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Intussusception in adults complicating specific inflammatory diseases of the intestine

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EDITORIAL SYNOPSIS  A patient with intussusception occurring in the course of a Salmonella typhi-murium infection is described, and a review given of the literature of intussusception as a secondary complication to inflammatory diseases of known origin.

Intussusception in the adult is a comparatively rare surgical emergency. Sanders and Kinnaird (1959) were able to trace 1,253 cases in the English and the American literature between 1892 and 1959 and since that time a further 126 cases have been reported. Brayton and Norris (1954) calculated that cases of adult intussusception formed only 0.003 to 0.02% of hospital admissions. Only about 10% of cases of intussusception are seen in patients over the age of 14, and these adults differ from the better known picture in infants in two principal respects. First, in infants intussusception is usually an acute episode. In adults, however, there is no characteristic syndrome, the early stages often being subacute and atypical, the condition pursuing a chronic course, and only later ending in an attack of acute intestinal obstruction. Secondly, 95% of cases in infants are idiopathic whereas in adults about 85% of cases are secondary to some demonstrable lesion, 67% being due to a tumour.

CASE REPORT

Mr. F., aged 34, was admitted as an emergency to Cardiff Royal Infirmary on 9 June 1960, under the care of Mr. A. C. Lysaght. He gave a history of severe colicky abdominal pain for the past 18 hours preceded by malaise, shivering, and headache. He had vomited once and passed a single dark tarry motion, his bowels having previously been normal. There was no other relevant history, and no similar previous episodes.

On examination he looked ill and was obviously in pain (pulse 90/min., temperature 99.4°F). The tongue was dirty with considerable halitosis but there was no clinical evidence of dehydration.

There was a visible tender swelling in the right iliac fossa, about the size of an orange, but no generalized tenderness or distension. There was nothing abnormal to be felt on rectal examination but the examining finger was stained with bright red blood.

A diagnosis of intussusception was considered, and arrangements made for a laparotomy to be performed.

Under general anaesthesia the abdomen was explored through a right lower paramedian incision. An ileocaecal intussusception was found which was fairly easily reducible. After reduction the terminal ileum did not appear abnormal, but the caecum felt very bulky, and in the belief that this was due to a benign tumour, it was amputated with the appendix, and the caecal stump oversewn. The wound was closed in the usual way, a drain being placed down to the caecal stump.

Subsequent examination showed that the ‘tumour’ consisted of oedematous mucosa only, this being over an inch thick, and later microscopical examination showed an area of infarction with acute inflammatory changes.

The patient’s post-operative course was extremely stormy. The abdominal pain persisted and within 48 hours he developed a profuse watery diarrhoea, passing 3 to 4 litres of offensive fluid daily for the next two weeks and similarly large amounts of gastric fluid were aspirated. Stool culture on several occasions showed the persistent presence of Salmonella typhimurium. He was treated with chloramphenicol, intravenous fluids, and gastric aspiration for the following two weeks. Ten days post-operatively he had a sudden episode of acute abdominal pain, tenderness, distension, hypotension, and a rapid rise in pulse rate. A clinical diagnosis of perforation was made, but in view of his precarious general condition, this was treated conservatively, and so no proof was obtained.

After two weeks he began to improve slowly, and had no further complications. His subsequent recovery was uneventful and he was discharged to a convalescent home on 6 July, four weeks after the original episode. Since that time he has been in good health and has had no further bowel symptoms.

DISCUSSION

Intussusception resulting from an acute inflammatory disease of the bowel is not uncommon in
children. Gross (1953) states that 'it is well recognized that intussusception occasionally makes its appearance during or shortly after acute enteritis, at which time the disturbed intestinal peristaltic movement may set the stage for the prolapse of one segment into another'. This, however, seems to be very uncommon in adults, particularly in more recent years. Thus, in a review of 745 adult cases reported in the English and American literature, in a series collected by Brayton and Norris (1954), only 16 were due to local inflammatory disease (typhoid, etc.) of the bowel, that is, 2-2% compared with 54% due to a tumour. Roper (1956), reviewing a further 134 cases, found 5-2% due to local inflammatory causes and 68% secondary to a tumour. These recent figures show a remarkable change from the 298 cases collected by Elliot and Corscadden in 1911. At that time 9-8% were associated with an inflammatory lesion but only 34-2% were secondary to a tumour. This change that appears to have taken place has of course occurred during a period in which malignant disease of the bowel has become more common and the incidence of such inflammatory diseases of the bowel as typhoid and dysentery has decreased.

In view of the unusual nature of the cases reported above, a search was made in the English and American literature of the last 70 years for similar cases. Thirty-seven cases associated with typhoid or with bacillary or amoebic dysentery were traced and in addition nine cases associated with tuberculosis and one each of regional enteritis, ulcerative colitis, and acute colitis. These collected series showed certain features peculiar to each specific cause.

**Typhoid (Enteric Fever)** Fourteen cases of intussusception occurring in the course of typhoid fever have been traced (Ash, 1902; Bryant and Bragg, 1905; Harte and Ashurst, 1904; Moreton, 1920; Ross and Page, 1907; Pernet, 1873; Quadflieg, 1901; Roper, 1956; Ross, 1905; Smith, 1909; Vincent, 1895; Watkins Pitchford, 1902). It is a rare complication. Huckstep (1960) did not see it in over a thousand cases of typhoid in Kenya, and does not mention it as a surgical complication of typhoid, even though intussusception is a comparatively common condition in adult Africans (Kark and Rundle, 1960). It usually arises as an acute episode late in the course of the disease, often in the convalescent phase, and in contradistinction to other surgical complications such as perforation, the prognosis with surgery is fairly good; seven out of nine cases submitted to laparotomy survived. It almost invariably starts in the small bowel, and may be associated with the presence of enlarged Peyer's patches. Diagnosis is difficult, as the features are similar to those of a perforation (Moreton, 1920). Both Ash (1902) and Ross (1905) in discussing the surgical complications of typhoid mention intussusception as an important differential diagnosis of perforation.

**Amoebic Dysentery** Eighteen cases of intussusception complicating amoebic dysentery have been found in the literature (Collins, 1939; Joly and Thomas, 1954; Kark and Rundle, 1960; Parry, 1945; Reddy and Rangam, 1946; Theron, 1947) and the features of this complication have recently been well reviewed by Kark and Rundle (1960). Unlike the picture in typhoid, intussusception occurs early in the course of an attack of amoebic dysentery, and the diagnosis is likely to be difficult, since amoebiasis itself is commonly associated with diarrhoea, mucus, blood, and sometimes a mass. When these are associated with colicky pain, the diagnosis of intussusception should be considered, and confirmation may be obtained by barium enema. The prognosis with surgery is again fairly good, there being 13 survivors in the 18 reported cases.

In countries where amoebic dysentery is common, as in Africa and India, large bowel carcinoma is a rare condition, and amoebiasis or an amoeboma is frequently suggested as a common cause of intussusception. Collins (1939) stated 'in view of the rarity of carcinoma of the colon, and the frequency of dysentery, it is possible that in tropical countries intussusceptions of the colon are more often the result of the latter condition than of the former'. However, though both amoebiasis and intussusception are common, the association of the two is still comparatively rare. Joly and Thomas (1954) found two cases of amoebiasis in 33 cases of intussusception, Kark and Rundle (1960) only five in 67 cases.

**Bacillary Dysentery** The picture in bacillary dysentery is quite different. It is now very rare for intussusception to occur, and no cases of intussusception in adults with this disease have been reported since 1908. At that time, of the six cases reported, five died, the only survivor having undergone laparotomy and lateral anastomosis (Maxwell, 1908; Muller, 1879; Pridmore, 1897).

A clinical condition closely related to dysentery is 'summer diarrhoea'. Perrin and Lindsay (1921) commented on the rarity of intussusception in summer diarrhoea in contrast to its frequent association with other gastrointestinal disturbances where lymphoid tissue is swollen.

**Tuberculous Enteritis** Though not specifically an acute inflammatory process, nine cases are included...
of intussusception complicating tuberculosis. As might be expected it may occur quite late in the course of the disease, and almost invariably affects the ileocaecal region. Of the nine cases reported, five survived following surgery; in one the outcome was not stated (Barrow, 1897; Brin, 1908; Cavaillon, 1901; Maxwell, 1908; Rydgyer, 1896).

**OTHER RARE INFLAMMATORY CAUSES** Ferrer (1950) reported a case associated with acute colitis but gave no details, and Tranisi and Parrillo (1958) successfully treated one occurring in regional enteritis. Bargen, Kerr, Hausner, and Weber (1937) found a case occurring in ulcerative colitis with secondary polypoidal changes, which was successfully reduced by enema. They could find no similar cases in the literature.

Ross and Page (1907) reported a case occurring in a man aged 27 due to 'ptomaine poisoning' after eating decayed crab. This may be of a similar aetiology to the case reported here, but, apart from this, there are no other cases occurring in acute epidemic gastroenteritis or Salmonella food poisoning. The rarity in adults is a little difficult to understand since there are frequent comments on its occurrence in children (Gross, 1953).

**AETIOLOGY** As in intussusception arising in other circumstances, in acute inflammatory disease a combination of a 'tumour' in the bowel wall associated with a disorder of normal bowel movements probably initiates the intussusception. Since Perrin and Lindsay's original paper in 1921, swollen aggregations of lymphoid tissue have been incriminated as the 'tumour' concerned. Certainly in cases occurring in typhoid, swollen Peyer's patches and typhoid ulcers have been mentioned as being present at the apex of the lesion (Bryant, and Bragg 1905; Ross and Page, 1907) though in the case reported by Vincent (1895), which occurred during the convalescent phase of typhoid, 'no typhoidal lesion was present in the bowel—all had repaired without trace, and no tumour, polyp or contraction was present'. Perrin and Lindsay (1921) went on to comment on the rarity of intussusception in summer diarrhoea, a condition closely related to dysentery, pointing out that in this condition lymphoid patches are actually decreased in size, a feature they ascribe to dehydration. This may also be the case in dysentery, accounting for the comparative rarity of associated intussusception. In amoebic dysentery the tumour concerned is in some cases an ulcer or an amoeboma (Reddy and Rangam, 1946) though here, as in other conditions there may be no demonstrable localized tumour.

Wangensteen (1942) pointed out that the growth curve for lymphoid tissue reaches its maximum at the age of 11, and thereafter the relative size of lymphoid follicles decreases. This might provide an explanation of the increasing rarity of intussusception as a complication of inflammatory diseases in adolescents and adults, though it does not provide an explanation for its comparative rarity in children about the age of 11.

For many years the occurrence of intussusception has been associated with abnormalities in bowel movements (Hunter, 1793; Treves, 1884; Nothnagel, 1884), a contraction ring becoming an intussusceptum into a distal relaxed or paralysed segment. Though there is little experimental evidence to confirm it, it seems likely that inflamed mucosa could stimulate a localized area of bowel contraction. With this factor in mind it becomes easier to understand the time of the incidence of intussusception in typhoid. In the early stages of the disease the bowel is swollen and oedematous, and invagination is presumably difficult for mechanical reasons. In the second week the bowel is dilated and paralysed, and colic is typically absent. Intussusception occurs after this phase, at the time when the previously distended and paralysed bowel is beginning to recover its function, a process which, it is understandable, is likely to occur in an irregular manner. Since the lesion occurs in a recovering bowel, this also provides some reason why the prognosis for surgery in intussusception is better than that for perforation. Similar conditions presumably apply in dysentery; in fact Griesenger is quoted by Treves (1884) as having shown that initial paralysis of the bowel wall is not uncommon. This does not apply in gastroenteritis, as in the case reported here, where colic is an early feature of the disease.

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