "rapid replacement protocol" which entailed the infusion of 40 ml/kg of isotonic saline over 4 hours in the isotonic saline arm of the study. The authors did not report on important known adverse effects associated with rapid infusion of isotonic saline which have been reported in previous randomised controlled trials of volume support with isotonic saline versus other fluids.

Rapid isotonic saline infusion predictably results in hyperchloraemic acidosis.<sup>2</sup> The acidosis is due to a reduction in the strong anion gap by an excessive rise in plasma chloride as well as excessive renal bicarbonate elimination.<sup>3</sup> In a randomised controlled trial with a mixed group of patients undergoing major surgery, isotonic saline infusion was compared to Hartmann's solution with 6% hetastarch and a balanced electrolyte and glucose solution. Two thirds of patients in the saline group but none in the balanced fluid group developed postoperative hyperchloraemic metabolic acidosis.<sup>3</sup> The hyperchloraemic acidosis was associated with reduced gastric mucosal perfusion on gastric tonometry.

Another double blind randomised controlled trial of isotonic saline versus lactated Ringer's in patients undergoing aortic reconstructive surgery confirmed this result; the acidosis required interventions like bicarbonate infusion and was associated with the application of more blood products.<sup>4</sup> Hyperchloraemia was found to have profound effects on eicosanoid release in renal tissue, leading to vasoconstriction and a reduction of the glomerular filtration rate.<sup>5</sup> The increased eicosanoid release may also explain the findings of reduced gastric perfusion in hyperchloraemia mentioned above.<sup>3</sup>

The main adverse effect of saline induced hyperchloraemic acidosis, however, may be the action which is taken to correct the abnormality. Acidosis is often seen as a reflection of poor organ perfusion and poor myocardial function, and a negative base excess may prompt the application of boluses of more saline containing fluids exacerbating the acidosis, the use of blood products, escalation of inotropic support and initiation of ventilatory support.<sup>6</sup>

The safety of hyperchloraemic acidosis has not been established in prospective studies and in patients with different types of critical illness. Particularly in critically ill patients with co-morbidities like renal disease, more physiological electrolyte solutions (e.g. Ringer's lactate solution) may be preferable to isotonic saline, and a slow replacement protocol safer than rapid infusions.

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## BOOK REVIEW

### Weight matters for children

Edited by Rachel Pryke. Oxon: Radcliffe Medical Press Ltd, 2006, £14.95 (US\$28 (approx.); €22 (approx.)), paperback, pp 215, ISBN 1857757718



It seems impossible to open a newspaper or turn on the television without the issue of childhood obesity being raised. The government has set targets to reduce the incidence of childhood obesity and school based programmes have been established, and yet the number of children who are obese continues to rise. What seems to be lacking and what this book sets out to provide is specific practical guidance for parents to follow as to what families need to be doing on a day to day basis to ensure children remain healthy and avoid becoming obese.

The primary focus is on parents and carers, and the author, who is a GP, frequently draws on her own experience as a mother to give examples of her own family life, which helps give credence to the messages which run throughout the book. The issue of choice is dealt with very well and discusses how important it is to give children choice and how parents can influence children to make the right choice. Parents are also encouraged to examine their own parenting styles to see how this influences the behaviour and eating habits of their children.

There are separate sections on preschool children and junior children, covering topics ranging from breast feeding and weaning to explanations about what constitutes a healthy diet. The advice about managing behavioural difficulties around mealtimes is particularly helpful, with sections such as "Tips to avoid becoming wound up at meal times" providing plenty of practical suggestions for families to try. There are also practical suggestions on encouraging physical activity in children and reducing television watching. The issue of dealing with a child who is already overweight is addressed, emphasising the need to take action early, placing this responsibility within the family context. There is also helpful information about understanding children's psychological wellbeing, examining the issue of low self esteem, bullying, and depression.

The final section covers nutrition and health problems. Basic nutritional information is provided to help make sense of the contents of our food and details what should constitute a healthy well balanced diet. There is a well written section on a wide variety of medical problems encountered which may affect a child's growth, ranging from cows' milk allergy to cystic fibrosis, and sources of further information are well referenced.

Overall I felt that this was a well written book, packed full with helpful practical suggestions. As a paediatrician, the advice contained within the chapters covered many of the food related problems seen in clinic. As a parent I also recognised many of the scenarios and remembered many meal times with young toddlers which were far from enjoyable! My one criticism of the book would be the general layout as I felt this was more in keeping with a medical textbook rather than a manual for parents. I'm not sure whether first impressions of the book would encourage parents to pick it up. However, this certainly won't deter me from recommending this book. I do feel this will be of benefit to all paediatricians to read as well as GPs, health visitors, school nurses, and of course also parents, especially those dealing with truculent toddlers!

**C Grayson**

## CORRECTIONS

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Principi N, Esposito S, Gasparini R, Marchisio P, Crovari P, for the Flu-Flu Study Group. Burden of influenza in healthy children and their households. *Arch Dis Child* 2004;**89**:1002–7.

This article has been retracted by the publisher because of significant overlap with Principi N, Esposito S, Marchisio P, Gasparini R, Crovari P. Socioeconomic impact of influenza on healthy children and their families. *Pediatr Infect Dis J* 2003;**22**(Suppl 10):S207–10.

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