

# AN UNUSUAL INFECTIOUS ILLNESS WITH AN UNUSUAL RASH

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Within a period of 2 months, 4 children suffering from a most unusual and bizarre infectious disease were encountered. The clinical picture at first suggested Coxsackie A virus infection, but as the disease pattern developed, it became apparent that this illness was different to any viral illness I had previously seen. All 4 cases presented a strikingly similar picture and once seen, it became possible to foretell the pattern of events in subsequent cases.

## CASE REPORTS

### Case 1

D.S., a male infant of 15 months, was admitted to hospital with a history of vomiting and refusing feeds for 1 day. Two days after admission he developed diarrhoea. This settled slowly over the subsequent few days; his treatment consisted of a diet and a short course of antibiotic and he was discharged 12 days after admission.

During his first period in hospital it was noted that he was extremely reluctant to feed and this became aggravated at home. For this reason he was re-admitted some 4 days later in an extremely irritable state. No other abnormalities were noted on clinical examination at this time, and lumbar puncture produced a normal fluid under normal pressure.

The symptoms continued in hospital and 2 days after admission he developed an ulcerative stomatitis involving the buccal and lingual mucosa. As a result of his refusal to feed and a recrudescence of his vomiting and diarrhoea, it became necessary to institute intravenous therapy, which had to be continued for 6 weeks. The stomatitis subsided after 10 days, but despite this he continued to refuse to take food. His temperature, which had been elevated on both admissions, continued to hover between 100 and 103°F.

Four weeks from the onset of the illness the pyrexia subsided, only to become elevated again 5 weeks from onset, when he developed a rash which began on the trunk and later moved to the face and extremities. The rash was at first red in colour and macular, but later became papular and hyperkeratotic, taking 6-7 days before it faded.

The acute infectious illness now subsided, but he was left in a dystrophic state, which continued for a further period of approximately 4 weeks, despite vigorous therapy, including the administration of anabolic agents, steroids and antibiotics. In this state he developed a mastoiditis which required drainage. The diarrhoea did settle eventually and he recovered. No infectious agent was recovered from his stool or blood.

### Case 2

G.S., a male infant aged 15 months, was admitted to hospital in a moderately dehydrated state as a result of diarrhoea and vomiting which he had had for a week. He was given intravenous fluids and a short course of antibiotic and put on a diet. The diarrhoea gradually settled and he was discharged 1 week later.

As with the first case, it was noted during his initial admission that he was reluctant to feed. This symptom became aggravated at home and he was re-admitted 4 days later, having again developed mild diarrhoea and vomiting. Intravenous therapy became necessary from the 3rd day of re-admission. Lumbar puncture done at this time showed no abnormality.

His temperature remained intermittently raised and the diarrhoea slowly settled. At 4½ weeks from the onset of the disease he developed a rash completely similar to that of case 1 (Fig. 1). Once the rash had developed his temperature subsided and he recovered, the rash taking 6 days to fade. No infectious agent was recovered from his stool.

### Case 3

K.M., a male infant of 8 months, was admitted to hospital with moderately severe diarrhoea and vomiting which he had had for 4 days. He was put on a diet and given a short course of antibiotic. He improved gradually, being discharged 1 week later, although he was reluctant to feed. This symptom became aggravated and when forced, he vomited. For this reason he was re-admitted to hospital some 5 days after discharge in an extremely irritable state. Lumbar puncture produced a completely normal fluid.

Refusal to feed and, to a lesser extent, vomiting and diarrhoea with intermittent pyrexia, persisted and 2½ weeks



from the onset of the disease intravenous fluids became necessary. The gastro-intestinal symptoms subsided slowly but the temperature continued, and with the reluctance to feed it became necessary to feed him intravenously again some 4 weeks from onset. Following this he appeared to improve and



Fig. 1. Distribution of the rash on the trunk (see text).

pyrexia subsided. At 5½ weeks from the onset of the disease his temperature rose and a rash similar to that seen in cases 1 and 2 appeared and lasted for about 60 hours. At this stage he improved and recovered from the illness. Neither bacteria nor viruses were recovered from culture of blood and stools.

#### Case 4

H.P., a male infant of 5 months, was admitted to hospital for investigation of suspected blindness and delayed milestones of development. Two days after admission he developed severe vomiting and diarrhoea and became severely dehydrated. An intravenous drip was set up and had to be continued for 4 weeks. He was given a short course of antibiotic at the onset of the disease.

The diarrhoea, vomiting and pyrexia continued, but most remarkable was his refusal to feed. At 4 weeks from onset he developed a mild loose cough and a week later a rash, similar to that seen in the first 3 cases, appeared and lasted about a week. Six weeks from the onset of his first symptoms he began to improve and after 7 weeks he was discharged, having fully recovered. No infective agent was isolated from his blood or stool.

The clinical and laboratory features of the 4 cases are summarized in Tables I and II.

#### DISCUSSION

All 4 cases described pursued a strikingly similar course and once the rash developed, it became apparent that

TABLE I. CLINICAL FEATURES

					No. of cases
Refusal to feed and irritability	..	..	..	..	4
Diarrhoea and vomiting	..	..	..	..	4
Intermittent pyrexia	..	..	..	..	4
Rash	..	..	..	..	4
Ulcerative stomatitis	..	..	..	..	1

TABLE II. LABORATORY INVESTIGATIONS

	Case 1	Case 2	Case 3	Case 4
Blood count	leucopenia	normal	leucopenia	leucopenia
Urine examination	normal	normal	normal	normal
Blood culture				
Bacteria	—ve	—	—	—
Viruses	—ve	—	—ve	—ve
Stool culture				
Bacteria	—ve	—ve	—ve	—ve
Viruses	—ve	—ve	—ve	—ve
Urine culture				
Bacteria	—ve	—ve	—ve	—ve
Viruses	—	—	—	—ve
CSF	normal	normal	normal	normal
Herpes antibodies	—ve	—	—	—
Toxoplasma antibodies	—ve	—	—ve	—ve
Leptospira antibodies	—ve	—	—ve	—ve
Rickettsial antibodies	—ve	—	—	—ve
Paul Bunnell	—ve	—	—ve	—ve
Urea and electrolytes	normal	normal	normal	normal
Serum proteins	normal	—	—	normal

these were all instances of the same disease.

There were 2 distinct phases to the illness, the first consisting of pyrexia, diarrhoea and vomiting and lasting about 10 days. Three of the 4 children were then discharged home but refusal to feed, which was evident from the onset, continued and re-admission to hospital became necessary within 3-4 days.

The second phase of the illness was also associated with intermittent pyrexia and lasted for about 4 weeks. During this period, irritability increased and vomiting and diarrhoea again appeared. However, when intravenous therapy became necessary it was mainly due to a refusal to feed. A loose cough also became evident at this stage. Between 4½ and 5 weeks from the onset of the illness, a characteristic rash appeared (Fig. 1). Initially the rash involved the chest and abdomen, spreading later to the back and eventually to the face and extremities. At first macular and morbilliform, the rash became papular and hyperkeratotic and lasted about 6-7 days. When the rash disappeared, the illness which had lasted about 6 weeks appeared to subside.

All 4 children were severely ill and one indeed became dystrophic, although he recovered eventually. Although no virus was recovered from the urine, stool or blood, it is felt that this may well be an example of a viral illness, which although seen in the same hospital, certainly developed in 3 of the 4 cases before admission.

#### ADDENDUM

I have seen 3 further cases of this disease since encountering the 4 described. The pattern of the disease was strikingly similar in all. It has been noted further that when these children begin to feed, the change from a state of anorexia is sudden and it is difficult to satisfy their appetites.