Psychiatric Presentation of Crohn's Disease

Diagnostic Delay and Increased Morbidity

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Four children presented to child psychiatric clinics with a variety of symptoms. They were all later recognised as having Crohn's disease. There was a significant delay between the onset of symptoms and diagnosis, compared with a control group of patients with Crohn's disease whose presentation was with predominantly gastrointestinal symptoms, which was associated with evidence of increased morbidity. Children with abdominal and psychiatric symptoms occurring in combination need serial assessments of physical status, including height and weight, and measurements of inflammatory and nutritional status.
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Crohn's disease in childhood and adolescence usually presents with the classical triad of abdominal pain, diarrhoea and weight loss (Burbidge, 1975). It is also well recognised that Crohn's disease may occasionally present with predominantly extra-intestinal symptoms, with a picture which is difficult to distinguish from anorexia nervosa. Delays in diagnosis may occur as a result (Gryboski *et al*, 1968; Jenkins *et al*, 1988). However, it is less well recognised that Crohn's disease sometimes presents with other apparently primary psychiatric symptoms without prominent anorexia.

We describe four children with Crohn's disease, each of whom presented between the ages of 10 and 13 years with mainly psychiatric rather than abdominal symptoms. Only one child was diagnosed as having anorexia nervosa. Depression and anxiety were prominent in all four subjects. All had abnormal eating behaviours and abdominal symptoms.

In order to determine whether a psychiatric presentation was likely to be associated with adverse outcomes, the duration of symptoms and indices of disease severity at diagnosis in the four subjects were compared with a group of 19 control children with Crohn's disease diagnosed during the same period but without predominant psychiatric symptoms. Our data suggest a significantly increased delay in diagnosis and in morbidity in our four subjects.

Case reports

Case 1

This boy was originally referred to his local child psychiatric services aged 13 years for 'appalling eating habits'. These included a year of eating extremely small amounts and refusing to eat with his family. His referring family doctor noted a haemoglobin count of 10.8 g/dl and iron and folate deficiency, which were presumed to be dietary (see Table 1).

Psychiatric assessment confirmed a long-standing eating problem which had worsened over the previous two years. He ate extremely small amounts and refused to join his family at meal times. He had irregular bowel actions and regular stomach aches before going to school but not during school holidays. He occasionally vomitted after meals. The psychiatrist described him as "thin, underweight, unhappy and withdrawn". His weight at that time was above the 25th centile and his height on the 10th centile. No diagnosis was recorded but he was managed as an out-patient with imipramine (which he did not take at first) and with a cognitive–behavioural approach aimed at improving his eating habits.

He attended clinic sporadically through the year and was reviewed at the age of 14 when his weight was found to have dropped below the third centile. He was referred to a paediatrician whom he saw two months later, but then, and subsequently, refused physical examination and blood tests, saying he was "sick of all these stupid doctors". He agreed to be examined later in the year and the paediatrician noted clubbing. A chest X-ray was normal but he still refused blood tests.

He continued to be followed up in the paediatric clinic, and at the age of 15 at last consented to have blood taken. This showed a haemoglobin count of 10.8 g/dl (normal range 11.5–14.5); an erythrocyte sedimentation rate (ESR) of 7 mm/hour; an albumin level of 23 g/l (normal range 30–50); a serum B_{12} level of 386 ng/l (normal range 150–1000); and a serum folate level of 1.0 ng/ml (normal range 3–17). He was advised to come into hospital. A barium meal was administered, and follow-through showed appearances consistent with extensive small-bowel Crohn's disease, particularly affecting the jejunum.

He was treated with sulphasalazine and prednisolone with a poor response. He was transferred to the regional paediatric centre one month later and had a laparotomy which confirmed extensive jejunal Crohn's disease, which was resected. One week later he was well and was discharged home. Follow-up showed him to have remained well with

complete resolution of his depressive symptoms although his bizarre eating habits remained. He still did not require medication when he was transferred to the care of an adult gastroenterologist the following year.

Case 2

This boy was originally referred to a child guidance clinic by his headmaster at 10 years, 5 months of age. There was a two-year history of school refusal. He complained of abdominal pain and diarrhoea, but only on school days, and asked to be excused from games because of a "sore bottom". School refusal was confirmed, with a background of anxiety and parental marital discord. A family interview was offered but his father was not willing to attend. The psychiatrist did not perform a physical examination but contacted the family doctor and asked him to do a full physical examination "to rule out any gastro-intestinal problems" (see Table 1).

After the initial interview, the patient and his parents ceased to attend the clinic and were re-referred the following year with "psychological abdominal pain" and school non-attendance. The family doctor wrote: "were he an adult my diagnosis would be endogenous depression". The child psychiatrist did not agree and noted considerable anxiety symptoms and gastrointestinal symptoms related to term-time. He prescribed propranolol. Symptoms were improved at follow-up and he was discharged three months later.

A further referral was made to a child guidance clinic the following year at the age of 12 years, 6 months, for continuing school refusal and depressive symptoms. Meanwhile, growth failure had been noted by his family doctor. His height and weight had fallen to the third centile and he was found to have a bone age five years behind his chronological age.

He was referred to a paediatrician whose working diagnosis was growth hormone deficiency. He was referred to the regional centre where he was admitted for investigations at age 13. Blood tests revealed the following: haemoglobin 9.4 g/dl (normal range 11.5–14.5); albumin 17 g/l (normal range 30–50); ESR 59 mm/hour; platelets 804×10^9/l (normal range 150–400). A small bowel enema showed marked mucosal oedema and ulceration of the terminal ileum and caecum, consistent with Crohn's disease.

He was treated with an elemental diet (Elemental 028, Scientific Hospital Supplies, steroids and sulphasalazine) but over the following two years he had a number of relapses. A laparotomy and resection of terminal ileum and caecum when he was 14 provided histological confirmation of Crohn's disease and a relief of symptoms.

His disease remained quiescent at follow-up but he was referred to the hospital child psychiatric service because of headaches and persistent school refusal. He was admitted to the child psychiatry unit at age 15. His symptoms improved considerably but headaches were noted to worsen during periods of home leave. At this time, his father was unemployed and the general level of family hygiene was unusually poor.

A year later his father was charged with sexual abuse of all the children in the family, although our patient never made a disclosure. He continued to receive follow-up from both psychiatric and paediatric services until he left school and was finally discharged to adult medical follow-up at the age of 17 years.

Case 3

This boy was originally referred to a child guidance centre at the age of 12 years. He had a six-year history of intermittent abdominal pain, poor appetite and refusal to eat or drink anything except milk. Over the previous years his weight had dropped from the 25th to the 3rd centile.

His medical history started when he was six years old. Since then he had been extensively investigated for abdominal pain by physicians and surgeons. He had a laparotomy at 11 years of age, when a normal appendix was removed. During the operation an inflammatory mass was noted near the ileo-caecal valve.

Following assessment by the child psychiatrist he was started on a behavioural/dietary regime, and, after initial failure, was admitted to a child psychiatry unit at the age of 12. He put on 2 kg in weight on the ward regime but regularly lost weight while on weekend leave. He went home for Christmas leave, but did not return to the ward. He was followed up by child psychiatrists until the age of 13 years, and during this time he did not gain weight. He also saw a surgeon and another paediatrician privately.

He was referred to a child psychiatry unit, who noted his weight to be well below the third centile. Masses (presumed faecal) were present in the left and right iliac fossae, and there was generalised abdominal tenderness and low mood.

He was seen monthly and when there was still no weight or height gain, he was referred to the growth clinic in the regional centre. The initial diagnosis was growth hormone deficiency, and oxandrolone was prescribed which produced 'satisfactory growth acceleration'.

His abdominal pain continued and he was seen in the regional paediatric gastroenterology clinic at the age of 14 years, 7 months. He had an ESR of 55 mm/hour; an albumin level of 32 g/l (normal range 30–50); and a bone age four years behind his chronological age. A barium meal was administered, and follow-through showed appearances consistent with Crohn's disease in the terminal ileum and caecum.

After initial, unsuccessful treatment with an elemental diet (Elemental 028, Scientific Hospital Supplies) and steroids, he had an ileo-caecal resection aged 15 years, 6 months. Apart from a brief relapse one year later, he has remained well, with improvement of mood. He was transferred to an adult gastroenterologist at the age of 18. Although his height was still far below the third centile at the time of transfer, good catch-up growth continued.

Case 4

This girl had a long history of eating problems. A week after her birth she had a partial gastrectomy for duodenal ulceration and, as an infant, she had several admissions to hospital with feeding difficulties. From the age of one year she had recurrent infections, including abscesses, and she had pneumonia at the age of two years. She had been investigated for feeding difficulties and failure to thrive ever

since. At seven years of age she had a barium meal, which was reported as normal. An educational psychologist treated her for behavioural difficulties at school.

When she was nine years old, her father left home. Her parents divorced shortly afterwards and she remained with her mother. About this time she stopped eating and her weight, which had been on the third centile, dropped well below it. She stopped growing. Cyclical neutropenia was diagnosed after admission to hospital later that year. She was followed up in out-patients and, towards the end of her 12th year, her weight started to fall precipitously again. A child psychiatrist found disturbance of body image, primary amenorrhoea, and refusal to eat, and noted intermittent abdominal pain and constipation. Abdominal tenderness was noted on physical examination. A diagnosis of anorexia nevosa was made. She was admitted to a child psychiatry unit at the age of 13 and was started on a strict behavioural programme. She put on weight while she was in the ward but she lost weight when she was allowed home on leave.

At the age of 14 she was referred to the regional paediatric gastroenterology unit for further investigation of her failure to thrive. Investigations showed her to have an ESR of 98 mm/hour; an albumin level of 27 g/l; and a haemoglobin count of 11.9 g/dl. B_{12} and folate levels were within the normal range. Subsequent barium studies were consistent with Crohn's disease and she was started on an elemental diet (Elemental 028, Scientific Hospital Supplies).

She had three relapses within the next 18 months and eventually had a resection of her terminal ileum and ascending colon at the age of 15. Later in that year she was referred to the hospital child psychiatry service with nightmares and school difficulties but did not attend. She was referred again when she developed nightmares associated with anxiety and occasional episodes of visual hallucinations. These symptoms were temporally related to the onset of left ventricular failure secondary to a mitral valve lesion and stopped after surgical treatment. A computerised tomography scan of the brain was normal. Her weight was still well below the third centile and body image disturbance persisted; she said she would like to be "just a little bit heavier".

Controls

The notes of all patients in whom a diagnosis of Crohn's disease was made at the Children's Hospital, Birmingham, between 1987 and 1991 were reviewed to select controls. Nineteen patients were identified in whom the disease presented with predominantly gastrointestinal symptoms. The age at diagnosis and the duration of gastrointestinal symptoms were recorded together with anthropometric, haematological and biochemical data at diagnosis.

Analysis of data

Comparisons between subject and control groups were made using non-parametric statistics (Mann-Whitney test). Variables compared at diagnosis were haemoglobin count, platelet count, ESR, height for age, and weight for age. Height for age and weight for age were expressed as standard deviations from the mean (Z-scores) for that age. Height-for-age and weight-for-age Z-scores in the control group were compared with the National Center for Health Statistic's reference population (Hamill *et al*, 1979) using Student's t-test for correlated samples. Duration of symptoms between initial presentation to the family doctor and diagnosis was also compared.

Results

There was a significant delay between initial presentation and diagnosis in the subjects (median 24.5 months) compared with controls (median 6 months; $P<0.05$). Subjects also had significantly lower height-for-age and weight-for-age Z-scores ($P<0.05$) than controls. The controls themselves had significantly lower height-for-age Z-scores ($t=3.10$, $P<0.05$) and weight-for-age Z-scores ($t=2.35$, $P<0.05$) than the reference population. Subjects had lower serum albumin levels than controls (median 23.5 g/l v. 33.0 g/l; $P<0.05$). Subjects tended to have lower haemoglobin counts (median 9.95 g/dl v. 11.20 g/dl), higher ESRs (median 57 mm/hour v. 35 mm/hour) and higher platelet counts (median 614×10^9/l v. 424×10^9/l) than controls, although differences were not significant.

Discussion

These data indicate that the diagnosis of Crohn's disease presenting with predominantly psychiatric symptoms may be significantly delayed and that the delay is associated with even greater growth failure and depressed serum albumin at diagnosis than controls.

The four children presented illustrate that organic illness may present with predominantly psychiatric symptoms. The symptoms in our cases included low mood, anxiety, school refusal, bizarre eating habits and body image disturbance. All our cases had additional features suggestive of Crohn's disease (see Table 1). The severity of growth failure in our cases suggests strongly that Crohn's disease had been present for several years.

The previous literature in this area has focused on Crohn's disease presenting with anorexia nervosa. Gryboski *et al* (1968) described 11 adolescent girls admitted to a psychiatric hospital over ten years with eating difficulties. All 11 had psychological symptoms characteristic of a diagnosis of anorexia nervosa (disturbed body image, fear of fatness). Three had hypo-albuminaemia and anaemia and these were subsequently shown to have small intestinal Crohn's disease. No indication was given of physical or psychiatric outcome.

Jenkins *et al* (1988) presented three teenage girls and one woman who had received a diagnosis of anorexia nervosa, although two had no evidence of distorted body image. Crohn's disease was

Table 1
Clinical symptoms and findings in four cases of Crohn's disease at time of diagnosis

Case	Sex	Age at diagnosis of Crohn's disease: years	Age at presentation: years	Symptoms	Physical findings	Area of involvement	Haemoglobin count: g/dl (normal range 11.5-14.5)	Platelet count: $\times 10^9$/l (normal range 150-400)	Albumin level: g/l (normal range 30-50)	ESR: mm/h
1	Male	15	13	Abdominal pain Weight loss Diarrhoea Depression Bizarre eating habits	Growth failure Clubbing Tender abdomen	Jejunum	10.8	500	23	7
2	Male	12	10	Abdominal pain Diarrhoea Anxiety Depression School refusal "Sore bottom"	Growth failure	Terminal ileum Caecum	9.4	804	17	59
3	Male	14	12	Abdominal pain No appetite Low mood	Growth failure Masses in iliac fossae Tender abdomen	Terminal ileum Caecum	12.5	457	32	55
4	Female	14	12	Abdominal pain Constipation Weight loss Amenorrhoea Body image disturbance	Growth failure Tender abdomen	Ileum Ascending colon	11.9	624	27	98

subsequently diagnosed after initial psychiatric treatment. All the cases had markedly abnormal biochemical and haematological markers including raised ESR and platelet count and reduced albumin and haemoglobin. In one of these patients, a recurring psychosis coincided with further exacerbations of the Crohn's disease, perhaps as a result of steroid treatment.

Even among physical illnesses, Crohn's disease can be difficult to diagnose. A *Lancet* editorial (1975) described it as "ousting syphilis as the great imitator". When growth failure was identified in two of our children, they received growth hormone treatment before Crohn's disease (certainly the cause of their growth failure) was recognised.

It is debatable whether or not earlier diagnosis and treatment of the Crohn's disease in our cases would have affected outcome. Treatment of Crohn's disease is largely symptomatic. Although some observations suggest that earlier diagnosis of Crohn's disease does not greatly affect outcome, Griffiths *et al* (1991) have suggested an improved prognosis over the following eight years for those who had earlier resections of diseased bowel. This is potentially important, as all our cases required resection after diagnosis. Furthermore, an earlier diagnosis of Crohn's disease would have led to effective symptomatic treatment including correction of growth failure. Moreover, the emphasis of psychiatric management would have been fundamentally different.

Retrospectively, there were several reasons for the delay in diagnosis of Crohn's disease. It was assumed that the psychiatric diagnosis precluded an organic explanation of the child's physical symptoms. Some of the subjects came from families with marked interpersonal difficulties which reinforced paediatric and psychiatric options that dynamic problems were of primary aetiological significance. Two of the subjects gained weight on the ward regime but lost it again on weekend leave, a factor which further reinforced those opinions.

In case 1, doctors complied with the child's unwillingness to be examined or investigated and the diagnosis was delayed further. In two cases the psychiatrists assumed that others would "rule out physical causes" but did not follow this up.

Crohn's disease may lead to secondary psychiatric disturbances. Bruce (1986) reviewed the literature and suggested that 60% of children and adolescents with Crohn's disease had recognisable psychiatric morbidity, compared with 17% of controls. He noted that siblings of affected individuals have a higher than expected psychiatric morbidity. He also described his clinical observation that children with Crohn's disease may experience exacerbations of physical symptoms at times of emotional distress. Our own observations bear this out. In two of our patients, gastrointestinal symptoms worsened at times of stress.

It is possible that all our patients' early symptoms were due to pre-existing psychiatric disorder and were independent of Crohn's disease. This seems highly unlikely but difficult to disprove. Many of the presenting symptoms and signs could be attributed to Crohn's disease and they resolved completely when it was adequately treated. For example, all our patients experienced low mood which, in retrospect, was probably related to pain, diarrhoea, malnutrition or to dynamic factors to do with illness. Depression has been associated with Crohn's disease in other studies. Helzer *et al* (1983) compared 50 subjects with Crohn's disease with 50 controls with chronic illness for prevalence of a number of psychiatric diagnoses. A diagnosis of depression was the only one of several diagnoses significantly more prevalent in the subject group.

Symptoms which persisted after treatment included bizarre eating habits (in case 1) and school refusal (in case 2). One of our patients (case 4) had a demonstrable fear of fatness and distorted body image which was present before and during treatment for Crohn's disease. In her case, Crohn's disease and anorexia nervosa probably co-existed. The implications for management are the same whether the children's symptoms were the earliest presentation of Crohn's disease or whether the insidious onset of Crohn's disease provided a new cause for the continuing symptoms of pre-existing psychiatric disorder.

Features of a history and examination suggestive of Crohn's disease, rather than primary psychiatric illness, include mouth ulcers, perianal disease, joint pain, erythema nodosum, abdominal masses, low height for age (or fall of height percentile), or low weight for height. Inflammatory markers (e.g. ESR, C-reactive protein) and platelets may be raised (Jenkins *et al*, 1988) and there may be more specific evidence of malnutrition (e.g. B_{12}, folate, iron and zinc deficiencies and hypo-albuminaemia) (Booth, 1991).

Hypoalbuminaemia is rare in children and adolescents with eating disorders. Palla & Litt (1987) described the medical complications of eating disorders in 65 adolescents with diagnoses of anorexia nervosa or bulimia nervosa. All subjects had serum albumin within the normal range. Moreover, ESR measurements in the subjects were low, with a mean ESR of 4 mm/hour for anorexics and 8 mm/hour for bulimics.

Serial reassessment of physical status is important in all children with a psychiatric diagnosis whose key symptoms are physical. Particular attention should

be paid to serial measurements of height and weight plotted on centile charts, physical examination and inflammatory markers. Continued cooperation between paediatricians and child psychiatrists is essential for successful diagnosis and management of these children.

Little is known about the eating habits of children with Crohn's disease, and a comparison of their eating behaviours with those of children with eating disorder without known organic pathology represents an intriguing possibility for future investigation.

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'Dyschronia' in a Patient with Tourette's Syndrome Presenting as Maternal Neglect

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We report an unusual and unsuspected cause of maternal neglect in a patient with Tourette's syndrome. An important cause of the behaviour appears to have been a form of dyscalculia characterised by a complete inability to appreciate the passage of time. To our knowledge this is the first case of its kind to be reported.
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Case report

A 23-year-old unmarried woman and her five-month-old baby son were referred to us by their local social services department for an assessment of the mother's ability to care for her child. She was the only daughter of elderly parents, and the family were isolated and inward-looking. They considered themselves psychic and took a practising interest in the occult. Her own birth had been complicated by a difficult labour. At the age of eight she was diagnosed as having Tourette's syndrome, with both motor and vocal tics and coprolalia, and was known in the family as the 'devil child'. She received no specific treatment. From her late teens she had been virtually free of symptoms. However, she had never held down a job and remained living at home. She had a history of impulsive and occasionally violent behaviour. When she became pregnant, she concealed the fact and only presented to the local medical services at 37 weeks.

Her baby was delivered in hospital weighing 3.2 kg. An obsessional concern with the sterilisation of bottles and the