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Special Problems of Children with ME/CFS and the enteroviral link

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Abstract

Since 1997, it has been known that ME/CFS constitutes the biggest cause of long term sickness absence from school, in both staff and pupils. The scale of the problem in children is substantial, and the pattern of illness in schools suggests a prominent role for virus infection, for example, the clustering of cases. The Dowsett/Colby study of 1997, researching long term sickness, reported on a school roll of 333,024 pupils and 27,327 staff, and found a prevalence of 70/100,000 in pupils and 500/100,000 in staff; 39% of cases were in clusters of 3 or more. The peak age was 14-16 years. The illness is known to be potentially very severe and chronic. In addition, the Tymes Trust has reported that many affected children struggle for recognition of their needs and feel bullied by medical and educational professionals. Children should have time to recover sufficiently before returning to school; sustainable, energy-efficient and often home-based education is important here, to fulfil legal obligations. Research is needed into viruses that trigger childhood ME, for example, enteroviruses, and into the neurocognitive defects caused by ME. We should recognise the value of previous biological research and records of outbreaks and I recommend that ME be made notifiable due to the encephalitic nature of the effects commonly reported in this illness.

Since 1997 [1] it has been known that ME constitutes the biggest cause of long term sickness absence from school, in both staff and pupils. The potentially severe and chronic disease now called ME/CFS by the Department of Health [2] isolates children and young people, who are often too ill to go out and need other ways of making contact with the world and of continuing their education [2].

Young people themselves are often their own best advocates. As long ago as 1989, two young people with ME started their own newsletter, 'TYMES' (to stand for The Young ME Sufferer). Their commendable initiative developed into a national charity, The Young ME Sufferers Trust (www.tymestrust.org) which has co-funded, with ME Research UK and Search ME, the first biomedical study of ME/CFS in children, now in progress at the University of Dundee to ascertain whether the abnormalities found in small blood vessels in adult CFS/ME patients, such as sensitivity to the endothelium-dependent vasodilator acetylcholine [3] are present in children. Over the last two years, the Trust's professional guides for doctors and teachers have been placed on a free website at www.tymestrust.org which has been chosen for national archiving by the Wellcome Trust. The Tymes Trust also runs a Professionals Referral Service (Table 1) where medical, educational and social service professionals can consult fellow professionals with experience of treating or educating children and young people with ME.

The Trust believes that it is important to remain true to the original aim of the children who started 'TYMES'. It has always placed emphasis on the value of the patient's personal experience of the illness, now validated by the government's Expert Patient scheme. Thus the Trust's Advice Line Team is unique in all having personal experience of ME, and particularly of ME in children, as well as the necessary professional guidance and information. In addition, the Tymes Trustcard pass card system (Table 1), supported by the Association of School and College Leaders, is being used successfully in schools to support pupils when they are able to attend. The children's own voices are also published, together with activities, and information for parents and older readers, in *Vision* (Table 1), the only ME publication designed for an all-age market.

The government's stress on the relevance of personal experience of chronic illness is one with which the Trust concurs; I value the combined approach running the Trust and continuing to develop its initiatives despite residual limitations. It was only in 1985, when I myself was diagnosed with 'Royal Free Disease' [4] and then 'ME', following a severe Coxsackie B virus infection, that I realised what having ME (the term used by the consultant microbiologist in my case) really meant. Nobody can prepare for such a dramatic and life-changing illness, not even someone who for many years has handled out of hours emergency calls in a busy urban area, as I had done for my husband's general practice. I had been awaiting the then housing Minister Ian Gow for a luncheon at the school where I was Headteacher when violent pains in my back heralded what might have been influenza, but was not. My memory of the next few months is mostly blank. I was unable to rise from my bed, my body was continuously wracked with pain, I lost a good deal of my vocabulary, and when I did manage to reach the staircase, my brain had 'forgotten' how to walk down it and I fell. It was very similar to what I know of polio [5-7]; my breathing was affected, I could not grip anything, and my legs refused to work. The pain continued, particularly in the large muscles of the back, stomach and thighs, which demonstrated visible fasciculations, and also in the chest, where the pain was thought to originate in the intercostal muscles, and not in the heart itself, although I am not personally convinced of that. I was almost too weak to chew and with no curative treatment available, my disabilities were still so profound five years later, as is typical of ME, [8] that I officially left my profession to rebuild my health.

The scale of the problem in children and the pattern of illness in schools is of particular concern. There is clustering of cases [1,9] frequently misunderstood as imitation of sick friends or siblings for secondary gain. Such gain is rare, with some children becoming at least as disabled as I was myself; less severe cases still lose their entire social lives, as relapse can occur if a person makes too much effort, which in some cases can be minimal [2]. The Dowsett/Colby study of 1997 [1], researching long term sickness reports on a school roll of 333,024 pupils and 27,327 staff, found a prevalence of 70/100,000 in pupils and 500/100,000 in staff; 39% of cases were in clusters of 3 or more. The peak age was 14-16 years. When the BBC began to take an interest in misunderstandings surrounding the illness, it ran its own survey (BBC Panorama Survey, 1999) which I co-designed. There was a 62% response from 741 families surveyed, revealing that 59% of families had been told that their child's illness was psychological; 15% of parents had been told that their own psychological problems were the cause, 7% had been subject to child protection procedures, and 4% had been 'labelled' as cases of Munchausen's Syndrome by Proxy. None of these suspicions were, to my knowledge, upheld and statistically they appear anomalous. It seemed that the children's illness was so severe for so long, that both medical and educational professionals were finding it impossible to believe that these were physical impairments when the usual tests were inconclusive. We now have ample evidence of physical abnormalities in this illness, and I cite only a fraction [10-12].

ME has not uncommonly been mistaken for school phobia, anorexia nervosa, neglect, child abuse, MSBP (FII), or Pervasive Refusal Syndrome (PRS). The common factor is often the perception of the illness as not capable of causing profound physical disability over a long period of time, with Cognitive Behaviour Therapy mistakenly employed as a 'cure' (Table 1). When an ME/CFS diagnosis is made, it is important to recognise that not only is the child's right to suitable education - ie. education in a manner suitable for their disabilities - enshrined in law, which includes education in the home where appropriate, but this education must legally be provided after 15 days of illness to any child unable to access school [13]. Sadly, in a study of 126 families by The Young ME Sufferers Trust in 2003 (Table 1) it was found that 87% had struggled for recognition of their needs and 81% had needed to move school to obtain recognition of their needs. Another concern was that 84% reported feeling bullied by the attitudes of professionals. Medical professionals can do a great deal to support the child's need for education in the home, whether it be by a teacher, or by an interactive online system that encourages contact with other students (Table 1).

The purpose of education is to educate; this may appear too obvious to state until one considers that attendance at school may be used as a measure of 'recovery' whereas it is simply a measure of how often the child was able to get into school, whether or not this was educationally useful. Children with ME are not strong until they have substantially recovered. Intellectual performance appears to be impaired and reduced cortical blood flow has also been noted [14]. If the child has truly 'recovered', then the time spent in school should lead to a comparable educational performance to that of a healthy child. Often, it does not. According to one Local Education Authority, the grades gained by children with ME using online education at home appear to be substantially higher than would normally be expected from children with ME studying through more conventional means (Warwickshire Local Educational Authority, 2005, personal communication). This may be due also to the typical pattern of intermittent attendance and absence that occurs when a child who is not robust keeps relapsing due to school attendance (Table 1). Therefore social considerations need to be balanced against legal rights to a suitable education to obtain qualifications. It is quite possible to provide for social contact separately from study, to gain the best of both worlds; children's health is thereby preserved, they can mix with friends when appropriate, and can also be proud of the qualifications they have achieved.

It is not commonly known that, although helpful and in some cases invaluable, there is no requirement for a consultant's recommendation for a child too unwell to access school to be provided with an alternative means of education. If there were, it is likely to be illegal, as few consultants could meet the 15 day statutory deadline (Table 1). The GP's recommendation, as a qualified doctor, is sufficient. After a meeting with the Trust, Parliamentary Under-Secretary of State Lord Andrew Adonis wrote: 'It is unfortunate if, as calls to The Young ME Sufferers Trust advice line would appear to indicate, the advice in *Access to Education* is sometimes being misinterpreted as insisting that local authorities obtain a report from a consultant before action can be taken to provide support to children with ME who are unable to attend school full time. Our guidance is quite clear. We say that ideally an early diagnosis should be made by a consultant paediatrician but we do not suggest that this is a requirement' [15].

As a former Headteacher, I have spent many years developing special principles of education for children with ME that are working in practice (Table 1). 'Energy-efficient Education' works by balancing the energy needed for healing against that needed for education. A child who is recovering is a child who can achieve educationally. Conversely, if the child becomes worse, education suffers. If educational methods *make* the child worse, then education has itself caused educational disadvantage. Education is something other than 'schooling'. One must set up a goal-oriented environment to help these children achieve what is not possible for them in an inappropriately structured one. One must also structure the curriculum appropriately, and make full use of special arrangements for academic examinations, including, if desirable, taking the examination in the home to maximise the grade. Special arrangements are a legal right. The provisions set out by the Qualifications and Curriculum Authority (QCA) and explained by the Trust do not aim to give preferential treatment to a child, but to level the playing field so that disadvantages in comparison with healthy children are, in so far as is possible, minimised. Medical professionals are of invaluable support to children with ME as facilitators, recognising that achievement can only occur when the environment is structured to enable it to happen. Success is a huge morale-booster. It helps sick young people to hold up their heads with pride.

There is a school of thought, one that appears to treat ME as being perpetuated by psychological problems, that says children with ME *need* to be in school or they will not recover. The individual experience of families whom I advise as a professional educator indicate the opposite. This aspect of ME in children needs further study. Since the first article on ME in children and educational considerations was published in the British Journal of Special Education [16] there has been a slow development in the recognition that children with ME need special educational provision, now enshrined in government statutory guidance [13] and that this and taking into account not just the severity but the stage of the illness helps health to improve (Table 1).

There is now a pressing need to consider developing our epidemiological knowledge of ME. The late consultant paediatrician Dr Alan Franklin had a maxim: if the child doesn't fit the theory, then the theory is wrong (Table 1). Suspicions among some physicians that groups of children with ME are copying one another, rather than physically ill, has not only led to misunderstandings and obstacles to special educational needs provision, but also to the sidelining of an epidemiological phenomenon; children may well be the epicentre of this illness. The Dowsett/Colby study [1] was conducted to determine 'whether the recognition of multiple cases of myalgic encephalomyelitis/chronic fatigue syndrome (ME/CFS) in one school is an unique experience'. In the 1098 schools situated in six English Local Education Authority (LEA) areas that were investigated, the diagnosis of ME/CFS was significantly associated with case clustering. The outbreak of 'Summer Flu' with gastroenteritis in a village school that had prompted the study had involved 70 out of 230 9-11 year old children. In the autumn term, some 10% of those affected in the summer complained of profound fatigue and cognitive disturbance that had not resolved on follow-up two years later. One child's encephalitic illness was shown to be due to an enterovirus infection in July but investigations in other cases were delayed and therefore unhelpful. Relapse in the same pupils occurred the following year.

Giving evidence at the Gibson Parliamentary Inquiry into ME, the Trust has proposed four priorities for research. These are:

- That an enteroviral study be set up as suggested by JKS Chia in his review of the role of enterovirus in chronic fatigue syndrome. [17]

Dr Chia explains that whilst 'initial reports of chronic enteroviral infections causing debilitating symptoms in patients with CFS were met with scepticism' and had been 'largely forgotten' for the past decade, 'observations from in vitro experiments and from animal models have clearly established a state of chronic persistence through the formation of double stranded RNA, similar to findings reported in muscle biopsies of patients with CFS'. He goes on to say that recent evidence has not only confirmed the earlier studies, but also clarified the pathogenic role of viral RNA through antiviral treatment.

Dr Chia summarises the available experimental and clinical evidence that supports the role of enterovirus – the virus family that includes the polioviruses. He quotes tests demonstrating that antibody concentrations against enteroviruses in patients with CFS were still raised more than 48 months after the acute phase. Pointing out methodological flaws with studies that once threw doubt on the enteroviral connection, he concludes:

Thus, renewed interest is needed to study further the role of enterovirus as the causative agent of CFS. [...] A well designed, randomised, controlled trial of antiviral treatment will ultimately provide crucial information on the pathogenic role of enterovirus in patients with CFS and other chronic diseases.

- That ME/CFS be made notifiable in schools – staff as well as pupils – in order to demonstrate the pattern in school-age children; there are mini-epidemics and clusters.

Encephalitis is already notifiable. In adopting the term Chronic Fatigue Syndrome we have blurred the distinction between those with the encephalitic condition known as ME as described by Ramsay, Dowsett and others [8] from those who have a more generalised condition.

- That all previous work on ME epidemics over the decades, eg during the whole of the twentieth century, be revisited; it has been sidelined in the adoption of the unsatisfactory name Chronic Fatigue Syndrome.

Many of the epidemics were in schools and this knowledge is vital to revisit and correlate with our increased knowledge of virology in the 21st century.

- That there be further study of cognitive impairments caused by ME/CFS

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Table 1. Special interest documents available at The Tymes Trust (www.tymestrust.org)

Special interest document	Internet link to document
Professional referrals service	www.tymestrust.org/pdfs/referralservice.pdf
The Tymes Trustcard	www.tymestrust.org/pdfs/trustcardinfo.pdf
Vision	www.tymestrust.org/pdfs/vision2006-1.pdf
Pushing the Boundaries in ME/CFS*	www.tymestrust.org/pdfs/pushingtheboundaries.pdf
Back to School?*	www.tymestrust.org/pdfs/backtoschool.pdf
Succeeding with ME	www.tymestrust.org/pdfs/succeedingwithme.pdf
The SENCO's key role in supporting pupils with CFS/ME†	www.tymestrust.org/pdfs/senco.pdf
GP's Good Practice Guide to Education for Children with ME	www.tymestrust.org/pdfs/gpguide.pdf
Ten Points on the Education of Children with ME*	www.tymestrust.org/pdfs/tenpoints.pdf
Care of CFS/ME in children‡	www.tymestrust.org/pdfs/careinchildren.pdf

*First published in Special Children by Questions Publishing

†First published in SENCO Update by Birmingham University School of Education

‡First published in GP Magazine by Haymarket Publishing

SENCO, Special Educational Needs Co-ordinator

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