Children with pervasive refusal

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Abstract
Four children are described with a potentially life threatening condition manifested by profound and pervasive refusal to eat, drink, walk, talk, or care for themselves in any way over a period of several months. The multiplicity and severity of the symptoms in these children do not fit comfortably into any existing diagnostic category. Long term and highly skilled nursing and psychiatric care is required to help these children to recover. The possible causes of this syndrome are discussed.

Child psychiatric disorders may vary from relatively mild, albeit distressing symptoms such as enuresis, encopresis, behaviour problems or school refusal, through to the devastating effects of psychotic illness or life threatening problems such as anorexia nervosa or severe depression with suicidal ideology and intent. We have recently seen a number of children with a profound and pervasive life threatening condition manifested by dramatic social withdrawal and determined refusal to walk, talk, eat, drink, or care for themselves in any way for several months. Careful history taking and physical examination and investigation at the time of presentation excluded any organic disease.

Case reports

CASE 1
A girl, aged 13, was transferred from a paediatric ward to our child psychiatric unit with a four month history of increasing pain and stiffness in her right leg, leading eventually to an apparent inability to walk. As her mobility diminished she reported that she could no longer see or swallow, and over the next few weeks she stopped speaking and lost the use of all four limbs. She refused, by screaming and vigorous physical endeavour, all attempts to assist her. Thus attempts at oral feeding led to her tightly closing her mouth, turning away her head, and spitting out any food or drink. There was no sleep disturbance. The only abnormality on physical examination was a finger sized hole in the hymen and abrasion of the right labium, with some vaginal discharge.

The girl's physical and intellectual development had been normal, but aged 4 she had been reported to be 'aggressive', and her premorbid personality was described as 'like an old lady, lacking childishness and fun'. She had a sister 10 years older than her, and her parents were in their 50s had both previously been treated for depression.

Despite intensive attempts to rehabilitate her she continued to deteriorate. She required nasogastric feeding and, on two occasions, manipulation of her joints under general anaesthetic to prevent contractures. The nursing care she required was that necessary for a patient with quadriplegia. She continued to resist all attempts to help her. Individual and family therapy shed little light on the cause of her illness, although an overweight and covert sexual relationship between the girl and her father was noted. Further investigation and exploration of this was unproductive.

After six months she slowly started to improve and one year after admission she could walk with minimal assistance, could obviously see although denying it, and could when necessary make herself heard. At this point she was transferred to the Bethlem Royal Hospital Adolescent Unit, with the expectation that she would require long term care. As her physical state continued very slowly to improve, her behaviour changed. She became far more outgoing, incited others to misbehave, stimulated interest in seances and black magic, and teased maliciously about sexual matters. On a number of occasions she ran away. She was finally discharged from hospital three years after her original admission and at three year follow up had remained well.

CASE 2
A girl, aged 11, was transferred to our unit from a paediatric ward with a one year history of panic attacks and prolonged episodes of screaming. For three months she had refused to eat, drink, walk, or take care of herself. She angrily refused to answer questions, described herself as very sad, and expressed a wish to die. Under no circumstances did she wish to go home. She resisted help with angry screams and at times physical aggression.

She was the second of three children and her physical and intellectual development had been normal. Her premorbid personality was described as 'anxious and perfectionistic'. Early separation problems had been noted, and at aged 7 there had been reports of 'sexually provocative behaviour'. There was no family history of significance.

The first three months of her admission were stormy with continuing and determined refusal to eat, drink, walk, or care for herself. She continued to scream whenever anyone communicated with her. Treatment consisted of a com-
bination of individual, group, and family therapy, and the use of the ward milieu. In individual interviews she eventually admitted to having a secret, and further painstaking exploration led to disclosure of prolonged and repeated sexual abuse, including the use of physical restraint and punishment. She refused to name the perpetrator(s). After the disclosure and further discussion of her experiences she slowly started to improve. She remained unable to allow a full physical examination but gradually became calmer and resumed eating, drinking, and walking.

She suffered a transient relapse during Christmas leave when she claimed to have seen the perpetrator. Subsequently she was made a ward of court and her parents were allowed only supervised access. Nine months after admission and 21 months from the onset of the illness she was discharged to a foster home, having fully physically recovered. She remains well at two year follow up.

CASE 3
A girl, aged 14, was admitted with a one year history of all the characteristic features of anorexia nervosa, including food avoidance, fear of fat, distorted body image, and weight loss of 8 kg. Six months before admission she had also become socially withdrawn, refused to sleep in her own bed, and had declined to open her Christmas presents. Full physical investigation had revealed no organic explanation for her condition and she continued to deteriorate. At the time of admission she was refusing to eat, drink, walk, talk, or care for herself. She was tearful with a stooped posture, and a terrified reaction to being touched, but there was no sleep disturbance. She would cringe when spoken to and cover her ears much of the time. She actively resisted all attempts to feed, wash, or comfort her.

She was the older of two girls and there had been no previous problems. Her physical and intellectual development had been normal and her premorbid personality was described as shy and conscientious. The only reported family history of significance was that the girl’s mother had an adoptive brother who was violent and alcoholic. She had witnessed episodes of his violence, although her parents denied that she had ever been a victim.

Despite intensive treatment including tricyclic antidepressants, individual, group and family therapy, and the use of the ward milieu, there was no improvement during the first three months of the admission. A change of medication to chlorpromazine made no difference and coincided with an acute abdominal crisis for which no cause could be found at laparotomy. An incidental observation was that even when very ill and in obvious pain she made no verbal communication. Medication was changed to sulpiride with no obvious immediate benefit, but gradual improvement commenced about three months later, nine months after admission.

She started tolerating small feeds and began to communicate by writing notes with a largely depressive content. Her self care and hygiene improved and she permitted touching, but for several more weeks retained her hunched, withdrawn posture and refused to speak. Approximately one year after admission she adopted a more upright posture, started talking, and ate and drank normal portions. Family and individual therapy focused on fears of violence, and although possible physical, emotional, and sexual abuse were investigated, none was elicited. She remained silent throughout these interviews. She was discharged, on no medication, to residential schooling 18 months after admission, and 30 months after the onset of her illness. At one year follow up she remains well.

CASE 4
A girl, aged 9, was transferred from a paediatric ward with a three month history of ill health, which started with a fall and cut lip, followed by complaints of sore and cracked lips, mouth ulceration, listlessness, and refusal to eat or drink. She became increasingly withdrawn and eventually stopped speaking, making only high pitched moans. On admission she was alert and interested in her surroundings but avoided all eye contact by covering her eyes whenever an adult came near her. She refused to walk or care for herself, and resisted all physical contact, including any form of feeding.

She was the second of five siblings of an Asian family in which there was no significant psychiatric history other than a first cousin with anorexia nervosa. Her development had been normal and she had had no significant health problems. Her parents described her as having been a hard working and popular girl who was very good with her younger siblings before her illness.

Treatment involved a combination of individual, group, and family therapy, and the use of the ward milieu. About 10 weeks after admission she began to show more overt interest by peeping through her fingers and making noises to draw attention to herself. She began to sit upright and started walking with support.

Her improvement slowly continued and nine months after admission all functioning was normal, except for a complete refusal to take anything by mouth, and a total rejection of her family.

Information from the family suggested that her mother had been physically and sexually threatened in the girl’s presence and that she had witnessed some marital violence. Furthermore the girl had shown fear of an uncle who occasionally looked after her. On one occasion she stated that she had been sexually abused by her parents but she was unable to give any further information. Attempts to explore this further have been inconclusive, although she had not retracted the allegation.

At the time of writing, 18 months after admission, she is living in a small group home and is eating normally.

Discussion
The most striking feature of these children is
their refusal to walk, talk, eat, drink, or take care of themselves. The word refusal has been used advisedly. In no case was there any neurological deficit to account for the symptoms. Neither was there any suggestion of a postviral state. These children demonstrated a wilful, angry, or frightened component to their nonfunctioning. What then is the diagnosis and why were these children so ill?

Much further work is required to develop a really satisfactory system of classification for child psychiatric disorders. In many instances it is difficult to fit a particular child’s clinical picture into a specific diagnostic category. Such a lack of fit applies particularly in cases as complex and severe as these. Each child had certain features of a variety of different diagnoses, including anorexia nervosa, elective mutism, anxiety or phobic state, depression, conversion disorder, stupor, and post-traumatic stress disorder. However, none of these children fulfilled the diagnostic criteria for any of these conditions as defined in the International Classification of Disease, 9th revision or the Diagnostic and Statistical Manual of Mental Disorders, 3rd edition.

Anorexia nervosa and elective mutism are insufficient diagnoses in that each refers to a specific area of functioning (eating or talking) and these children had far more pervasive problems. Anxiety, phobic responses, and depression were undoubtably features of the mental state of all these girls, although none of these terms alone seems adequate to describe their feeling state or their determinedly rejecting and self-punitive behaviour. There were similarities with conversion disorder, which is characterised by alteration or loss of physical function that suggests physical disorder, but instead is apparently an expression of psychological conflict or need. Our children manifested a very obvious wilfulness in their symptoms, however, and in particular a pattern of refusal. Such determined and wilful refusal negates the diagnosis of conversion disorder, which is manifested by inability rather than volition.

Stupor is described by Lishman as a state of akinnesia and mutism but with definite evidence of relative preservation of conscious awareness (p 8) and sometimes of a volitional nature. In ‘psychogenic stupor’ signs of conversion hysteria are commonly in evidence, the condition may wax and wane, and there may be an appreciable emotional reaction when sensitive subjects are discussed. Complete passive dependence on others for feeding and toilet functions is rare, and the patient may show signs of irritation when moved against his will (p 198).

Although these children certainly fit these diagnostic criteria to some extent, we are not convinced that stupor is the correct diagnosis, if only because the criteria are so wide, and because the children showed such a wilful negativism.

A paper by Berrios on stupor may also be of help in understanding this condition. He draws parallels between stuporous states and ethological models of freezing reactions, trance states, animal hypnosis, and tonic immobility, the latter being considered as a final step in the fear response against predation. Thus we might hypothesise that these children have adopted this posture as a final resort to escape an intolerable situation. This view is reinforced by the fact that we are currently treating two more children with a similar clinical picture, one of whom has finally been able to disclose that she has been sexually abused, but is clearly terrified of saying more. She has indicated that she and her mother would die if she told anyone exactly what had happened, and that she only feels safe in hospital having contact with no-one other than her mother and hospital staff.

The diagnostic criteria for post-traumatic stress disorder include the existence of a known stressor, re-experiencing of the trauma, numbing of responsiveness, or reduced involvement with the external world, and the presence of other symptoms such as hyperalertness, guilt, sleep disorder, cognitive difficulties, and avoidance behaviour. The obvious problem with making this diagnosis is the absence of any known definite stressor in all but one of the children. It would not be unreasonable to describe intrafamilial violence as a stressor, although by no means all children so exposed develop such serious illness.

Given that there is no completely satisfactory diagnosis it is clearly important to attempt to understand the aetiology. At presentation there were no obvious precipitating factors. Investigation of the intrapsychic state of these children during the course of their admission revealed a fairly uniform picture.

Each child denied conflictual feelings, projected anger, and showed fear of attack. Their families were characterised by denial of any conflicts or other problems and yet had histories of intrafamilial violence.

The contribution of sexual abuse to these children’s problems has to be considered. It was disclosed in one girl (case 2) and, given the physical signs, exceedingly likely in another (case 1). The diagnosis of sexual abuse is only valid if the suggestion of abuse by the family of case 3, sexual threats in that of case 4, and intrafamilial violence in both, necessitates a high index of suspicion, but there is a major problem in obtaining corroborative information from mute, withdrawn and frightened children. The findings of Kiser et al seem to be of some relevance here. They describe post-traumatic stress disorder when a reaction to sexual abuse as having three different forms of presentation:

(1) Avoidance: (a) restriction of affect, (b) withdrawal, (c) loss of interest, (d) aggression, (e) reluctance to talk, and (f) avoidance.

(2) Re-experiencing: (a) sex play, (b) talk and behaviour, (c) nightmares, (d) day dreams, and (e) flashbacks.

(3) Autonomic hyperarousal: (a) irritability, (b) aggression, (c) sleep problems, (d) anxiety, and (e) impaired concentration.

All the children fit into category 1, avoidance, with their pervasive refusal to participate in the normal activities of daily life, and each girl manifested some features of the other categories. It is also possible to conceptualise the refusal to eat and the resistance to washing and toileting as avoidance phenomena.

The aetiology of this condition is not comple-
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tely clear but may involve a two-stage process. The first is the trauma of sexual abuse, perhaps of a particularly sadistic nature, and the second is fear, induced by either a violent family member, threats, or the likely consequences of disclosure. The powerful silencing effects of threatened violence to a child cannot be underestimated. Summit vividly describes the plight of such children in his description of the child sexual abuse accommodation syndrome. The syndrome is composed of five categories: (1) secrecy, (2) helplessness, (3) entrapment and accommodation, (4) delayed, unconvincing disclosure, and (5) retraction.

Three cases described by Brown and Perkins bear some similarity to ours in that all were girls who experienced a profound deterioration in their physical and mental state necessitating urgent hospitalisation and assessment for a degenerative disorder. Their much younger ages, two being 5 and one 6-5 years old, allowed them to manifest uninhibited sexual play that led to rapid disclosure of sexual abuse in each case.

In older children the normal developmental decline in spontaneous play makes communication through that medium unlikely, and their cognitive development means that they can begin to weigh up the pros and cons of disclosure. At this age the children are caught between the developmental need to be within a family setting, while having the cognitive awareness that a disclosure is likely to break up their family. They are too old to disclose spontaneously, as did the children in the series of Brown and Perkins, and too young to be able to live independently of parents.

Discussions with colleagues have identified five more very similar cases to ours in girls aged between 11 and 15 (D Steinberg and D Black, personal communications) and we currently have two more such children on our ward. One of us (MT) has also recently seen a girl of 13 who had been mute and withdrawn for 10 years. After placement at a special boarding school she disclosed that she had been repeatedly and ritually sexually abused by several members of her extended family and threatened with death if she disclosed.

CONCLUSIONS
The multiplicity and severity of the symptoms in these children do not fit comfortably into any existing diagnostic category. The symptom complex can perhaps be understood as an extreme variation of so-called avoidance behaviour seen in post-traumatic stress disorder. The inability to disclose even the existence, let alone the nature, of the trauma because of the threatened consequences may account for the extreme severity of the disorder. While awaiting further elucidation of how children react to and cope with such experiences, we suggest that it may be useful to recognise this discrete syndrome of pervasive refusal, as manifested by refusal to eat, drink, walk, talk, or participate in any activity including self care or toileting. It is most likely to occur in girls between the ages of about 9 and 14 years, in whom there is a possibility of sexual abuse, but who are terrified of disclosure because of threats of extreme violence.

Treatment must be in a hospital setting, where the child feels safe and can stay for as long as is necessary. Skilled psychiatric care and the availability of staff who are expert in exploring the possibility of sexual abuse are essential. There is no one specific form of treatment for children as ill as these. The mainstays of management include a combination of individual and family therapy, the use of the ward milieu, and medication as required. Whether or not such children can return home must depend on individual circumstances. The decision will be made on the basis of what is considered to be in the child's best interests. If the child expresses a clear wish to return home and there is no reason to believe that the child will be harmed at home, and the parents are able to accept and work on the fact that there is a psychological explanation for their child's illness, then a gradual return to the family home is indicated. If these circumstances do not apply serious consideration must be given to alternative placement. In any event such complex decisions are best made in the context of a series of case conferences held under child protection guidelines.

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