LETTERS TO THE EDITOR

The Family Fund database - an underused research resource

EDITOR,—I wish to bring to the attention of paediatricians, and other health and social workers interested in childhood disability, the existence of a unique source of information with great research potential.

The Family Fund (now known as the Family Fund Trust for Families with Severely Disabled Children) was established in 1973 to provide grants to families with one or more children whose disabilities cause very severe handicap. Funding is provided by the Department of Health. Until 1995 the administration of the fund was under the direction of the Joseph Rowntree Foundation, but it is now a free standing registered charity. The fund is available to families with disabled children up to the age of 16 years who are resident in the UK (England, Wales, Scotland, and Northern Ireland). Applications can be initiated by parents as well as by health and social work professionals.

The qualifying medical condition is the degree of disability rather than the specific medical diagnosis. In order to make the best use of the funds available, grants are not normally available to families that can afford the relevant costs. Although the award of a grant requires that the medical and economic criteria are both satisfied, and that the items requested are appropriate, *all* children whose families apply for help are included in the computerised records. The database is in no sense a comprehensive register of all disabled children in the UK, nor is it necessarily representative of them. It has been estimated that 50–75% of eligible families apply to the fund for help. Selection bias arises principally from:

- Local awareness of the existence of the fund. However, at regional level, application rates tend to reflect known differences in the prevalence of disability.
- Professional awareness of the Family Fund criteria for making grants. Medical and social work staff who are experienced in the use of the Family Fund will not waste time on fruitless applications.
- The fund restricts its grants to families whose gross income is below the national average wage. Although applications are sought as widely as possible, this restriction tends to be known by medical and social work staff making referrals or recommendations to the fund.

The fund's register holds data on over 150 000 applicants. New applications are now approaching 10 000 annually. It is therefore self evident that the main strength of the database is its sheer size, which is reflected by very large numbers of children with common disorders and substantial numbers of relatively rare conditions. For example, data are held on approximately 30 000 cases of non-specific mental handicap (learning difficulties), 20 000 cases of cerebral palsy, 12 000 cases of Down's syndrome, 7000 cases of deafness, 4000 cases of childhood malignancy, 2000 cases of metabolic disease (other than diabetes), 2000 cases of cystic fibrosis, 1000 cases of disabilities caused by head injury, 700 cases of juvenile chronic arthritis, and 500 cases of brittle bone disease.

For the large majority of cases there is a detailed social work report. The main weakness of the database for medical research purposes is that doctors' reports exist for only a minority of children. For reasons of confidentiality, names and addresses cannot be given to researchers without permission of the parents concerned. However, Family Fund staff are prepared to approach parents on behalf of researchers to request permission for medical records to be studied, and experience has shown that the great majority of parents are only too happy to help in this way.

For further information about using the Family Fund as a research resource, please contact: Dot Lawton (Research Fellow), Social Policy Research Unit, University of York, Heslington, York YO1 5DD; tel: 01904-433608, fax: 01904-433618.

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Cellular profile of bronchoalveolar lavage fluid in pulmonary tuberculosis

EDITOR,—Bronchoalveolar lavage (BAL) has been used to study the immunopathogenesis of several respiratory diseases. The aim of our study was to determine the inflammatory changes occurring at the site of a tuberculous lesion in the lung in children.

Eighteen children (mean (SD) age 5·1 (3·2) years, range 1·5–12 years) attending the tuberculosis clinic at the Institute of Child Health and Hospital for Children, Madras who had signs and symptoms suggestive of pulmonary tuberculosis with a persistent radiographic abnormality had early morning gastric lavage and bronchoscopy and BAL done under local anaesthesia. The bronchoscope (3·5 mm Pentax) was wedged into the involved segment and BAL was performed by instilling 2 ml/kg sterile saline in three equal aliquots. There were no complications encountered and informed consent was obtained from the parents. The study was approved by the institutional ethics committee.

The total cell count showed wide variation between cases and the mean (SD) total count (74 (45)$\times 10^6$/100 ml) was not significantly different from reference values (80 (84)$\times 10^6$/100 ml).[1] The percentage of macrophages was significantly reduced in those with tuberculosis compared with reference values (56 (25)% *v* 84 (8)%, $p<0{\cdot}01$). The BAL fluid from cases of tuberculosis compared with reference values had a greater mean number of lymphocytes (22 (17) *v* 10 (6)$\times 10^6$/100 ml, $p=0{\cdot}02$) and eosinophils (10 (17·5) *v* 0$\times 10^6$/100 ml, $p<0{\cdot}001$). Bronchial epithelial cells accounted for 6 (4·5)% of total cells. Due to the obvious ethical limitation of enrolling controls, we have used the BAL data on healthy children of Ronchetti *et al* for comparison.[1]

Several studies looking at BAL cellular profile in adults with pulmonary tuberculosis have demonstrated increased lymphocyte counts.[2 3] Nowakowski *et al* found that the CD4/CD8 lymphocyte ratio was decreased in BAL fluid but increased in blood in children with pulmonary tuberculosis.[4] In this study, we have found a relative lymphocytosis and eosinophilia at the site of the lesion in children with pulmonary tuberculosis. There was a proportionate decrease in the percentage of alveolar macrophages in BAL fluid. More research into the local immunopathology of tuberculosis is required.

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1 Ronchetti R, Villani A, Dotta A, *et al*. Pediatric BAL - reference values. *Am Rev Respir Dis* 1992; **145:** A556.
2 Ainslie GM, Solomon JA, Bateman ED. Lymphocyte and lymphocyte subset numbers in blood and in bronchoalveolar lavage and pleural fluid in various forms of human pulmonary tuberculosis at presentation and during recovery. *Thorax* 1992; **47:** 513–8.
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Screening for biliary atresia

EDITOR,—Having discussed this issue with colleagues several times over the past year without a clear consensus emerging I welcomed the recent article by Mowat *et al*.[1] Obviously nobody would seriously argue with the proposition that it is beneficial if children with biliary atresia are identified and treated early, certainly by 40 days after delivery. However there are a number of questions that need to be answered. How good is the screening test proposed in detecting cases of biliary atresia at 2, 3, and 4 weeks and what percentage of the normal population are still slightly jaundiced at these ages? Every paediatrician seeing small infants will frequently see infants of 2 weeks of age who are still slightly yellow, thriving, often breast fed, with normal yellow breast milk stools.

Probably equally difficult for me, in the case being advanced for screening, was the cost benefit analysis attempted. While allowing for a certain amount of advocacy can you seriously introduce a national screening programme involving someone at least looking or checking on the degree of yellowness of every infant at two weeks after delivery and in addition obtain a urine sample from those thought to be still jaundiced, without any additional personnel? I seriously doubt it. The logistics involved in effectively screening a population are considerable and well documented. I enjoyed the two five minute phone calls to track results and don't believe any laboratory test costs £3.50 (for direct bilirubin) in the real world where heating, lighting, insurance and salaries, travel expenses, etc have to be

paid. The cost savings from non-transplanted livers were equally impressive even at the discounted price of £30,000 per transplant. Are we to believe that these spare livers would not be used for some equally deserving cases thus resulting in no net saving to the health service? As a paediatrician I remain unconvinced by the arguments advanced that a national screening programme at two weeks after delivery will solve this clinical dilemma.

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1 Mowat AP, Davidson LL, Dick MC. Earlier identification of biliary atresia and hepatobiliary disease: selective screening in the third week of life. *Arch Dis Child* 1995; **72:** 90–2.

Professor Mowat and Dr Dick comment:
We are please to have Professor Matthew's support in trying to achieve surgical treatment for all infants with biliary atresia by 60 days of age. Because we share some of the concerns he expresses, we do not advocate screening for biliary atresia but selective screening or more correctly case finding by detecting conjugated hyperbilirubinaemia in jaundiced infants to detect all forms of hepatobiliary disease. Most will have other hepatobiliary disorders for which early and specific treatment is desirable. By screening at the same time as the infant is being assessed by community health care professionals much of the cost and logistic difficulties will be minimised.

King's Healthcare Trust is undoubtedly in the real world. Next year the cost for a direct bilirubin will increase to £4.00 including all overheads! Since submitting our paper an infant aged 18 days with biliary atresia was 'overlooked' by a member of our junior staff. The total serum bilirubin concentration was 72 μmol/l. We cannot stress too strongly the infant with biliary atresia in the first weeks of life appears well. The only constant abnormal clinical feature is jaundice which may be very mild and urine which is persistently yellow and never colourless. In the last two years 25 infants and children in UK died while on waiting lists for liver transplantation. If any of these were alive because a selective screening made transplantation unnecessary for one child with biliary atresia, would any paediatrician object?

Because the optimum time for screening is controversial, community staff in our district are testing for conjugated hyperbilirubinaemia in jaundiced infants of different ethnic backgrounds. This study funded by the Children's Liver Disease Foundation will clarify logistical difficulties and the prevalence of benign jaundice in the third and fourth week after birth.

Double blind placebo controlled trial of pizotifen syrup in the treatment of abdominal migraine

EDITOR,—Now and then the concept 'abdominal migraine' appears in the literature as if it were a fact. I have always been reluctant to accept it as a special entity. The only thing that distinguishes it from recurrent abdominal pain in Apley's definition is the exclusion of the milder cases.[1 2] The demonstration of a special visual evoked response pattern in children with migraine and abdominal migraine is of course interesting.[3] But it is necessary to do this test in an *unselected* group of children with recurrent abdominal pain, to see if it delimits a special group among these children, or if it is a common phenomenon in children with recurrent abdominal pain. Even if it should delimit a special group it might just be a question of severity.

I am not able to refute the existence of abdominal migraine. But until now nothing except severity seems to justify the concept. Migraine in a close family member is a prerequisite for the diagnosis abdominal migraine.[2] But not even this criterion seems to be of any help, as accumulation of several kinds of presumed psychosomatic symptoms including headache is very common in children with recurrent abdominal pain and in their families.[4] I would still prefer the expression recurrent abdominal pain for all bellyachers, at least until we know more about aetiology and pathogenesis.

These reflections should be seen as a comment on the paper of Symon and Russell showing effect of pizotifen in children with abdominal migraine.[5] It is of course important to show that pizotifen does work. But the paper gives rise to two important questions. How does pizotifen work on all children with recurrent abdominal pain? And does the effect of pizotifen in a group of children with severe pain justify the migraine diagnosis?

Aetiology of recurrent abdominal pain is not known with certainty, but it is likely that psychosomatic mechanisms are operative. In the complex pathogenesis different peptides and motility may be important factors.[6] It is in this context that the effect of pizotifen should be considered.

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1 Apley J. *The child with abdominal pains.* 2nd Ed. Oxford: Blackwell, 1975.
2 Symon DNK, Russell G. Abdominal migraine: a childhood syndrome defined. *Cephalalgia* 1986; **6:** 223–8.
3 Mortimer MJ, Good PA, Marsters JB, Addy DP. Visual evoked responses in children with migraine: a diagnostic test. *Lancet* 1990; i: 75–7.
4 Christensen MF, Holm E, Sahlholdt I. Recidiverende mavesmerter hos danske skolebørn. *Ugeskr Laeger* 1984; **146:** 2690–5.
5 Symon DNK, Russell G. Double blind placebo controlled trial of pizotifen syrup in the treatment of abdominal migraine. *Arch Dis Child* 1995; **72:** 48–50.
6 Lindberg T. Recurrent abdominal pain in childhood. *Acta Paediatr* 1994; **83:** 775–6.

Dr Symon and Dr Russell comment:
Recurrent abdominal pain is a symptom and not a diagnosis. We find no difficulty in accepting that children with recurrent headaches may be suffering from a wide variety of different diseases, including migraine, tension headaches, and even cerebral tumours. Similarly recurrent abdominal pain may be the final symptom of a wide variety of disease processes. In our practice the commonest cause of recurrent abdominal pain is constipation. The concept that all recurrent abdominal pain is psychosomatic in origin has been discredited by the absence of any statistically significant differences between children with recurrent abdominal pain and pain free children with regard to various psychological variables thought to be associated with psychogenicity.[1]

The children whom we treated in our trial were not 'bellyachers' but were suffering from recurrent severe disabling symptoms. Unlike bellyachers their symptoms came in discrete attacks with complete normality between episodes. We accept that the term 'abdominal migraine' is not universally accepted and the arguments for this were fully rehearsed in a recent clinical controversies article.[2] Perhaps there would be fewer objections if the syndrome had a different eponymous name such as Buchanan's syndrome,[3] as some people wish to reserve the term migraine solely for headaches on the basis of its presumed etymological derivation from hemicrania.

We would not expect pizotifen to be of benefit in all children with recurrent abdominal pain and logically we feel that it is unlikely that pizotifen would be of value in recurrent abdominal pain other than abdominal migraine. We are not aware of any trials of the use of pizotifen in recurrent abdominal pain other than our own trial in abdominal migraine.

To lump together all children with recurrent abdominal pain as having psychosomatic pathology is to do grave disservice to those patients who come to us seeking relief of their symptoms.

1 McGrath PJ, Goodman JT, Firestone P, Shipman R, Peters S. Recurrent abdominal pain: a psychogenic disorder? *Arch Dis Child* 1983; **58:** 888–90.
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3 Buchanan JA. The abdominal crises of migraine. *J Nerv Ment Dis* 1921; **54:** 406–12.

Medicalisation of the normal variant – treatment of the short, sexually immature adolescent boy

EDITOR,—I enjoyed Christopher Kelnar's annotation but as a non-endocrinologist am unhappy about his advice for delayed puberty in the absence of disease that 'boys over 14 years of age ... who have impaired self image and social withdrawal not responding to reassurance' should be considered for treatment which 'should not be denied when appropriate'.[1]

There are two issues. Firstly the widespread use of potent endocrine agents for a self limiting condition. Can we really be sure that there will be no long term adverse effects during the lifetime of the individuals concerned or, indeed, of their progeny? 'Patients need to know whether they want to take the risks and doctors need to be accountable', states Brendon Nelson, the president of the Australian Medical Association, in considering the unexpected long term consequences of another endocrine intervention, Creutzfeldt-Jakob Disease.[2] The prospect of permanent gross dwarfism probably, even in retrospect, justified the, at the time unpredictable and thus unquantifiable, long term risk. Does the transient and common phenomenon of delayed puberty? We must surely include permanence as well as severity and incidence in any therapeutic cost benefit analysis.

Secondly, and more importantly, we need to be careful, as paediatricians, not to narrow the range of accepted normality and to medicalise normal variation. A teenager with delayed puberty may have impaired self image and social withdrawal at the age of 15. Where is the evidence that short term manipulation of the situation with drugs is of long term benefit to the psychological health of the future man, quite apart from its implications