Intractable diarrhoea of infancy and latent otomastoiditis

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SUMMARY In 16 infants with intractable diarrhoea, latent otomastoiditis was found in 9 (3 at necropsy and 6 at myringotomy-antrotomy). In 5 of the 6 operated group, surgery was followed by a striking cessation of the diarrhoea and with weight gain. It is concluded that (1) latent otomastoiditis may be a perpetuating factor in intractable diarrhoea; (2) myringotomy-antrotomy should be considered if other forms of treatment have failed, and especially if there is leucocytosis; (3) mastoiditis with diffuse osteitis seems to be associated with a poor prognosis.

The association of ear infection with diarrhoea and malnutrition in infancy has been known for more than a century,¹ but it was only from the late 1920s that articles began to appear in the medical journals drawing attention to the relation between acute, or latent, otomastoiditis and diarrhoea, either acute or prolonged, with severe malnutrition.^{2–11} With the advent of sulphonamides and other antimicrobial drugs, most of the ear infections were able to be controlled, and, in the course of time, the concept of latent otomastoiditis has almost been forgotten. In two review articles on intractable diarrhoea of infancy,^{12–13} latent otomastoiditis was not mentioned, nor is it mentioned in a well-known and much used book of paediatric gastroenterology.¹⁴ The aim of this paper is to recall attention to a problem which seems far from resolved.

Patients and methods

16 infants with intractable diarrhoea were treated between January 1977 and July 1979 in this unit. 'Intractable' diarrhoea was defined by the criteria of Avery *et al.*¹⁵ but with an age limit of up to 3 months. Mean age was 41 (range 7–120) days, and mean weight on admission 2.90 (range 1.80–4.58) kg. Each infant required total parenteral nutrition from a minimum of 11 to a maximum of 95 (mean 43) days; many of them were also treated with hydrocortisone and cholestyramine. Each received between 3 and 5 antibiotics during his long stay in hospital; these were given either alone or in combination. Hydrocortisone was used mainly during periods of severe clinical deterioration. Treatment with

antibiotics was instituted for intercurrent infections, if corticosteroids were given, or if an infection was strongly suspected on clinical grounds. None of the infants had been given breast milk with the exception of Cases 11 and 16, who received some breast milk together with formula for the first 1½ and 3 weeks of life. Each infant had at least one blood culture (maximum 11), three stool cultures (maximum 10), and two urine cultures (maximum 4). The material obtained at surgery from the ear cavities of Cases 11, 12, 13, 15, and 16 has been cultured.

Results

Four infants (Cases 2, 3, 4, and 7) had an uneventful recovery after the institution of total parenteral nutrition. Two (Cases 5 and 6) died unexpectedly from complications of unsuspected duodenal ulcer—perforation and massive bleeding.

Ten infants (Cases 1 and 8–16) had very protracted clinical courses and treatment seemed to have no effect on either the diarrhoeal or the nutritional condition; there was a leucocytosis in 8 of them. Three of the 10 (Cases 1, 8, and 9) died, and necropsy showed the existence of bilateral purulent mastoiditis; earlier otoscopies had been repeatedly normal. Because of this it was decided to submit each of the remaining 7 infants (in whom diarrhoea and malnutrition were equally resistant) to bilateral myringotomy, and if no pus was encountered, to follow with bilateral antrotomy. This was done despite completely normal otoscopies. Two infants died: one (Case 14) died 10 hours after an antrotomy which had shown mastoiditis on the right side with

Fig. 1 Weight charts of 10 infants with protracted diarrhoea resistant to medical treatment. Cases 10-13 and Cases 14-16 were subjected to myringotomy and antrotomy.

osteitis: the other (Case 15) died 38 days after antrotomy which had shown bilateral mastoiditis with diffuse osteitis (a left-sided mastoiditis was still present at necropsy). Five infants (Cases 10-13 and 16) recovered; clinical improvement immediately followed myringotomy-antrotomy in the first four and was followed after some delay in the remaining infant. In Case 10 no lesion was found on antrotomy; three (Cases 11, 12, and 16) had unilateral mastoiditis (plus diffuse osteitis in one case); one (Case 13) had bilateral otitis media. After the myringotomy-antrotomy procedure in these five infants, the diarrhoea cleared in 1-4 days, and the fever (present in two) by the next day; weight improved rapidly in all except Case 16, in whom improvement was more delayed (Fig. 1); and the number of leucocytes progressively fell to normal (Fig. 2). Stool, blood, and urine cultures were all consistently negative. Cultures from the ear cavities showed a *Klebsiella* sp. in Case 11, a *Staphylococcus aureus* coagulase-positive in Cases 12 and 13, and were negative in Cases 15 and 16.

Discussion

The frequent coexistence of acute or protracted diarrhoea in infancy with parenteral infection,

otitic or other, is a fact well known to paediatricians, although the relation of the two remains obscure. The diagnosis of otitis media is usually made by otoscopy, while treatment with antibiotics resolves most cases. But in young infants and particularly in those severely malnourished, otoscopy may be unreliable: an infected middle ear, antrum, or mastoid may exist without discernible local signs.¹⁴ This is the condition that has been termed latent otomastoiditis. In relation to diarrhoea, the occult ear infection may be primary, secondary, or simply accompanying, but in any event, once installed it may perpetuate its course.^{7 17}

Some 40 years ago many paediatricians did not hesitate to advise myringotomy or even antrotomy for a marasmic infant with prolonged and resistant diarrhoea. With the advent of potent antimicrobial drugs this problem has substantially changed, and references to it have not appeared in the columns of Western medical journals, at least during the last 10 years. This contrasts with the situation in some east European countries, where the importance of ear infection with infantile diarrhoea, and of antrum drainage in its treatment, seems to persist.¹⁷⁻²⁰

In 16 infants successively admitted to our unit with malnutrition and intractable diarrhoea, latent

Fig. 2 Leucocyte counts in the 10 infants with protracted clinical course and resistance to medical treatment.

otomastoiditis was found in nine. In the remaining infant (Case 10) no otitic infection was detected, but diarrhoea and fever ceased, and weight gain started immediately after antrotomy. Similar beneficial effects of surgery, even in the absence of manifest infection, have been reported.^{4,11} Of the 3 infants in whom mastoiditis was associated with osteitis, two (Cases 14 and 15) died, and one (Case 16) had a delayed recovery after surgery, suggesting that diffuse osteitis may be associated with a poor prognosis. Latent otomastoiditis has proved to be remarkably uninfluenced by antimicrobial therapy; indeed it has been claimed that antibiotics may actually worsen the clinical condition of infants with

intractable diarrhoea.¹⁵ Myringotomy and antrotomy are simple procedures in the hands of a skilled otologist, and we suggest that the procedure should be considered after other treatments of the diarrhoea have failed.

The fact that so few of these infants were breast fed contrasts with the 53 and 30% incidence found in infants from 1 and 2 months in Lisbon,²¹ suggesting that human milk may have some effect in protecting the infant against intractable diarrhoea.

Conclusions

Latent otomastoiditis may be a perpetuating factor

in intractable diarrhoea in infants. Myringotomy-antrotomy should be considered after other forms of treatment have failed, and especially if a leucocytosis is present. Simple 'ventilation' by surgery of apparently intact ear cavities may favourably influence the course of the disease. Mastoiditis with diffuse osteitis seems to be associated with a poor prognosis.

Addendum

Since submitting this paper four more infants have presented with intractable diarrhoea and latent otomastoiditis. Antrotomy showed bilateral mastoiditis in each, with osteitis in two. Cultures from the ear cavities grew a *Klebsiella* sp. in one and were negative in two. After surgery, diarrhoea stopped within 4 days and weight improved rapidly.

References

- Parrot J. Note sur l'otite de l'oreille moyenne chez le nouveau-né. *Bull Soc Med Hop Paris* 1869; 6: 82-4.
- Jeans P C, Floyd M L. Upper respiratory infection as a cause of cholera infantum. *JAMA* 1926; 87: 220-3.
- Spahr M B. The bacteriology of mastoiditis in infants. *Am J Dis Child* 1929; 37: 541-5.
- Burghi S E. Rinofaringitis aguda simple en la infancia. *Arch Pediatr Uruguay* 1938; 9: 653-98.
- McConkey G, Couper E C R. Otitis media and mastoiditis in infancy. *Arch Dis Child* 1938; 13: 137-44.
- Oreggia J C. La otomastoiditis del lactante. *Arch Pediatr Uruguay* 1939; 10: 585-604.
- Schneegans E. L'otite et l'antrite du nourisson et leurs répercussions sur l'état digestif. *Rev Fr Pediatr* 1939; 15: 425-56.
- Couper E C. The Schilling haemogram in otitis media and mastoiditis of infancy. *Arch Dis Child* 1940; 15: 27-42.
- Penichet G C, Castilho B D, Hernández F B. Otitis del lactante. *Bol Soc Cubana Pediatr* 1941; 13: 9-29.
- Lévesque J, Perrot R, Renault. La ponction de l'antrite dans le diagnostic des oto-mastoidites des nourissons (abstract). *Arch Fr Pediatr* 1942; 1: 100-2.
- Salazar de Sousa C. A oto-mastoidite do lactente. *Rev Port Pediatr Puer* 1943; 6: 203-24.
- Sunshine P, Sinatra F R, Mitchell C H. Intractable diarrhoea of infancy. *Clin Gastroenterol* 1977; 6: 445-61.
- Bransky D. Intractable diarrhea of infancy. In: Lenbenthal E, ed. *Digestive diseases in children*. New York: Grune & Stratton, 1978: 351-65.
- Roy C C, Silverman A, Cozzetto F J. *Pediatric clinical gastroenterology*, second edition. St Louis: Mosby, 1975: 197.
- Avery G B, Villavicencio O, Lilly J R, Randolph J G. Intractable diarrhea in early infancy. *Pediatrics* 1968; 41: 712-22.
- Martinho da Rocha J, Fortes H. *Propedêutica Pediátrica e Semiologia Infantil*. Rio de Janeiro: Livraria Atheneu, 1958: 318.
- Prîșcu R, Maiorescu M, Rociu O, Petrescu C. Considerations of present aspects in infantile otomastoiditis (in Rumanian). *Pediatria (Bucuresti)* 1969; 18: 481-94.
- Gurdzil Z, Modzelewska I, Pliszczynska-Brennenstuhl M, Czapska M. Analysis of indications of antrotomy in infants and its results in the light of personal observations from the period 1965-1974 (in Polish). *Wiad Lek* 1975; 28: 1963-9.
- Yevdoshchenko E A, Melnik A M. Toxicosis resulting from oto-antritis (in Russian). *Zh Ushn Evos Gori Bol* 1976; 2: 70-5.
- Dyachuk V V. The course of otoantritis in chronic nutritional disturbances in nurslings (in Russian). *Vestn Otorinolaringol* 1978; No 4, 68-72.
- Videira Amaral J M, Espinosa L, Lopes W. Alimentação com leite materno na área urbana de Lisboa. *Rev Port Pediatr* 1979; 10: 155-78.

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Commentary

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Intractable diarrhoea of infancy as defined by Avery *et al.*¹ is a fairly rare but a disproportionately resource-consuming problem in the UK. Although many contributory factors have been identified, the underlying aetiology is often obscure and management is reduced to life support by intravenous feeding and gradual weaning on to a hypoallergenic diet when possible. The mortality rate is high.

I have no personal experience of an association between this condition and subclinical mastoiditis or antral infection. It may be argued that I have never looked for it, and this would be perfectly true. Professor Salazar de Sousa claims that response to myringotomy was striking and that the procedure itself was relatively trivial. Most British paediatricians trained in the antibiotic era will have had little experience of myringotomy, although it was widely practised in North America where I worked 15 years ago. Perhaps if we looked carefully at necropsy for chronic middle ear infection in these babies we should find it. Perhaps too, if we took our courage in our hands, or in those of our ENT colleagues, we might also find that latent middle ear infection was often present in babies with intractable diarrhoea. Rapid recovery of the patient from his diarrhoea would be gratifying and would support Professor Salazar de Sousa's claims. Perhaps some readers of the *Archives* have had similar experiences, and I know that the Editors would welcome correspondence.

Reference

- Avery G B, Villavicencio O, Lilly J R, Randolph J G. Intractable diarrhea in early infancy. *Pediatrics* 1968; 41: 712-22.