OSTEOMYELITIS OF THE SUPERIOR MAXILLA IN INFANTS

A REPORT ON 24 PERSONALLY TREATED CASES*

BY

FLORENCE CAVANAGH, M.B., B.Sc., F.R.C.S.Ed.
Manchester

In the course of about 100 years there have been reported (so far as I can discover) only 152 cases of this condition. Yet in 17 years I have myself seen and treated 24. This disparity surprised me. Even the more recent reports show some variety in treatment, and the end-results are often unsatisfactory, so that the condition cannot be said to lack interest. It is impossible, in fact, to avoid the suspicion that many cases are not correctly diagnosed. Yet the results of wrong diagnosis and inadequate treatment are by no means negligible. They include sacrifice of teeth, disfiguring scars round the eye, and underdevelopment of the affected maxilla, leading to asymmetry of the face.

I first became interested in this condition in 1940, when I saw a 4-year old boy who had suffered from osteomyelitis of the superior maxilla in the first fortnight of his life. He had scars round his left eye, his face was markedly asymmetrical, as much of the superior maxilla had been removed, and he had no upper teeth on that side. I hoped that when I came to treat one of these babies I should avoid this disfigurement, and it has been most gratifying to find that this is indeed wholly possible if the case is seen at a sufficiently early stage.

Literature

The earliest case in the literature was reported by Rees in 1847. In 1904 Brown Kelly collected 16 records of cases, though 10 of them were entitled "empyema of the antrum of Highmore." In 1922 Marx, an ophthalmologist, discovered 35 recorded cases, and in 1939 Asherson found 70. By 1945 Jacoby and Sagorin had noted 80. I have found a further 72—that is, 152 cases in 111 years.

Most of the papers report isolated cases. Asherson (1939), Reed *et al.* (1953), and Vogt (1956) each reported four cases; Haworth (1947), Schenk (1948), Petrocini and Bullio (1956), and Clerici and Bellina (1957) had five patients each. Dennison (1955), in a paper on osteitis of neonates, mentions eight cases of osteomyelitis of the superior maxilla—occurring over a period of 10 years. Bonnet (1954) saw 15 cases in 20 years, but remarked that this might be due to the fact that he is an ophthalmologist. Incidentally, he reported a mortality rate of 15% in his series.

Clinical Picture

In looking through my case notes and on reading the literature, I was amazed to find how typical most of the histories and findings are; and yet it remains a sad fact that many of these children are treated at eye hospitals as cases of dacryocystitis or are seen by general surgeons who incise the pointing abscess at the outer canthus and never examine the mouth or nose.

The baby with osteomyelitis of the superior maxilla is usually only a few weeks old. Table I shows the age

of my patients at the onset of the disease. (It is worth noting that half were under the age of 2 months and 19 out of 24 were under 3 months.)

The affected baby has usually been quite well until about the day before the doctor is called. He suddenly refuses his food and develops a high temperature. Within 12–24 hours his eyelids have become so oedematous that it may be difficult to see the conjunctiva. There may be chemosis and proptosis. An

TABLE I.—Age at Onset

<1 month ..	8	19	8 months ..	1
1 " ..	4		19 " ..	1
2–3 months ..	7		26 " ..	1
6 " ..	1		30 " ..	1

TABLE II.—Presenting Signs

Swelling of eyelids ..	18
" " cheek ..	13
" " alveolus ..	14
" " palate ..	8
Pus in nostril ..	21
Fistula in alveolus ..	14
" " palate ..	6
" " at inner canthus (or abscess pointing there) ..	2
" " outer canthus (or abscess pointing there) ..	7
" " in lower lid ..	3

abscess may be pointing at the inner or outer canthus—or even at the mid-point of the lower eyelid. The cheek is swollen. There is pus in the nostril of the affected side (and none in the other nostril). The alveolus on the affected side is grossly thickened and there is usually a fistula on its outer aspect in the region of the canine fossa or the first deciduous molar. The palate on the affected side is often boggy and bulges downwards, and there may be a palatal fistula. If pressure is applied to the cheek there is an increased flow of pus from any of the fistulae.

We see, therefore, an ill baby with a swollen eye, and it is easy to understand why he is sent to an eye department.

Table II shows the presenting signs in my patients. The two most frequent signs are the swollen eye and pus in the nostril of that side.

FIG. 1.—Swollen eye seen in babies with osteomyelitis of the superior maxilla.

Diagnosis and Prognosis

Diagnosis is easy if the condition is borne in mind. When a doctor sees a young baby with one swollen eye he should always examine the nose. If he finds pus in the nostril of the affected side he should look at the alveolus to see whether it is swollen and has a fistula on the outer surface. If he presses gently over the cheek pus will run out of the nose and fistula. The diagnosis is then made without any doubt. These babies are often sent for x-ray examination of the maxilla, but personally I have never had any help from such films. Fig. 1 shows the swelling around the eye. Fig. 2 shows the swelling of the hard palate.

The prognosis to-day is excellent. When Asherson (1939) wrote his paper the mortality rate was at least

*Paper read to the Section of Laryngology, the Royal Society of Medicine, London, on December 5, 1958.

25%, and Bonnet (1954) gave a rate of 15%. The morbidity rate was much higher. The surviving children continued to have discharging fistulae—on the face, alveolus, or palate. Sequestra were discharged periodically over many years. Some patients developed ectropion—which is a most unsightly condition. Many

FIG. 2.—Swelling over the hard palate—right side.

had absent or dead teeth on the affected side, and in several children the cheek was reduced in size and the face therefore asymmetrical. To-day these babies stay in hospital for about 10 days, and if diagnosed and treated early there should be no subsequent abnormality—even the milk teeth should be normal.

There has been one death among my 24 patients. This baby came into hospital extremely ill and had a chest infection in addition to the osteomyelitis of the superior maxilla. X-ray films of the chest showed several abscess cavities. These ruptured later to give first left and then right pyopneumothorax. The pus from the lung abscess and the pleura contained *Staphylococcus aureus*, as did the pus from the nose.

The Infection

The source of the infection remains obscure. Table III contains the information available in my 24 cases. As the illness often begins in the neonatal period, it has been thought that the child could have picked up the organism in transit through the vagina or perhaps from an abrasion of the gum caused by the midwife's

TABLE III.—Source of Infection

Mother	Puerperal pyrexia	1
	Breast abscess	2
Child	Paronychia and infected umbilicus	1
	Large abscess in palm	1
	Styes and boils on cheek	1
Other babies in nursing-home		1

TABLE IV.—Bacteriology

<i>Staph. aureus</i>	16
Haemolytic streptococcus	2
Sterile	1
Not recorded	5

cleansing of the mouth at birth. Cracked nipples, breast abscesses, and septic spots on the mother or nurse have all been blamed. In most cases, however, it is impossible to find the cause.

The infecting organism is almost always *Staph. aureus*. This was so in 16 of my 24 cases (Table IV). In one case a haemolytic streptococcus was found in association with *Staph. aureus*. In another case the culture was

sterile because the child had had antibiotic treatment before I saw him. In five of my cases the report has been lost or the pus was not sent to the laboratory.

Pathology

The pathology of these cases is still not perfectly clear. Some authors maintain that the lacrimal sac is first affected, others that the maxillary sinus is the original site, and a third group believe that the focus is in the dental sac of the first deciduous molar.

Fig. 3 shows the relationship of the antrum to the dental sac of the first deciduous molar. Figs. 4 and 5 show the antrum in longitudinal section and in horizontal section. Fig. 6 is an x-ray picture of the dried neonatal skull, and the antrum is clearly shown.

I am indebted to Mr. Asherson (1939) for the use of these last four illustrations. The dental sac is certainly very large in the neonate, but it is separated by a thick layer of tissue from any superficial abrasion which may have occurred on the gum. Wilensky (1932) claims that the infection is blood-borne. He states that the internal maxillary artery supplies the upper jaw in a series of

FIG. 3.—Coronal section showing the maxillary antrum (M.A.) and the tooth socket (T.S.) of the first deciduous molar. M.T.=middle turbinal; I.T.=inferior turbinal; E.C.=ethmoid cell.

FIG. 4.—Longitudinal section to show the maxillary antrum (M.A.) of the newborn.

loops and that the maximum site of damage depends on which branch carries the infected embolus. The dental sac of the first deciduous molar has a very rich blood supply and would therefore be liable to infection by the blood stream, just as the rich blood supply to the growing end of a long bone determines the site of osteomyelitis.

Poncher and Blayney (1934) and Lederer (1935) discuss the post-mortem findings of one case in which serial sections were taken and the infection was followed over a wide field. They claim that there is "incontestable evidence that the site of entry was the left inferior

FIG. 5.—Horizontal section showing the maxillary antrum (M.A.) of the newborn. N.L.D.=nasolacrimal duct.

FIG. 6.—X-ray picture of a dried neonatal skull to show the maxillary antrum.

turbinal" because the lesion of the inferior turbinal was older and more advanced than that in any other part.

Whatever the route by which the infection enters the superior maxilla, we do know that it spreads quickly. The dental sac is certainly infected soon, and in some cases the tooth is forced down by pressure of the pus. It may even be extruded spontaneously. This was described in the older case reports as "premature eruption of the first molar."

The antrum is also involved early—perhaps from rupture of the dental sac into the antrum, or perhaps from the presence of pus already in the nostril. The infection spreads to all surfaces of the maxilla—orbital, facial, alveolar, palatal, nasal—and may spread beyond these parts into the malar bone and the zygoma.

If a fistula develops at the inner or outer canthus, a probe passed through it will enter the body of the maxilla. So will a probe passed through an alveolar fistula; and if a third probe is passed through the nose into the antrum it will meet the other two. This central cavity is a fused abscess cavity involving the dental sac, the antrum, and the bone surrounding them.

Fig. 7—taken from Vogt's (1956) paper—shows forceps entering the alveolar fistula and projecting from the one at the outer canthus after traversing the body of the maxilla. D'Arcy Power (1897) had noted that a probe which he inserted at the inner canthus fistula appeared at the fistula in the mouth.

Treatment

It is because this central cavity forms early that treatment via the nose is so effective. The great advantage of draining the abscess through the nose is that it avoids any scar on the face and also avoids any further damage to the teeth. I have never made an incision on the face. Even if the child has an abscess about to point on the face when first seen it has been possible to avoid incision by putting a "polythene" tube through the nostril into the antrum and aspirating the pus twice daily.

Again, I have never used incisions in the alveolus, as I have great regard for the teeth—even the milk teeth, which, after all, hold open the spaces for the permanent teeth. I was horrified to read, in a paper published as late as 1955, that the author had incised the whole length of the alveolus and removed all the dental sacs of the milk teeth.

My own treatment of these infants is now extremely simple. Systemic antibiotic treatment is begun immediately and is continued for 7–10 days. For local treatment, a trocar and cannula is introduced into the

FIG. 7.—Forceps passing through the alveolar fistula across the body of the maxilla to appear at the left outer canthus.

antrum, a polythene tube is then passed down the cannula, and the tube is strapped to the cheek so that the nursing staff can wash out the cavity twice daily with saline and instil the appropriate antibiotic. Pus is aspirated before insertion of the polythene tube—so that the causal organism can be determined as a guide to the antibiotic treatment. The polythene tube remains in position until no pus has been obtained for two or three days; it is then withdrawn.

If the baby should pull out the polythene tube it can be reinserted with very little trouble. If this cannot be done at once, then simple suction applied to the nostril will suffice to withdraw some pus, and this can be repeated several times daily with a minimum of distress.

Illustrative Cases

The following are some points from the case histories of four of my patients.

Case 1

A male infant was first seen in 1944, at the age of 8 months, with osteomyelitis of the right maxilla. Six months previously he had had similar trouble on the left, treated elsewhere. This is the only patient I have had who suffered from the condition on both sides. The original side was still troublesome; sequestra were often removed by his mother from the four fistulae—two on the face, one in the alveolus, and one in the palate. The right side was seen early (three days after the swelling of the eye started), but before we had antibiotics. His antrum was washed out as part of the initial treatment, and it was interesting to see how quickly the right side recovered. He had no fistulae and no sequestration on that side.

FIG. 8.—Case 1. Boy aged 11 with gross thickening of the left upper alveolus and hard palate. The permanent teeth have been affected by the disease.

Fig. 8 shows the gross broadening of the left alveolus and the stumps of discoloured *permanent* teeth at 11 years.

Case 2

A female infant was first seen at the age of five weeks by her general practitioner, who made one incision under the right eye and a second at the outer canthus.

The fistulae continued to discharge, and at 10 weeks she was admitted to hospital. At that time there was an energetic but inexperienced R.S.O., who proceeded to enlarge the fistulae and curette the bone underneath—on three separate occasions. The fistulae continued to discharge and ectropion developed. She was readmitted, and one of the general surgeons excised the scar and removed a sequestrum.

Two months later the fistulae were still discharging, and I was then asked to see the child. She was 15 months old.

She had a dirty nasal discharge on the right, and when I examined the alveolus I found an erupting dead tooth. There is no note that the alveolus was looked at previously, though the tongue and pharynx were examined at each admission.

I washed out the antrum, which was full of pus, and put in a Malécot catheter (polythene tubing was not available at that time). After a course of washouts and penicillin instillations, the fistulae on the face closed, and remained closed for some time.

Unfortunately, whenever the child had a cold the infection flared up and the fistulae discharged again. She was admitted for two further periods of antral drainage.

FIG. 9.—Case 2. Skin graft at outer canthus which has corrected an unsightly ectropion. Unfortunately the graft is of different colour from the surrounding skin and is adherent to the underlying bone.

Eventually, when the fistulae had been dry for two years, she was referred to the plastic surgeon for correction of the ectropion. She has now had a free graft and the unsightly ectropion has been corrected, but the graft is different in colour from the surrounding skin and is adherent to the bone. A further plastic operation will be carried out later, but I think the child will always have some disfigurement. Fig. 9 shows the appearance at present.

Case 3

This boy was desperately ill when first seen as a baby and had had only systemic antibiotic treatment. He is of special interest because he developed osteomyelitis of the left humerus on the second day after admission—the only complication of this type that I have seen. (In the literature, however, Posey (1912) reports a case in which osteomyelitis of the tibia developed; and White (1935) records the case of a patient who had secondary osteomyelitis of the humerus. Gregson and Yeoman (1957) saw a patient who developed osteomyelitis of the calcaneus.)

Four of the milk teeth in the left upper jaw were affected. They were small and yellow, and soon crumbled away to mere stumps. Fig. 10,

FIG. 10.—Case 3. Showing asymmetry of the face following osteomyelitis of the left superior maxilla.

taken at the age of 6, shows the marked asymmetry of the face—resulting from destruction of much of the left superior maxilla.

Case 4

A male infant had severe involvement of the teeth. He was admitted to an outside hospital when 2 weeks old and a tooth bud was removed. He was readmitted three weeks later and a further tooth was removed. After that, various small sequestra were picked out by his mother.

The child was nearly 5 months old when I first saw him; he then had a large swelling over the outer and upper part of the cheek. There was pus in the nose and in the alveolar fistula. The central and lateral incisor teeth never erupted; the canine and first molar were yellow and crumbling.

Summary

A review is given of 24 cases of osteomyelitis of the superior maxilla in infants treated personally since 1941. This number is surprising, as, so far as can be discovered, only 152 cases have been reported during 100 years. It is possible, therefore, that many cases are not correctly diagnosed.

Staphylococcus aureus is the usual organism found, and when the infection is contracted in a large maternity unit it is usually insensitive to one or more of the antibiotics.

At an early stage of the disease a central abscess cavity develops in the superior maxilla, and this can be drained by inserting a polythene tube through a cannula which has punctured the lateral nasal wall under the inferior turbinal. By this means antibiotics can be instilled locally. The baby is also given systemic antibiotics.

No incision should be made in the alveolus, or the teeth will be further damaged and incisions around the eyes should be avoided, thus preventing unsightly scars.

This paper is published in the *British Medical Journal* by kind permission of the Editors of the *Proceedings of the Royal Society of Medicine*. Figs. 3-6 are included by kind permission of the editor of the *Journal of Laryngology and Otology* and of Mr. N. Asherson (1939). Fig. 7 is reproduced by permission of U. Vogt (1956) and the publishers, Springer-Verlag.

BIBLIOGRAPHY

- Achar, M. V. R. (1957). *A.M.A. Arch. Otolaryng.*, **66**, 248.
 Asherson, N. (1939). *J. Laryng.*, **54**, 691.
 Barker, G. N. (1942). *Ibid.*, **57**, 157.
 Bass, M. H. (1928). *Amer. J. Dis. Child.*, **35**, 65.
 Bernheim, M., Chavanne, L., and Roux, J. (1947). *Pédiatrie*, **36**, 232.
 Bloch, A., Loroque, and Gruber, C. (1937). *J. Laryng.*, **52**, 194.
 Bonnet, P. (1954). *Arch. Ophthal. (Paris)*, **14**, 343.
 Clerici, E., and Bellina, M. (1957). *Boll. Mal. Orecch.*, **75**, 123.
 Cohen, M. M. (1949). *Oral Surg.*, **2**, 50.
 Collet, F. J. (1922). *Arch. int. Laryng.*, **28** (n.s. 1), 1041.
 Dennison, W. M. (1955). *Lancet*, **2**, 474.
 Dey, D. L. (1957). *Med. J. Aust.*, **2**, 648.
 Douglas, A. (1898). *Brit. med. J.*, **1**, 368.
 Dunphy, D. L., and Frazer, J. P. (1947). *Yale J. Biol. Med.*, **19**, 877.
 Faulkner, E. R. (1923). *Laryngoscope*, **33**, 557.
 Fleming, T. N. (1928). *Minn. Med.*, **11**, 252.
 Forbes, R. P. (1931). *Colo. Med.*, **28**, 317.
 Franchini, Y., Turro, O. R., Ribo, J., and Ferrero, N. (1956). *Rev. Asoc. méd. argent.*, **70**, 234.
 Galli, P. (1926). *Clin. pediat. (Bologna)*, **8**, 604.
 Gittins, T. R. (1925). *Ann. Otol. (St. Louis)*, **34**, 1259.
 Goldbloom, A., and Bacal, H. L. (1937). *Canad. med. Ass. J.*, **37**, 443.
 Gollnitz, H. (1958). *H.N.O. (Berl.)*, **6**, 289.
 Green, W. T., and Shannon, J. G. (1936). *Arch. Surg. (Chicago)*, **32**, 462.

- Gregson, A. E. W., and Yeoman, P. M. (1957). *J. Laryng.*, **71**, 703.
 Haas, L. (1955). *Brit. med. J.*, **2**, 245.
 Harlowe, H. D. (1941). *Laryngoscope*, **51**, 464.
 Harry, P. A. (1919). *Brit. J. Ophthal.*, **3**, 557.
 Haworth, J. C. (1947). *Arch. Dis. Childh.*, **22**, 175.
 Howard, J. E., and Robinson, A. (1948). *J. Mt Sinai Hosp.*, **15**, 101.
 Hughes, E. E. (1921). *Brit. J. Dis. Childh.*, **18**, 89.
 Jacoby, N. M., and Sagorin, L. (1945). *Arch. Dis. Childh.*, **20**, 166.
 Jervoy, J. W. (1937). *J. S.C. med. Ass.*, **33**, 258.
 Kägebein, P. (1956). *Disch. med. J.*, **7**, 574.
 Kelly, A. Brown (1904). *Edinb. med. J.*, **16**, 302.
 Lacy, N. E., and Engel, L. P. (1939). *Arch. Otolaryng. (Chicago)*, **29**, 417.
 Lederer, F. L. (1935). *Surg. Gynec. Obstet.*, **60**, 645.
 Lust, M., Petit, R., and Durant, P. (1950). *Nourrisson*, **38**, 157.
 Macbeth, R. G. (1951). *Proc. roy. Soc. Med.*, **44**, 1030.
 McCash, C. R., and Rowe, N. L. (1953). *J. Bone Jt Surg.*, **35B**, 22.
 McGovern, F. H., and Newman, S. (1946). *Virginia med. Monthly*, **73**, 219.
 Magnus, J. A. (1944). *Brit. J. Ophthal.*, **28**, 135.
 Mallardi, A. (1957). *Lattante*, **28**, 248.
 Marx, E. (1922). *Brit. J. Ophthal.*, **6**, 25.
 Mayer, E. (1901). *Med. Rec. (N.Y.)*, **60**, 210.
 Mowlem, R. (1944). *Brit. med. J.*, **1**, 517.
 Offret, G., and Meaux, J. (1947). *Arch. Ophthal. Paris*, **7**, 250.
 Oringer, M. J. (1948). *Oral Surg.*, **1**, 842.
 Petrocini, S., and Bullio, D. (1956). *Minerva pediat. (Torino)*, **8**, 662.
 Phelps, K. A. (1923). *Trans. Amer. Acad. Ophthal. Otolaryng.*, **28**, 115.
 Poncher, H. G., and Blayney, J. R. (1934). *Amer. J. Dis. Childh.*, **48**, 730.
 Posey, W. C. (1912). *J. Amer. med. Ass.*, **59**, 1020.
 Power, D. A. (1897). *Brit. med. J.*, **2**, 808.
 Reed, H., Spiro, I., and Wilson, B. D. R. (1953). *Brit. J. Ophthal.*, **37**, 629.
 Rees, G. A. (1847). *Lond. med. Gaz.*, **4**, 859.
 Roehm, H. R. (1931). *Amer. J. Dis. Childh.*, **42**, 1171.
 Röpke (1899). *Arch. Otol. (N.Y.)*, **28**, 259.
 Roure, M. (1898). *Arch. int. Laryng.*, **11**, 137.
 Schenk, R. J. (1948). *Pract. oto-rhino-laryng. (Basel)*, **10**, 472.
 Schmiegelow, E. (1896). *Arch. Laryng. Rhin. (Berl.)*, **5**, 115.
 Self, E. B. (1948). *Pediatrics*, **1**, 617.
 Shea, J. J. (1928). *Ann. Otol. (St. Louis)*, **37**, 509.
 Stephenson, S. (1918). *Brit. J. Ophthal.*, **2**, 416.
 Stuart, D. W., and Pitt-Steel, C. A. (1954). *J. Laryng.*, **68**, 712.
 Terracol, J. (1928). *Arch. int. Laryng.*, **34** (n.s. 7), 532.
 ———, Guerrier, Y., and Chalmerton, J. (1947a). *Ann. Oto-laryng. (Paris)*, **64**, 44.
 ———, ——— (1947b). *Ibid.*, **64**, 40.
 Turner, J. H. (1942). *Arch. Pediat.*, **59**, 1.
 Vogt, U. (1956). *H.N.O. (Berl.)*, **5**, 275.
 White, H. D. (1935). *Arch. Dis. Childh.*, **10**, 85.
 Wilensky, A. O. (1932). *Amer. J. Dis. Childh.*, **43**, 431.
 Wolf, I. J. (1933). *Ibid.*, **45**, 1040.
 Wood, F. C. (1897). *Brit. med. J.*, **2**, 1259.
 Zarfl, M. (1920). *Z. Kinderheilk.*, **25**, 266.

"I would like to express once again the hope that young West African scientists in ever-increasing numbers may be attracted to the field of medical research. There is a feeling that research workers are oddities. Perhaps there are a few—you can't do without some of them. Some three or four years ago on my arrival in Dar-es-Salaam a medical administrator who met me remarked after an awkward silence, 'You know, you people are a race apart. You're quite different from other people.' Later, when I was sounding a very experienced medical administrator in East Africa as to whether he would be willing to become Secretary of our East African Council for Medical Research, he commented, 'But surely you want one of those young men with beards and sandals.' It is true that we have our prima donnas, but we have no monopoly of them; there must be many in other vocations than medical research. It is true that we have to include the peculiar and the unorthodox, because very often they have something of vital importance to contribute. But, taken by and large, the stigmata of the medical research worker can be summed up, as I have already mentioned, in the fact that he is imbued with the simple worth-whileness of finding out why something happens, and with the zeal to pursue that quest." (Dr. R. Lewthwaite, Presidential Address, Conference on Tuberculosis and Leprosy, Northern Nigeria.)