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Children's sleep disorders: a case of serious neglect

There can be few more striking examples of the gap between clinical need and provision of services than sleep disorders medicine, especially concerning children. Sleep problems are endemic and yet their recognition, diagnosis and treatment constitute a blind spot in medical and other healthcare education. The scope and importance of sleep disorders medicine is grossly underestimated by the limited (often perfunctory) accounts of children's sleep and its disorders in textbooks of paediatrics and child psychiatry.

In clinical practice, major errors are made in all the main categories of sleep disturbance. Sleeplessness is frequently treated symptomatically without considering the underlying cause; excessive sleepiness is commonly misconstrued as laziness or some other form of psychological shortcoming; and the many types of episodic disturbances of behaviour associated with sleep are regularly confused with each other diagnostically. The consequences to patients and their families of these mistakes are inevitably serious.

There are several reasons for this unfortunate state of affairs. One major factor must be the wide-ranging nature of the study of sleep and its disorders which crosses many of the boundaries between conventional disciplines and medical specialties. This poses problems for the more traditional teaching and training programmes where learning and the acquisition of skills are still confined to separate compartments with little attempt at integration. In this setting, multidisciplinary areas of discourse and clinical practice are likely to be poorly represented. This is especially so with the ever-increasing pressure on professional curricula, particularly in medicine in view of the spectacular advances in molecular medicine, imaging techniques and other technologically sophisticated and prestigious fields.

Sleep disorders medicine is likely to make increasing use of such impressive advances but, as a specialty, it has difficulty competing for time in teaching

courses, especially in view of its unfamiliarity to teaching programme planners. The scientific study of sleep and its disorders is very recent and with the emphasis still firmly on adults. Indeed, reviews of the history of sleep disorders medicine hardly mention children at all. It is a tribute to the imagination and diligence of both basic and clinical sleep researchers that so much information has accumulated in such a short period of time. However, the imbalance remains, with children's sleep disorders medicine still under-represented in clinical practice and research.

A more fundamental reason why sleep problems are not taken sufficiently seriously by professionals may be the supposed basic familiarity of such problems. Everyone experiences difficulties with their sleep at some time or other. Usually, the problem is short-lived and more a nuisance than anything more serious. Many parents are only too familiar with their young child's refusal to go to bed at the required time, or being woken in the night for attention. This is easily seen as part of the mixed blessing of parenthood and something to be endured until matters (hopefully) improve. In other words, there may be a common feeling that sleep problems do not merit a great deal of professional attention compared with other difficulties. This rather casual approach to sleep problems appears to be quite widespread.

In fact, any such attitude flies in the face of the evidence that, at all ages, sleep disturbance is the cause of much suffering, disadvantage and risk. This claim does not rest simply on the overvalued idea of sleep disorders medicine enthusiasts, it is supported, for example, by the closely argued case for more educational and clinical interest and more research funding presented to the US Government in the Wake Up America documents (National Commission on Sleep Disorders Research 1993, 1994).

A main point in these and less extensive accounts to the same effect (such as Dement & Mitler, 1993), is that persistent sleep disturbance often causes much personal distress, poor educational and occupational performance, and social and recreational difficulties. It is also implicated in many types of accident, including man-made disasters. Physical- and mental-health problems may also result. The cost to the national economy is considered to be huge. There is no reason to believe that the impact of sleep disorders is any less than universal, or that adverse effects are confined to adults on whom, again, emphasis has been placed in these publications.

These facts need to be brought home to the public and professionals alike. Those suffering from sleep disturbance (and their relatives) should be encouraged to recognize that such problems are not only troublesome but potentially serious, and that effective treatment (if based on accurate diagnosis of the underlying cause) can be available. The idea that medication is the only available treatment

needs to be corrected and the value of psychological treatments more widely known. Raised public expectations that help should and can be available would act as a spur to the better provision of clinical services for sleep problems. This, in turn, would require better training for providers of those services. The basic need is better education all round.

Despite the quite entrenched reasons why sleep disorders medicine has been a Cinderella specialty, optimism that matters will improve is justified. It is the experience of teachers in the field that the topic is invariably interesting to audiences, whether lay or professional. Those with a sleep problem of their own, parents and other relatives, teachers, medical students, neurologists, paediatricians, psychiatrists and psychologists invariably want to know more when sleep and its disorders is presented as the intriguing subject that it is, with important practical and clinical aspects, as well as many research possibilities.

This demand for further information is only partially met at the present time. The neglect of sleep and its disorders in professional education has been a self-perpetuating problem with relatively few enthusiasts (capable of enthusing others) entering the field. However, a trend towards more professionals from various backgrounds developing a special interest in the area is evident from the increasing membership of national and international sleep societies and the establishment of new national groups.

A number of textbooks, published in recent years, will have made their own contribution to increased awareness of and knowledge about the sleep disorders field, although their main appeal seems likely to have been to those already specially interested in the subject. Books written for sufferers themselves and relatives have also been a welcome development. The present text is intended to fill the gap between books for professionals with a specialized interest in children's sleep disorders and those written for parents. It emphasizes practical approaches to the recognition, diagnosis and treatment of sleep disorders in infancy, childhood and adolescence, and is intended to be useful mainly to paediatricians, psychiatrists, psychologists and others who are professionally concerned with child health and welfare.

To maintain the essentially clinical rather than academic nature of the text, referencing is restricted. However, sources of more detailed information are cited in the form of selected review articles and chapters, together with more specific publications to supplement or update the more general accounts, or to emphasize certain points. Heavily technical accounts of limited appeal to the nonspecialist reader have been avoided. Personal clinical impressions are also included on aspects which have not been the subject of empirical research.

For the most part, the references chosen are concerned with studies or observations on children or adolescents, but some reports on adults are included where

the content seems likely also to be relevant to younger patients. The case histories throughout the book are drawn from the author's own clinical practice, preserving the anonymity of each child and family. Although the patients have mainly attended a special sleep disorders clinic, the basic principles that they illustrate, and the treatment approaches described, are relevant to children with sleep disorders in general, seen typically in non-specialized clinical services. *The main aim is to provide the reader who does not have a specialized interest in sleep disorders with an overview and a suggested clinical approach to the many conditions covered in children's sleep disorders medicine.* The fine details of treatments are not attempted but can be found in the references cited. Necessarily, many statements are subject to revision in the light of further research.

In places, the child is referred to as 'he' purely for convenience. The layout adopted, with bullet points and other subdivisions, is intended to facilitate access to the information contained in the text.

Chapter 2 consists of a more detailed account of the main general points made in these opening comments.

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General issues

History of sleep disorders in children and adolescents

Those of an historical inclination might be interested in the provenance of present-day practice and research in children's sleep disorders. There is, in fact, not much to say, and what can be said is a patchwork of historical, literary and clinical references which illustrates that children's sleep disorders have long been recognized but without any systematic study until recent times.

The starting-point might best be described as 'prescientifically paediatric'. In *The Boke of Chyldren* (1545), Thomas Phaire included in his list of 'the manye grevous and perilous diseases' with which children of his day were afflicted 'terrible dreames and feare in the slepe' (caused by 'the arysing of stynkyng vapours out of y^{e} stomake into the fantasye, and sences of the brayne') and also 'pissing in the bedde'. To what extent Phaire's suggested remedies would meet the requirements of present-day evidence-based practice is doubtful. For example, his observations of childhood nightmares were accurate, but the treatments he recommended included 'a lytle pouder of the seedes of peonie and sometimes triacle'. 'The poudered wesande [windpipe] of a cocke' and 'the stones of an hedgehogge poudred' were some of his recommendations for bedwetting.

To do justice to Phaire, it must be said that at least he was drawing attention to the special medical problems of the young at a time when, in general, few if any concessions were made to children, who were viewed as small adults and dealt with accordingly (Pinchbeck & Hewitt, 1969). Just as childhood is a relatively new concept, paediatrics has largely been emancipated from general medicine only recently. Examples remain, in the provision of various clinical services, of inadequate acknowledgment of the special needs of children.

Some of the best early clinical descriptions of sleep disorders are available in the novels of Charles Dickens. Cosnett (1992) has discussed how Dickens took an interest in sleep problems, possibly because of his own insomnia. He described them in several of his characters in such detail that they must have been based on

real people. The best-known example is the Fat Boy Joe in *Pickwick Papers* (Dickens, 1836/7). The usual diagnosis of Joe's excessive sleepiness is obstructive sleep apnoea, although his obesity (seen in only the minority of children with this condition), overeating and periodic behavioural disturbance raises other possibilities, notably the Kleine–Levin syndrome.

The short history of the scientific study of sleep and sleep disorders has been described by Thorpy (2000). Significantly, this and other historical accounts contain little about children and adolescents. It is the case, however, that some time ago the Stanford University group drew attention to the clinical importance of sleepiness in children and chided physicians for usually not taking any interest in the problem until significant complications had arisen (Anders et al., 1978). In fact, there had been a much earlier warning about the potentially serious effects on young schoolchildren of lack of sleep. The anxieties expressed by Dukes (1905) were overdramatic, but his concerns anticipated similar warnings expressed by later writers. He complained that 'younger pupils are all offered the same number of hours as the seniors for sleep. What this means to children is lowered vitality, apathy, bloodlessness, and diminished growth of body and brain. It renders the child an easy prey to disease, and causes slight fainting attacks resembling those cases of epilepsy termed petit mal.' Not a great deal of notice has been taken of Dukes nor, indeed, Anders and his colleagues, except, to some extent, regarding the common problem of sleepiness in adolescence which became the special interest of Dement and Carskadon at Stanford (Dement, 1990).

As mentioned already, there has been a determined attempt more recently (especially in the USA) to bring to public, professional and governmental attention the importance of sleep disorders in adults, but with little mention of children in whom sleep problems are common and for whose development persistent sleep disruption can be expected to have particularly important implications. The International Classification of Sleep Disorders (American Sleep Disorders Association, 1997) is essentially adult based. It contains limited recognition of the differences between adults and children (discussed later) in the pattern of occurrence of sleep disorders, the way basically the same condition can present itself clinically or physiologically, and the different implications for psychological and sometimes physical development.

That is not to say that much is not already known of considerable clinical importance and research interest regarding children (Ferber & Kryger, 1995). There is still an urgent need to incorporate this knowledge widely into clinical practice and to pursue answers to the questions and uncertainties that remain concerning the nature and consequences for young people of the many ways in which sleep can be disturbed.

Prevalence of children's sleep disorders in the general population

Because of failures of recognition and limited referral of cases for medical attention, it is not known with accuracy how many people of any age suffer from sleep disorders. Much greater awareness and diagnostic sophistication are required before reliable figures can be expected. In the meantime, it is reasonable to assume that the usually quoted figures are underestimations. Even so, there is ample evidence that sleep disorders are very common.

Overall, it seems likely that at least 20–30% of children from infancy to adolescence have sleep problems that are considered significant by them or their parents (Mindell, 1993). Intriguingly, the pattern of reported problems can vary from one nation to another according to a survey of 11–16-year-olds in eleven European countries (Tynjälä et al., 1993).

Perhaps the most reliable figures for more specific sleep problems are those based on population studies of settling and night-waking problems in toddlers, about 20–25% of whom pose such problems to their parents most nights and sometimes every night (Richman, 1987). The importance of collecting information from more than one source about the occurrence of sleep problems is illustrated in the survey of sleep habits and sleep disturbance in 4–11-year-old children by Owens et al. (2000). By using a battery of sleep questionnaires for parents, teachers and children themselves, it was possible to identify a range of types of sleep disturbance some of which might well have been missed if only parental information had been collected.

Severe sleep problems show a further high occurrence rate in adolescence, including those due to erratic sleep–wake patterns and the delayed sleep phase syndrome (DSPS) (see later) which causes a combination of inability to get off to sleep and great difficulty getting up in the morning. Although at a lower level, the reported prevalence rates for some other conditions causing excessive daytime sleepiness in young people also illustrate that these disorders are by no means the rarities that might be supposed. For example, upper airway obstruction during sleep is said to occur in about 2% of children from infancy onwards (Carroll & Loughlin, 1995a). If the suggested prevalence estimates for narcolepsy (4–6 per 10,000 in the United States) are correct, then the rate of this disorder in children and adolescents must be at least a third of this figure because of the general agreement that the first symptoms frequently appear at this early age (Stores, 1999a).

Prevalence rates for the third main type of childhood sleep problem (i.e. in addition to sleeplessness and excessive sleepiness), namely episodes of disturbed behaviour associated with sleep (the parasomnias), are difficult to judge. Only a small proportion seem to come to medical attention, partly because some occur

infrequently in perhaps the majority of children. Accurate prevalence rates are also made difficult to obtain by the fact that different parasomnias are often not distinguished from each other very clearly, with a tendency to use the term 'nightmare' for any that involves dramatic changes of behaviour. Again, it seems that the frequency with which children are seen professionally for help with this type of sleep disturbance is no guide to the occurrence of such problems in the community at large.

Children at high risk of sleep disturbance (Stores & Wiggs, 2001)

Sleep disturbance is common enough in children in general, but some groups are particularly prone, namely certain children with various types of physical or psychological disorder including those with a learning disability (mental retardation). However, there are some subgroups within the general population who are at special risk because of developmental factors.

General population

Some of the changes in sleep physiology in the period between infancy and adult life may predispose children to sleep disorders at certain stages of development. Other important changes having the same effect concern parenting or changes in life style.

- Rapid Eye Movement (REM) sleep is prominent in *early infancy*, possibly explaining in part why sleep seems to be fragile then as this type of sleep is less sound than nonREM (NREM) sleep.
- In the *toddler years* a basic aspect of parenting is to establish satisfactory sleep patterns. Failure to do so is common, resulting mainly in settling and night-waking problems in 20% or more of children of this age.
- Deep NREM sleep is particularly prominent in *early childhood*. This may be a reason why arousal disorders (such as sleepwalking), which arise from this form of sleep, occur mainly at this age.
- In contrast, the amount of slow wave sleep (SWS) decreases in *adolescence*. Of apparently greater importance, however, is that (out of keeping with the progressive decline at earlier ages) sleep requirements do not decrease at puberty. Also, the overnight sleep phase becomes delayed partly for biological reasons, judging by the fact that the extent of the delay is related to stage of puberty and that the timing of melatonin secretion changes in early adolescence in relation to this sleep-phase delay (Carskadon et al., 1997). Increasingly, as adolescence progresses, these physiological factors, combined with strong social influences (e.g. to stay up late especially at weekends) frequently cause sleep deprivation, daytime sleepiness and various adverse effects on mood, behaviour and performance (Carskadon, 1990).

The complex origins of the particularly high rates of sleep disturbance seen in adolescence illustrate the need to discover more about the psychosocial and other factors which put certain groups of children at particular risk of serious sleep disturbance and its consequences.

In general, current information about the origins of children's sleep difficulties is only preliminary. Infant's sleep problems are the most researched (Sadeh & Anders, 1993). A number of important biological factors including perinatal complications and temperament have been implicated, as well as socioeconomic factors such as overcrowding. Parenting practices and standards are obviously relevant, and also parental psychiatric illness (Seifer et al., 1996). However, outside grossly distorted or abnormal circumstances, precise prediction of which infants will develop serious sleep problems is difficult because of the many interacting influences involved. As will be mentioned later, relatively subtle factors have been suggested by recent research, such as mothers' attitudes and emotional reactions to their children's sleep patterns, perhaps based partly on their own experience of sleep as infants or their feelings of competence as parents (Morrell, 1999). Insight into such influences might explain why objectively the same degree of sleep disturbance is seen as a serious problem by some parents and not at all by others.

Chronic physical illness, disorder or disability

As part of being generally unwell, or because of discomfort or distress, acute illness usually affects sleep, mainly for the duration of the illness although the experience of being ill, receiving treatment or being *admitted to hospital* (White et al., 1990) can cause a sleep disturbance which persists for some time afterwards. For example, this has been described in children who have undergone intensive care, even for a relatively short period (Cureton-Lane & Fontaine, 1997).

It is difficult to think of any chronic disorder which is not complicated by long-standing sleep disturbance. It seems, however, that this is often overlooked because the physical aspects of the condition occupy the attention of those caring for the child. Parents may share this preoccupation, or consider that the sleep problem is inevitable and untreatable which is rarely true. Failure to identify and treat a child's sleep problems may well mean that the opportunity is missed to improve his overall well-being (and that of his carers) and to help him to cope with the other difficulties imposed by the basic condition.

Sleep may be disturbed by physical conditions in various ways.

- Examples of the effects of *night-time discomfort or pain* include severe atopic dermatitis (Stores et al., 1998d), juvenile rheumatoid arthritis (Zamir et al., 1998) and malignancy (Miser et al., 1987). Pain is, in fact, related to sleep disturbance and its possible psychological and physical consequences in a number of ways (Lewin & Dahl, 1999). These include direct adverse effects on

the duration and quality of sleep and the influence on sleep of the emotional upset caused by the painful condition. This is well illustrated in the consequences of burn injury where sleep disturbance can be very persistent (Lawrence et al., 1998). Promotion of better sleep is likely to improve the child's ability to cope with illness and possibly to hasten recovery from it (Adam & Oswald, 1984).

- *Some disorders worsen at night.* For example, this happens in asthma (possibly more often than usually supposed) in which the main sleep disruption appears to be frequent awakenings for variable lengths of time including brief periods which are likely to be undetected clinically (Stores et al., 1998c). The worsening of respiratory function in asthma (and also in bronchopulmonary dysplasia and cystic fibrosis) has been discussed by Loughlin and Carroll (1995). Childhood epilepsies, even in relatively mild form, are associated with high rates of sleep complaints, especially those suggesting poor quality of overnight sleep (Stores et al., 1998e). In some cases the cause appears to be nocturnal seizures, but other factors are also likely to be responsible, such as the underlying cause of the epilepsy and the accompanying psychological problems of many children with epilepsy (Cortesi et al., 1999).
- As will be discussed later, sleep disturbance caused by *upper-airway obstruction* (*UAO*) at night is reported to be common in the general population but it is particularly associated with a range of specific disorders. Main examples of this is learning disability (discussed shortly) and also some forms of cerebral palsy (Kotagal et al., 1994), neuromuscular disorders (Attarian, 2000), craniofacial abnormalities (Hoeve et al., 1999), myelomeningocele (Kirk et al., 1999), achondroplasia (Tasker et al., 1998) and sickle cell disease (Samuels et al., 1992). UAO has been implicated in the final events leading to sudden infant death syndrome (cot death) although the factors, including sleep mechanisms, leading to this dramatic outcome have yet to be made clear (Cornwell, 1995).
- *Sensory impairment* can affect sleep in different ways. Severe visual impairment is likely to lead to circadian sleep–wake cycle disorders because light perception is the main cue to establishing and maintaining a normal 24-hour sleep–wake pattern. However, other types of sleep disturbance, both physical and behavioural in origin, can also be expected in visually impaired children in view of the diversity regarding the cause of their condition and also comorbidity (Stores & Ramchandani, 1999). The very limited research on children with a hearing impairment suggests they also have a range of sleep disorders apparently including circadian sleep–wake cycle abnormalities (Oishibashie et al., 1993). Accompanying conditions, such as tinnitus (Rizzardo et al., 1998) or psychological disturbance (Roberts & Hindley, 1999), are likely to affect sleep.