



## AN UNUSUAL CASE

## More than a mouthful

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A 39-year-old man with trisomy 21 developed acute tongue swelling, initially thought to be an allergic reaction. He was treated with hydrocortisone injection and promethazine. The patient was epileptic, was maintained on carbamazepine, and was allergic to penicillin. On further deterioration a tentative diagnosis of hereditary angioneurotic oedema was made and epsilon aminocaproic acid (EACA) and antibiotics, namely metronidazole and clindamycin, were added to the treatment.

Six days after onset of the swelling the patient was referred to our department. He was totally unco-operative and non-communicative. He was afebrile and had a massively enlarged tongue protruding from his mouth that was immobile and not fluctuant (Figs 1 and 2). There was no airway obstruction and his chest was clear. His oral cavity could not be extensively examined because of the tongue swelling, but peg-like upper dentition was noted. He had a raised white cell count (WCC) of  $36,4 \times 10^9/l$  with a differential count showing 78% neutrophils. His erythrocyte sedimentation rate (ESR) was 70 mm. Complement (C3 and C4) as well as C1 esterase inhibitor levels were within the normal range.



Fig. 1. Pre-operative view showing massive tongue swelling.

Attempts at imaging were inconclusive because of lack of patient co-operation and reluctance to sedate him fully owing to his at-risk airway. He underwent examination under anaesthesia. Oral intubation was relatively easy as most of the tongue swelling was found to be outside the oral cavity. A

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Fig. 2. Intra-operative view with patient orally intubated.

tracheostomy was performed. The main swelling was in the anterior two-thirds of the tongue, with a bead of pus noted on the mid-dorsal surface.

An incision in the midline of the dorsal tongue surface was made into a large abscess cavity, which extended to the ventral surface and several centimetres anteriorly. About 20 ml of pus was drained and a pus swab taken. A corrugated rubber drain was inserted through the tongue to ensure drainage. Dental indentation was seen on the ventral surface of the tongue and the superior and inferior teeth were carious, loose and peg-like. Complete dental clearance was performed.

The patient responded well, with the tongue decreasing in size over a few days so that it was mobile and could be retracted fully into the mouth. The drain was removed postoperatively on day 3 and tracheostomy on day 6. The WCC fell to  $13,5 \times 10^9/l$  and the pus swab revealed *Streptococcus agalactiae*, *Klebsiella ozaenaena* and *Candida albicans*. Co-trimoxazole, clindamycin and nystatin were commenced in accordance with sensitivity results. The patient was discharged on day 8.



## Discussion

The diagnosis of lingual abscess in the anterior two-thirds of the tongue is simple in view of the characteristic symptoms and signs.<sup>1</sup> Our patient's initial diagnosis was confused, possibly because of his lack of communication and the fact that he was afebrile. Hereditary angioneurotic oedema, caused by C1 esterase inhibitor protein deficiency, usually presents with a triad of symptoms and signs, namely abdominal pain, peripheral non-pitting oedema, and laryngeal oedema.<sup>2</sup>

Lingual abscess is an extremely rare condition.<sup>3</sup> The tongue is very resistant to infection due to factors such as a thick keratinised epithelial layer, resistance of muscle to infection, cleansing action of saliva, abundant vascularity, paucity of submucosal areolar tissue, and contact exposure of the tongue to bacteria resulting in immunity.<sup>4</sup> The aetiological factors in the formation of tongue abscesses include dental or foreign body trauma,<sup>4</sup> acute parenchymous glossitis, infected circumvallate papillae, upper respiratory tract infections,<sup>5</sup> and carious teeth as present in our patient. The symptoms include severe tongue pain, decreased movement of the tongue (protrusion), hyper-salivation, dysphagia and voice change.<sup>1</sup>

The most common site is the anterior two-thirds of the tongue.<sup>4</sup> Abscesses of the lingual tonsil, extension of infections from the molar teeth,<sup>6</sup> and intralingual thyroglossal duct cysts need to be borne in mind when an abscess is located within the tongue base.<sup>3</sup> Pyrexia is not necessarily present<sup>7</sup> and the WCC and ESR are variable.<sup>6,7</sup> The organisms responsible are usually *Staphylococcus aureus* and *Streptococcus haemolyticus*.<sup>4</sup> Both ultrasound<sup>3</sup> (US) and computed tomography (CT)<sup>1</sup> have been utilised to aid diagnosis.

As in our patient with tongue abscess and delay in diagnosis, treatment includes incision and drainage, airway monitoring, antimicrobial therapy, and addressing causative factors.

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