RESEARCH DESIGN

Lessons from psychosocial studies of chronic renal failure

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Medical advances have resulted in considerable improvements in life expectancy and in associated morbidity for many childhood chronic conditions, but what is the cost in terms of the quality of life for the child and family and in the child's psychiatric adjustment? In many of these illnesses (for example renal failure and cystic fibrosis) treatment only ameliorates the illness. In others (for example cancer) although 'cure' of the primary disease is possible the risks of second malignancies, late recurrence, and late side effects of treatment remain. Thus even successful treatment, whether it is curative or palliative, has its burdens. There is an obligation to identify the burdens of the condition and its treatment so that all available social and psychological supports and treatments may be provided and appropriately targeted. Problems may arise that affect the child or family. They vary in complexity ranging from relatively simple problems such as coping with specific procedures and family stress to more complex problems such as the recognition of psychiatric conditions whose presentation is modified by the physical illness or non-adherence that might crucially affect the outcome. Increasing attention has been directed to this area in recent years.¹⁻⁴ There are a number of problems in designing studies in this area. This review discusses some of the potential pitfalls and possible solutions in the context of studies of the psychosocial consequences of renal failure in children.

A further purpose of this report is to emphasise the collaborative nature of such research. Such cooperation in research studies is facilitated by close liaison between the paediatric medical team and mental health professionals in day to day practice. This allows the training and expertise in research methodology, which is a strength of psychiatric personnel, to be integrated with paediatric clinical expertise.

Review of the Manchester studies

A number of studies of the psychosocial consequences of renal failure have been undertaken in this unit. The purpose of this brief review is not to attempt a comprehensive overview of psychosocial research in renal failure in chil-

dren but rather to set the scene for the subsequent discussion of methodological issues.

An initial study compared children on hospital haemodialysis in end stage renal failure (ESRF) with two other groups of children: children with renal failure not yet requiring ESRF management and a group of healthy children matched for age, sex, and socioeconomic characteristics.⁵⁻⁸ Subsequently a number of these assessments were repeated on patients after transplantation and thus at a further different stage of illness.⁹ The long term outcome is obviously of great importance for childhood illnesses and further studies assessed the effects of ESRF management in childhood upon function in adulthood by using a matched group of healthy young adults.^{10,11} Referral patterns of children with renal failure will clearly have a major impact on the outcome of studies such as these. We have previously reported details of the ESRF programme for children in Manchester where these studies were conducted.¹²

These studies demonstrated that psychiatric morbidity is increased in children with ESRF when compared with controls, whatever the stage of illness. But for the most part psychiatric problems are short lived, taking the form of either adjustment disorders or minor symptomatology not seriously handicapping to the child's emotional state or quality of life. For children in the predialysis stage these minor psychiatric problems were manifested primarily in the school setting and through mood changes, but there were no deleterious effects on the child's self concept.^{5,7} The more severely ill children on haemodialysis manifested their psychiatric problems primarily in the family and problems were not reported in the school environment, they also had prominent mood changes, and in this group self esteem was adversely affected.^{5,7} The transplantation stage was characterised by improvement in the child's mood and self concept in parallel with parental perceptions of improvements in general physical and psychological health, but children still showed higher levels of minor psychiatric morbidity at home than healthy children.⁹ Social development, measured by the presence of a special friend, showed a clear gradient according to severity of illness: thus

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30% of children on haemodialysis reported having a best friend compared with 55% of those at the predialysis stages, 68% after transplantation, and 82% of healthy controls.^{6,7,9}

Young adult survivors of ESRF did not have increased psychiatric morbidity when compared with healthy matched adult controls,^{10,11} thus the increased minor psychiatric morbidity was confined to childhood and adolescence. If young adults demonstrated psychopathology congruent with the child findings, they tended to have mood disorders. Adult patients did show a lowering of self esteem, which was linked to earlier onset of illness and lower educational achievements. Additionally, fewer patients than controls were married or had established intimate relationships outside the home. Those who had established such relationships tended to report more stress from intimate relationships than healthy controls in similar situations. This may have been related to fewer ill children having special friends in childhood together with their closer dependence on parents and family relationships. Fewer patients were in full time employment or were living outside the parental home but the majority were satisfied with their circumstances.

The effects of the illness on parents and other family members may be regarded as an additional measure of adjustment. Our work showed that parental mental morbidity and family stress were closely related to the stage of illness with more problems in the families of the more severely affected children.^{5-7,9} Transplantation was characterised by much better parental mental health: these mothers in fact compared favourably with the mothers of healthy controls: this may have been related to a sense of euphoria commensurate with the improved family freedom and physical well being in children with functioning transplants. In spite of this mothers of transplanted children reported increased behavioural problems in these children at home over those of healthy control children.

Methodological issues

NEED FOR CONTROL GROUPS

Our studies underline the need to study a control group of subjects. The conclusions of all the studies were influenced very significantly by the findings in the comparison group. The constitution of the control group depends very much on the type of question being asked. Basic questions that have vital importance for our knowledge include whether there are psychological risks associated with having a particular chronic physical disorder, what is the nature of the psychological consequences and what are the effects of these consequences on the psychological functioning of the child and family, and following from this what resources are required in providing for these children, emotionally, socially, educationally, and medically. Comparison with a normal population rather than other ill children is relevant in these circumstances. Using this approach we have shown differential rates of psychological difficulty between ill children and healthy con-

trols.⁷ If the question is whether different treatments of a specific illness or illness stages carry different psychosocial risks then comparison of homogenous groups with the same condition is appropriate.^{6,7,9} Different questions may require other types of control groups. For example studies of the efficacy of non-disorder specific psychosocial support might appropriately pool together children with different chronic illnesses provided that similar levels of disability can be documented across the different conditions.

PROBLEM OF SMALL NUMBERS

Many chronic illnesses, such as renal failure, are rare and this is another problem in designing studies. There are a number of ways of circumventing this problem.

Different illnesses may be compared with regards to psychosocial aspects if they share a number of characteristics involving disruption and distress, provided that they differ in one crucial aspect which is likely to have a powerful effect on psychosocial functioning. For example it has long been established that chronic illness that affects the brain is considerably more likely to be associated with child psychiatric illness than other illnesses not affecting the brain.³

If the researcher wishes to study very specific aspects of treatment of a rare severe condition with small samples in individual centres, this will often only be possible by recruiting multi-centre samples rather than comparing a treatment group in one centre with untreated controls in another centre. The study of the psychological consequences of growth hormone treatment in chronic renal failure might be an example of such a specific aspect of treatment.¹³ In such studies it is important to be confident that aspects of intervention such as take up rates for treatment, socioeconomic characteristics, severity of illness, patterns of medical support, and available psychosocial support are comparable between the centres.

National and international registries are another way of overcoming this problem. The paediatric registry of the European Dialysis and Transplant Association (EDTA) has fulfilled this function in Europe. Since 1971 it has produced annual reports; these include psychosocial adjustment in patients.¹⁴⁻¹⁶ Registry data has contributed enormously to understanding in the psychosocial and physical areas and is the only way in which large numbers of patients can be surveyed. Registries are appropriate for objective measures (for example crude employment status, presence of other major handicaps, marital status, etc) and are powerful tools for answering certain questions because of the patient numbers involved. Thus they are invaluable in providing broad measures of mortality, morbidity, and rehabilitation but some of the 'social' measures may be influenced very heavily by cultural and socioeconomic factors making invalid comparisons between countries and over long time periods. To describe in more detail the psychosocial impact of illness it is often necessary to complement these surveys with questionnaires

about behaviour and interview derived information.

USE OF QUESTIONNAIRES OR INTERVIEWS

Questionnaires about behaviour are commonly used to assess psychiatric disturbance in ill children but their limitations need to be recognised. As children's normal behaviour changes with development standardised instruments need to be used which have established norms for different age groups. Questionnaires cannot measure the extent to which parents have imperceptibly 'excused' changes in the child's behaviour as 'understandable' in view of the illness or conversely amplified them nor to what extent behavioural symptoms are an expression of physical ill health. Most instruments are not designed for chronically sick populations but for those without major physical or cognitive limitations. Questionnaires about behaviour are far less flexible than interviews in assessing the severity and resulting handicap of any psychological symptomatology; these are key features for the assessment of childhood psychiatric disorder. Conversely, exclusive reliance on interviews may miss definite but minor psychological symptomatology in the children. One consequence is that the concordance between questionnaire and standardised interview assessment of psychiatric morbidity is only modest, which may be interpreted by the naive as an inconsistent result. In our study of children with ESRF, parental interviews and questionnaires were discrepant in their identification of psychological problems in the child. This was because some severely affected children had few but very handicapping symptoms, whereas less affected children tended to have more symptoms that were comparatively mild in terms of severity and associated handicap.⁷ A comprehensive measure of the illness burden must examine the whole spectrum.

SITUATION SPECIFICITY

A related issue is the setting or situation in which the child's adjustment is being assessed. Using questionnaires it is common for parents and teachers to identify different children as having psychological difficulties.¹⁷ In our study we found that questionnaire assessed school problems were increased among the less severely affected children at the predialysis stage but not among children on hospital haemodialysis.⁷ These problems were only identified by the research questionnaires, the teachers had not previously recognised the problems. Study of the children in a different setting, namely the haemodialysis unit, showed that a number manifested problem behaviour at the unit, but these problems were not reported to the same extent by their parents.⁸ This problem behaviour was often provoked by the demands of the treatment programme both within the unit (needling for haemodialysis, physical demands of the dialysis session, etc) and outside the unit (particularly adherence to dietary regimens and medication).

TIMING OF ASSESSMENTS

Chronic illness is punctuated by episodes of stress with deterioration in the condition, admissions to hospital, or the introduction of new treatments. Few studies are explicit as to whether or not they include children at times of acute stress. It is well known that 'adjustment reactions' are frequent at those times.^{18,19} Adjustment reactions are a defined category of psychiatric disorder characterised by emotional and/or behavioural symptoms clearly linked in onset to a stressful event and time limited in manifestation.¹⁹ These may greatly confound any measurement of the psychosocial impact of more chronic difficulties. In our studies we included only children who had been stable for at least six months, that is with no change of treatment status or major crisis whatever the level of severity of their renal failure. Our findings indicate that had we included children at times of crisis the rates of psychiatric morbidity would have been considerably higher. The point prevalence of psychiatric disturbance in ill children in childhood was 28%, whereas prevalence encompassing the whole of childhood was 47%, a considerably higher rate than in healthy controls (17%).⁷ This suggested that a considerable degree of disturbance in ill children was short lived and may be regarded as adjustment reactions arising at stressful times in the illness.

AREAS OF FUNCTIONING ASSESSED IN STUDIES

Formal psychiatric disorder, behaviours reported in response to questionnaire, self esteem, peer relationships, parental mental health, and family coping are all important in measuring the impact of illness but they are not synonymous even though there may be overlap and most of these variables (especially the last four) describe broad areas of functioning, not easily measured in a single simple instrument. Studies should be explicit about the areas being addressed. Our research into childhood renal failure has identified differential effects of illness on these different aspects of psychosocial functioning of children and their parents.

PROTECTIVE FACTORS

The effect of chronic illness on children's adjustment is complex² and not always adverse.²⁰ If illness mobilises factors that are psychologically protective this may facilitate the child's favourable adjustment.¹ In our studies we showed that as well as leading to family stress chronic renal failure in the child enhanced supports in a number of social areas.⁶ Chronic illness may lead to heightened maternal empathy towards and sympathy with the child and in young children it may not affect the security of parent-child attachments in the absence of repeated potentially stressful separations.²¹ Moreover, distressing experiences, if adequately handled may promote coping or at any rate not adversely effect it. This is suggested for example by the finding, against expectations, that in our older school-children and adolescents with ESRF requiring repeated hospital admissions, the number of admissions was not associated with the child's

psychiatric adjustment.⁷ Thus studies should always take account not only of the stresses produced by illness but also the possibility that experience may help children cope with stress and that supports might be mobilised to alleviate these stresses.

OBJECTIVE AND SUBJECTIVE OUTCOME MEASURES

The adult outcome studies highlighted interesting issues in terms of the definition of good and poor social outcome.^{10,11} What may be objectively regarded as poor outcome in terms of normalisation (fewer intimate relationships, less independent living, more unemployment) may be associated with a realistic acceptance and response to limitations together with a positive response from the family and represent good outcome in terms of the subjects' psychological health and satisfaction with life. Conversely what the EDTA¹⁴⁻¹⁶ and other studies²² have appropriately considered good outcome (high employment rates, independent living) may in fact be at the cost of greater stress for some of those individuals, a poorer subjective outcome. We suggest therefore that both objective and subjective measures are used. When this is done in our studies, the results suggest that most survivors are well adjusted, probably because of adjusted expectations to their work and social roles and their satisfaction with their lives is not substantially affected by illness. These results cannot, however, be applied to other samples without due regard being taken of other factors such as severity of illness, social circumstances, and socio-economic factors in the population being considered. The task of re-examining objective and subjective outcome measures and forming a judgment as to whether the cost of normalisation is too high for some individuals will remain until renal failure can be prevented or cured.

Future research

A number of important clinical areas can be identified for further exploration.

- (1) *Non-adherence to treatment regimens.* Poor treatment adherence was associated in our studies with psychiatric disturbance. This confirms findings from many years ago. Nearly 20 years ago Korsch *et al* highlighted the problems of non-adherence and suggested that identifying high risk patients and planning intervention programmes might reduce non-adherence.²³ Despite this, loss of transplants because of non-adherence remains an important problem both in Europe²⁴ and the USA.²⁵ In the American study 13% of recipients lost their grafts because of non-compliance.²⁵ Twenty two of 47 adolescents were judged to be non-compliant of whom 56% lost some of their graft function and 15% lost their grafts. Future work should address more specifically the links between child psychiatric adjustment, adherence to treatment, and psychosocial variables.

- (2) *Ways of optimising the children's educational progress and fostering their social development* should be explored.
- (3) Detailed study of *children's and families' coping at stressful periods in the illness* will assist in developing interventions to increase psychological resilience and coping.
- (4) With younger children with ever more complex problems being taken on to ESRF programmes the burdens of care are increasing and *the particular burdens in young children* merit detailed study. It is clear from our clinical work that these families are less able to mobilise support and the stresses will be more disabling.
- (5) Clinical experience also suggests another at risk group of families are *those with major psychosocial disadvantages before the start of ESRF*. These families almost certainly require earlier identification and monitoring of their needs. Evaluation of proactive interventions in this group is another area in urgent need of research.

Some of these issues may not be disease specific and may, therefore, be suitable for research across different chronic illness conditions.

Conclusions

This series of studies has sampled a well defined population of children treated by the same unit; stages of illness and severity of the condition have been documented and controlled for, a longitudinal perspective has been taken, and comprehensive measures of psychiatric and social adjustment have been used. This provides a model of thinking and of research that may be usefully extended to other conditions.

In our view future studies should address specific questions. Study design should utilise developmentally appropriate well validated instruments complemented, if appropriate, by semistructured interviews. The situation in which the assessments are conducted and their timing also needs careful planning. Use of appropriate comparison groups is essential. Where stresses are identified counterbalancing supports need to be explored. Assessments of outcome should include subjective and objective measures.

There is a danger, of course, that such well designed scientific studies might fail to capture an important dimension(s) of the burden of chronic illness. This tension between the detail possible in anecdotal reports and the possible loss of detail imposed by more rigorous scientific designs has been highlighted by McFadyean and Altschuler who suggested that these two approaches should be seen as complementary.²⁶ The least helpful way of approaching these problems in future would be further cross sectional studies using inappropriate or incomplete methodology.

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