Medical Memoranda

Familial Angioneurotic Oedema: Two Fatal Cases after Dental Extractions

The five cases reported below convey a warning of the risks of dental trauma in subjects of angioneurotic oedema.

Case 1.—A woman aged 20 had an uneventful medical history until the age of 16, when she began to suffer from recurrent superficial swellings of the face, hands, and feet for which she received medical treatment. When 18 she had a tooth extracted under local analgesia, after which swelling of the lips and face occurred, which disappeared in two to three days. On March 22, 1951, at 11.30 a.m., she had four teeth extracted under nitrous oxide and oxygen—Nos. 5 and 8 from the lower right jaw, No. 6 from the right upper jaw, and a supernumerary tooth between 2 and 3 on the upper left jaw. She recovered completely and was able to go home by tram and went to bed. By 4.30 p.m. there was some swelling of the lips, and by 6.30 p.m. this was so obvious that her doctor was sent for and he gave an adrenaline injection. By 10.30 p.m. the neck was visibly swelling and breathing suddenly became difficult; she got up and went to the opened window to get air, but in a few minutes collapsed and died, gasping for breath.

The next day one of us (R.R.) performed a post-mortem examination. The body was that of a young well-developed and well-nourished woman showing puffiness below the eyes, swelling and protrusion of both lips, tongue protruding through the teeth, swelling of the neck, and swelling and pitting of left forearm and hand. On dissection, the whole of the back of the throat, with the uvula and soft palate, was swollen, particularly the epiglottis, while the vocal cords were so oedematous as to occlude the airway completely (see photograph). The neck tissues were also oedematous. Both lungs were tense, deeply congested, and oedematous, and the blood was dark and fluid. The heart was normal. Apart from deep congestion of the abdominal organs and oedema of the brain, nothing significant was found. Death was certified as due to asphyxia from oedema of the glottis occurring in a patient subject to angioneurotic oedema, the present attack being precipitated by dental extractions.

Case 2.—A man aged 28. One of us (R.R.), as medical referee to the cremation authority, received on April 26, 1956, papers for this deceased person, who died on April 24, 1956, in which the cause of death was given as angioneurotic oedema and oedema of glottis, and in answer to the question, "Was there any other cause that contributed to or accelerated death," the answer was given, "Had molar tooth extracted on April 23, 1956." The coincidence of a comparatively uncommon cause of death associated with dental extraction aroused curiosity, and naturally led to further investigation, which revealed that this man was the brother of Case 1, and that he had had one tooth extracted under an analgesic ("unacaine" with adrenaline). He, too, had a history of

attacks of angioneurotic oedema for some years. His death occurred under circumstances similar to those in Case 1, 14 hours after the dental extraction.

These patients were the second and third eldest of a family of nine in which it was stated that neither father nor mother had so suffered, nor had any of the rest of the family except another brother (Case 3). He was duly warned of the risks of dental treatment, and when such became necessary he was seen by the consulting dental surgeon to the North-Eastern Regional Area, and admitted to hospital, where the precautionary measures were taken.

Case 3.—A young man, aged 19, was admitted to hospital on December 3, 1956, for extraction of carious teeth. He gave a history of oedematous swellings of various parts of the body following injury even when not severe—for example, swelling of eyelids of both eyes after a blow on the head. He never had difficulty in swallowing or breathing. His history was otherwise negative and his physical examination revealed nothing of note. He did, however, show marked weals on skin-scratching, and therefore for four days he was treated with mepyramine 50 mg. twice daily, this reaction then ceasing. Before operation he had 50 mg. of pethidine and 0.64 mg. of atropine sulphate with nitrous oxide and oxygen anaesthesia. Immediately after he was returned to bed; at 11.30 a.m. he had 50 mg. of mepyramine, repeated at 5 p.m., and 25 mg. at 9.30 next morning. He showed nothing but slight local swelling of extraction sites and made an uninterrupted recovery.

Case 4.—This patient, a boy aged 14, had no history of angioneurotic oedema or of asthma, eczema, or other allergic manifestation, but because of the family history he was admitted to hospital under the care of the consultant dental surgeon. Skin-scratching showed some weal formation, and he received mepyramine 50 mg. for two days, and immediately after operation again received 50 mg. of mepyramine. Recovery was uneventful.

Case 5.—This patient, aged 33, stated that he had had slight oedematous swelling as a younger man but none for several years. He, too, however, was admitted to hospital and for two days before operation received 25 mg. of promethazine with 50 mg. of pethidine and 0.64 mg. of atropine sulphate, and immediately after operation 50 mg. of mepyramine. Again recovery was uneventful.

Within a comparatively short time we have therefore had five cases requiring dental treatment—all members of a family with a definite history of angioneurotic oedema—two of which proved fatal. The third, by intensive treatment, was probably saved from serious trouble, while the other two would, in all likelihood, have had only minor upset even if untreated. Such a history, however, emphasizes the necessity of strict precautions in carrying out dental treatment in patients with a familial history of angioneurotic oedema, as apparently it was the trauma from the dental extractions and not the anaesthetic which triggered off the acute attack of angioneurotic oedema.

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Post-mortem specimen from Case 1.

A Unesco report on the education of atypical children in various countries, *Statistics on Special Education*, is available, price 10s., from H.M.S.O., P.O. Box 569, London S.E.1. The report is chiefly concerned with statistics about the education of children who are handicapped physically, mentally, or morally, but some countries have sent in statistics on highly intelligent children and itinerant children. These figures are also included.