

UNUSUAL SITES OF DIPHThERITIC MEMBRANE AND CERVICAL OEDEMA

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Abstract The clinical picture in four children (5½-8 years of age) who showed a diphtheritic membrane in an unusual site - during an outbreak of diphtheria in Khartoum - is reported and discussed in the light of similar reports in the literature. Three patients showed the rarely reported finding of a labial diphtheritic membrane unassociated with involvement of either the nose or throat. In two patients, the membrane covered the floor of the mouth and in a third patient also the inferior surface of the tongue. The fourth child had a membrane on the floor of the mouth as well as in the tonsillopharyngeal region. This extended membrane was accompanied by severe cervical oedema giving the classical 'bull-neck' appearance.

Key words Diphtheria; Membranes; Mouth mucosa; Tongue; Oedema; Neck

INTRODUCTION

At many hospitals of the Developing World, including several in the Sudan, the diagnosis of diphtheria is essentially clinical, since specific bacteriological and serological investigations are not feasible^{1,2}. Although the appearance of the throat does not specifically distinguish the disease from a number of other infections, the typical diphtheritic membrane in the tonsillopharyngeal region forms a corner-stone in the diagnosis. The accompanying cellular infiltration - presenting as oedema of the neck - is a helpful clinical sign, when present. In its severest form, the swelling forms a distinct collar, reaching from ear to ear and filling out the whole space beneath the jaw, giving a 'bull neck' appearance. This entity has received special attention, since it has been found to be associated with high morbidity and mortality^{3,4}.

McClosky et al⁵ observed another type of oedema in an epidemic of diphtheria in San Antonio, and called it 'erasure oedema'. This was characterized by obliteration of the sternomastoid muscle border, the mandible and the median border of the clavicle.

In this paper we report four cases presenting during an epidemic of diphtheria in Khartoum with an unusually situated diphtheritic membrane in the oral cavity, associated in three of them with cervical oedema which was confined to the submental region.

PATIENTS AND METHODS

The patients were seen amongst 107 children who were admitted with respiratory diphtheria, during an epidemic, to the Children's Emergency Hospital and Khartoum Teaching Hospital in the period 3 September to 20 December 1978. A full account of this epidemic is given elsewhere^{6,7}.

Case reports

Case 1. A six-year-old boy had sore throat and swelling of the neck for 3 days before admission. On direct questioning he denied earache, and was said not to have dental problems or skin infection. Nor did he have

nasal discharge. No antibiotic therapy had been provided before he reported to the hospital. His past medical, family and nutritional histories were insignificant. He had had three doses of triple (DPT) and oral polio vaccines but had not received a booster dose.

On examination, he looked ill and toxic. His temperature was 38°C and his pulse was 102/minute and regular. He displayed a 'bull neck' appearance, with the cervical swelling extending between the mandible and the anterior sternomastoid border on either side and filling out the space beneath the jaw. The swelling was non-pitting, warm to touch and tender on palpation. Throat examination revealed large congested tonsils with a creamy-greyish adherent membrane covering both tonsils and the uvula. There was another membrane in the floor of the mouth, 1½ cm in diameter, which left a bleeding raw surface on an attempt to remove it. Throat and nasal swabs were positive for *Corynebacterium diphtheriae* subsp. *gravis*. The organism was of the toxigenic *gravis* strain with the unusual property of fermenting sucrose.

The patient received 100,000 units of diphtheria antitoxin on admission and 1,000,000 units of procaine penicillin intramuscularly daily for 7 days. He made a good recovery and on discharge on the tenth day all symptoms and signs had subsided, apart from the cervical oedema, which was then confined to the submental region. The swelling was still hard, but not tender. It was associated with bilateral, non-tender, palpable submandibular lymph nodes measuring 2x3 cm.

On follow-up 4 days later, the submental swelling had the same characteristics, but had become soft and fluctuant in the middle (Fig 1). It was surgically drained and the aspirate revealed no bacterial growth. The patient was put on ampicillin + cloxacillin intramuscularly for 2 days and then orally for another five days. Subsequently, he made an uneventful recovery and in 2 weeks the cervical swelling had completely disappeared.

Case 2. A 5½-year-old girl presented with submental swelling of 10 days' duration, accompanied by mild fever but no sore throat. She had received six injections of penicillin in a health centre without regression of the

swelling. She had not been immunized and her social history revealed that she had been in contact with a boy who had been admitted with diphtheria during the epidemic.

She was ill, with a temperature of 38°C and a regular pulse of 123/min. A hard mass, 5x4 cm in size and warm to touch, occupied the submental region (Fig 2). In addition, non-indurated submandibular lymph nodes, 3x4 and 2x2 cm in size, were palpable on the right and left sides respectively. Throat examination revealed tonsillar hyperaemia, but neither an exudate nor a membrane was visible. A creamy membrane about 1 cm in diameter was adherent to the floor of the mouth. A swab from the membrane, tonsils and both nares revealed no bacterial growth. The patient was given antidiphtheritic therapy similar to that of Case 1 and made an uneventful recovery, with gradual resolution of the submental swelling in 3 weeks.

Case 3. This 7-year-old girl had fever, sore throat and dysphagia for 3 days before reporting to hospital. Before admission she had received ampicillin syrup which had been obtained by her family from a pharmacy without a prescription. She had not been vaccinated.

On examination, she was febrile (temperature 39.1°C) and her pulse was 132/min and regular. A warm, tender, hard swelling occupied the entire submental region (Fig 3). Submandibular and anterior cervical lymph nodes were palpable bilaterally - 3x4 cm on the right side and 2x3 cm on the left. Throat inspection revealed slightly enlarged tonsils, which were hyperaemic but not covered by a membrane. However, a membrane similar to that in Case 2 was attached to the floor of the mouth. Throat, buccal and nasal cultures were negative. The patient responded well to 100,000 units of diphtheria antitoxin combined with a 7-day course of procaine penicillin intramuscularly. The submental swelling subsided completely in 2 weeks.

Case 4. A boy of 8 years presented with pyrexia, sore throat, dysphagia and nasal discharge of 2 days' duration. He had been seen in a health centre near his home, where he had received one intramuscular injection of procaine penicillin for a presumptive diagnosis of tonsillitis. Subsequently, his temperature rose and he



Figure 1. Submental swelling in case 1 following surgical drainage



Figure 2. Submental swelling in case 2 three days after starting treatment.

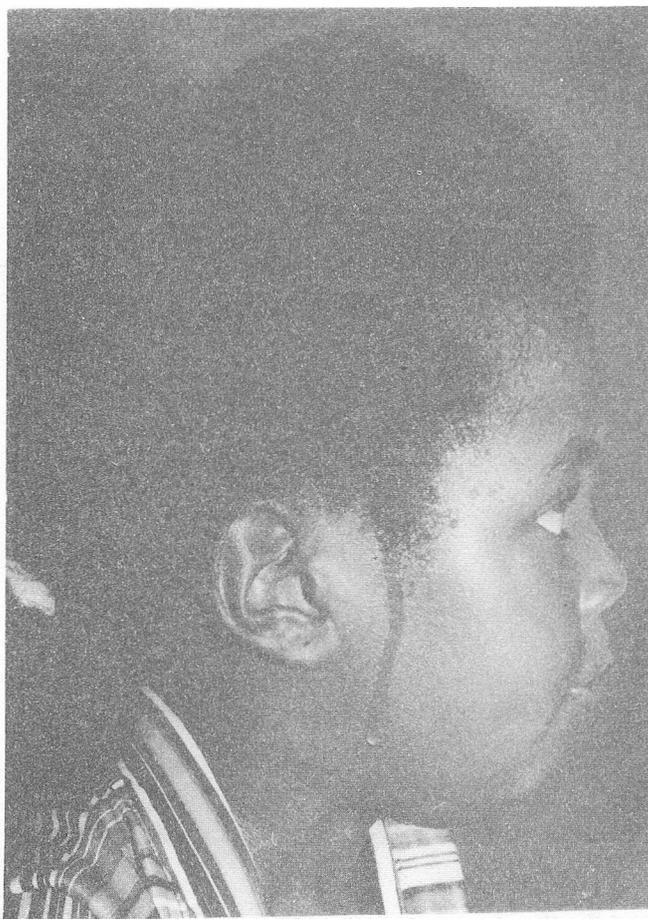


Figure 3. Case 3 Showing submental swelling three days after initiating specific treatment.

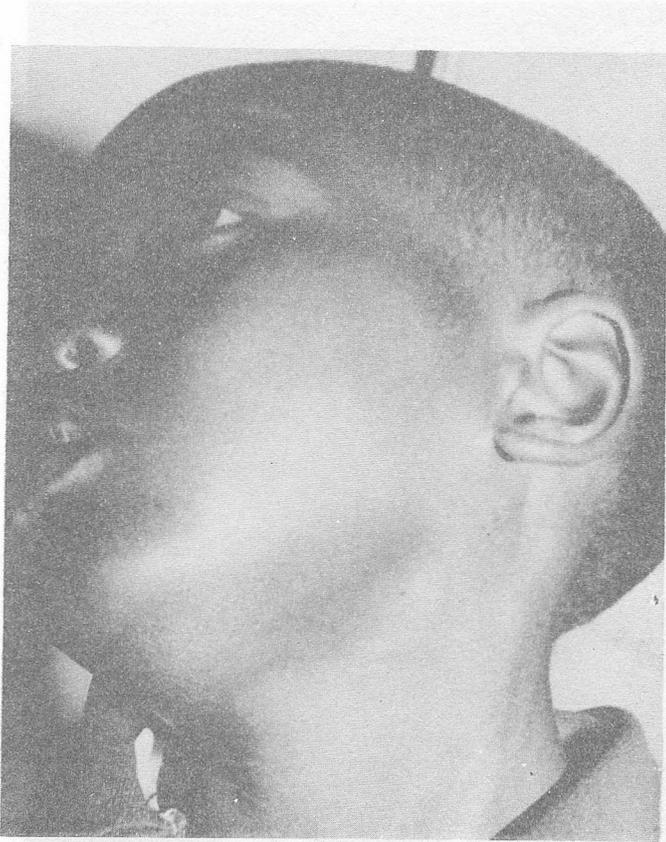


Figure 4. Submental Swelling in case 4 six days after starting antidiphtheritic therapy.

developed pronounced submental swelling, and his parents decided to take him to hospital. He had not been immunized before.

He looked toxic and had marked pyrexia (temperature 40.2°C). His pulse was 100/min and examination of the cardiovascular system revealed no abnormality. The tonsils were enlarged and hyperaemic, but no tonsillopharyngeal membrane could be seen. His tongue was also hyperaemic and swollen. There was a creamy-whitish membrane about 1 cm in diameter covering the inferior surface of the tongue and another one, 1½ cm in diameter, in the floor of the mouth. A pronounced swelling occupied the whole of the submental region and extended down the neck to the level of the cricoid cartilage (Fig 4). It had a similar consistency that in Cases 1-3 and was warm to touch. Submandibular lymph nodes were palpable on both sides.

This patient was managed in a similar way to the other patients with diphtheria and the submental swelling regressed dramatically in 3 days. He was discharged on the seventh day in good condition, with residual submental swelling about 3 cm in diameter. He did not report again for follow-up.

DISCUSSION

These four patients illustrate the fact that the diphtheritic membrane can involve parts of the oral cavity other than the tonsillopharyngeal region, an observation that was well recognised when the disease was still common in Europe. It was thought to arise most commonly from failure to arrest the membrane in late severe faucial diphtheria⁸, and in Hamburg, 49 patients with lip lesions were recorded among 7314 fatal cases⁹. In a 24-year-old female, Fleming¹⁰ reported the occurrence of the diphtheritic membrane on the floor of the mouth, the root of the tongue, the lower gum, and in between the gum and lower lip. She had a membrane in the anus, in the perianal area and in the sulcus between the labia majora and labia minora, extending over the urethral surface and into the vagina.

The first patient in the present report is an example of cases where the membrane involves the floor of the mouth in addition to the tonsillopharyngeal region.

It is noteworthy that this extended membrane was accompanied by severe cervical oedema giving the classical 'bull neck' appearance. It would therefore seem wise to inspect the other parts of the oral cavity together with the tonsillopharyngeal region when examining a case of diphtheria presenting with severe cervical oedema of the 'bull neck' type.

Unfortunately, bacteriological proof of diphtheria could be documented only in the first case. The cervical swelling in this patient was subsequently complicated by the development of a submental abscess, possibly due to secondary bacterial infection with either *Streptococcus pyogenes* or *Staphylococcus aureus*. Such secondary infection (classically due to *Streptococcus pyogenes*) is known to occur in diphtheria¹¹. Initial throat and nasal cultures were negative for these pathogens, but these results cannot be considered conclusive, as it is now well documented that needle aspiration of the enlarged lymph nodes is superior to throat and nasal cultures in isolating the aetiological agent causing cervical lymphadenitis. In studies where such a procedure was used, the organism was identified in 60-80% of the cultures¹². Our patient did not undergo such an investigation and the sterile aspirate of the submental abscess is explainable by the preceding penicillin chemotherapy. A change of the antibiotic to cloxacillin and ampicillin was mandatory following the drainage of the abscess, in order to guard against the possibility of *S. aureus* infection, which is the agent most frequently isolated when cervical lymphadenitis progresses to abscess formation¹³.

The other three cases are examples of the development of a labial diphtheritic membrane unassociated with involvement of either the nose or throat. This finding is very infrequent. Riddell¹⁴ reviewed 24 such cases and also reported another 15 with an associated respiratory localization. It is interesting to note that in three of our four cases the cervical lymphadenitis and associated oedema were confined to the area of lymph node drainage, i.e. the submental region. It might therefore be of importance to inspect the floor of the mouth in a patient presenting with submental cervical swelling, especially in such a vulnerable population as the unimmunized group of Sudanese children.

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