

Title: Tongue Protrusion as the Presenting Symptom of Parapharyngeal Abscess

Authors: Timothy Visclosky, MD^a, Sarah Tomlinson, MD^a, Lauren Bohm, MD^b, Andrew Hashikawa, MD^a

Author Affiliations:

^a Department of Emergency Medicine, University of Michigan, Ann Arbor, MI, USA

^b Department of Pediatric Otolaryngology, University of Michigan, Ann Arbor, MI, USA

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Abstract:

Deep neck space infections are commonly seen in the pediatric population. The diagnosis, however, can be challenging to make and requires a high degree of suspicion due to developmental and age-related factors in children, and non-specific presenting symptoms. Diagnosis becomes further complicated in patients whose comorbid conditions mask some of the more severe systemic symptoms. We present a case of a two-year-old female with Trisomy 21 who presented with a chief concern of “tongue swelling” per parents. After initially failing treatment for presumed angioedema caused by an ACE-inhibitor, she was ultimately diagnosed with a parapharyngeal abscess with extension and mass effect causing tongue protrusion. This case represents a novel early presentation of a common infection and highlights the challenges of diagnosing deep neck space infections in children.

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Introduction:

Deep neck space infections are relatively common with an annual incidence of 4.6 cases per 100,000 children in the United States [1]. While a number of clinical signs and symptoms can alert providers to these infections, the presentation can be insidious and extremely variable, making diagnosis challenging in pediatric patients [2-5]. Delayed diagnosis can result in severe complications including airway compromise, septic thrombophlebitis, and carotid artery erosion [2]. Emergency physicians must maintain a high level of suspicion and understanding of neck and oropharyngeal anatomy to avoid missing subtle, and occasionally misleading, presentations of serious neck infections in children. We present a case of parapharyngeal abscess presenting as tongue protrusion.

Case-Report:

A two-year-old female with Trisomy 21, Tetralogy of Fallot status post repair, and hypothyroidism, presented to the Emergency Department (ED) with concern of “tongue swelling” for one day. Parents reported nine days of intermittent fevers to 102°F with mild, clear rhinorrhea. Four days prior to presentation, the patient was seen at an Urgent Care center, where she had negative testing for COVID-19, influenza, and RSV. She was diagnosed with a viral upper respiratory infection (URI). Following that visit she began having multiple episodes of non-bloody, non-bilious emesis per day with watery stools. When she woke up on the morning of her presentation to the ED, her parents noted her tongue was protruding from her mouth. They estimated that her tongue appeared twice its normal size and stated that she was unable to bring it back into her mouth. The patient was otherwise acting normally with an unchanged activity level. Parents denied any difficulty or noisy breathing, cough, vomiting, or rash. She had been on daily enalapril for the past eight months for afterload reduction with decreased left ventricular function. Enalapril dose was last increased two months prior to this presentation. Her medications also included daily aspirin and levothyroxine. She had no personal history of macroglossia.

On the initial presentation, the patient was active and comfortable appearing. She was afebrile and all other vital signs were age-appropriate. She was breathing easily without retractions, nasal flaring, or stridor on room air with clear lung sounds bilaterally. She was able to manage her oral secretions well and no drooling was noted. Her exam was most notable for a beefy but non-tender red tongue, protruding ~two centimeters from her mouth (Figure 1). During her admission, parental permission was obtained to use the photography in this case report. Her mucous membranes were dry with no oral lesions, lip swelling, or neck masses. The floor of her mouth was soft and she demonstrated full and comfortable range of

motion of her neck. The remainder of her oropharyngeal exam was limited due to her tongue findings and her tonsils were not able to be visualized.

Her labs were notable only for leukocytosis (WBC 21.7 K/uL with neutrophil predominance), and an elevated CRP (6.3 mg/dL). After a review of her medications, there was concern for bradykinin-induced tongue angioedema secondary to enalapril. She was started on dexamethasone, diphenhydramine, and famotidine and admitted to the Pediatric Intensive Care Unit. Pediatric Cardiology was consulted and agreed with discontinuing the enalapril. Pediatric Otolaryngology was also consulted and were similarly unable to visualize the posterior pharynx but agreed with the plan. Her tongue protrusion showed slow, but gradual improvement back to baseline and no rebound was observed after discontinuing steroids. She was subsequently discharged home on hospital day two.

Two days after discharge, however, the patient's parents called the primary cardiologist to report progression of symptoms. She continued to have fevers and her tongue was again protruding from her mouth. She was also more fatigued than before and parents saw new right-sided neck swelling. She was directly admitted to the hospital where her exam was now notable for right-sided neck fullness extending to the jawline, and limited range of motion of the neck. At this time a CT neck was obtained that revealed a parapharyngeal abscess (3.3 cm x 3.2 cm x 2.7 cm) with extension into the sublingual space causing mass effect (Figure 2). It became clear that what was initially interpreted as "tongue swelling", was in fact tongue protrusion secondary to mass effect. The patient was quickly taken to the operating room by Pediatric Otolaryngology. The parapharyngeal abscess was drained via a transoral approach and she was started on intravenous ampicillin-sulbactam. Cultures from the drained fluid grew Group A Streptococcus and Prevotella melaninogenica. Following drainage, the patient had no further fevers and her tongue protrusion and other airway symptoms resolved. She was discharged home on hospital day two after transitioning to oral amoxicillin-clavulanate.

While continuing the oral antibiotics, she did develop a right upper extremity abscess. An endocarditis workup was negative and it remains unclear if this represented a contiguous infection or seeding from transient bacteremia. The abscess was drained but cultures showed no growth. Her antibiotics were broadened with the addition of trimethoprim-sulfamethoxazole. Following this, her symptoms showed total improvement, inflammatory markers normalized, and she has had no recurrence of infections since that time. A subsequent immunologic workup after completing treatment demonstrated decreased T and B cell function, believed secondary to her history of Trisomy 21.

Discussion:

Parapharyngeal abscesses are commonly seen in children under 5 years of age [6], and are often secondary to contiguous spread from local sources of infection, such as tonsillitis [7]. They result in growing infection within the potential spaces between the deep fascial planes [4]. As reflected in our patient, parapharyngeal abscesses rarely cause localizing symptoms until the infection spreads beyond these anatomic boundaries [4]. Consequently, the most common presenting symptoms of parapharyngeal abscesses include fever, neck pain/mass, neck stiffness, and odynophagia [5, 8]. Yet even these symptoms can have widely variable penetrance and severity among children and can be further obscured by any comorbid conditions or immunosuppression [5]. Unlike other deep neck space infections, such as Ludwig's angina and even retropharyngeal abscesses [9, 10], parapharyngeal abscesses are unlikely to have extension that is significant enough to result in mass effect beyond localized neck swelling and stiffness. To our knowledge, there are no previous case reports of abscesses of primarily parapharyngeal origin presenting with a chief concern of tongue protrusion.

Patients with varying levels of immune deficiencies are known to have more subtle presenting symptoms and potentially even diminished signs of systemic toxicity [1]. It is likely our patient's infection was able to undergo such extensive spread as to result in tongue protrusion because of her depressed B and T cell function. She continued to be energetic and non-toxic throughout her presentation, with the only localizing symptom being her oral examination, which was initially interpreted as tongue swelling rather than tongue protrusion. She had no history of similar infections or prior imaging to suggest a lymphovascular malformation that may have suggested a predisposition to deep neck space infections. At the same time, the misinterpretation of tongue protrusion as tongue swelling, and the fact that she had at least one known risk factor for developing angioedema lead the team to an incorrect initial diagnosis.

While the majority of angioedema cases in pediatrics are mast-cell mediated and are associated with other allergic symptoms, bradykinin-induced angioedema can result in more isolated symptoms [11]. ACE-inhibitors are a known cause of angioedema in children and can have a latency period between one day and 10 years [11, 12]. Given her daily enalapril use, this was considered high on her initial differential diagnosis.

Differentiating between isolated angioedema and other causes of tongue protrusion can be challenging with the clinical examination alone, particularly in the pediatric population and in populations with developmental delays who are often unable to fully communicate their

symptoms and frequently have difficulty cooperating during the evaluation [5]. These challenges can be further exacerbated in patients with underlying oropharyngeal abnormalities such as the macroglossia often associated with Trisomy 21 and other genetic syndromes [13]. Emergency physicians should maintain a high degree of suspicion and use advanced imaging when the diagnosis remains unclear, and consider tongue protrusion as a possible early sign of deep neck space infection. For this, CT imaging remains the gold standard by providing quick and accurate anatomic information without the field-of-view or technician-related limitations associated with ultrasound [4].

Another important component of this case was the role of cognitive biases. In the setting of an impressive exam finding that seemed isolated to the tongue, the medical teams anchored on ACE-inhibitor induced angioedema. The intermittent history of fevers for nine days (but absent on presentation) and leukocytosis, while considered, were attributed to a confounding previous diagnosis of a URI, but should have prompted a fever work-up for occult infection. The elevated CRP was difficult to interpret as this has also been documented with ACE-inhibitor induced angioedema [14]. There was also likely a recency bias as this was in the midst of respiratory season and prolonged, intermittent fevers were being seen frequently in children with back-to-back viral infections. Physicians throughout her admission, anchored on angioedema at the exclusion of other etiologies, particularly as the patient showed improvement on steroids with no respiratory distress.

This case represents both a unique early presentation of a common but serious disease process, and demonstrates the clinical challenge of evaluating tongue abnormalities and the dangers of cognitive biases in pediatric patients. EM providers should remember that tongue displacement may mimic glossal edema, and maintain a high degree of suspicion for deep neck space infections when evaluating young children who are unable to provide a complete description of symptoms or a cooperative examination. When the diagnosis is unclear, advanced imaging should be employed as the consequence of missed or delayed diagnosis can be severe.

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Figure 1: Photograph of patient demonstrating tongue protrusion.

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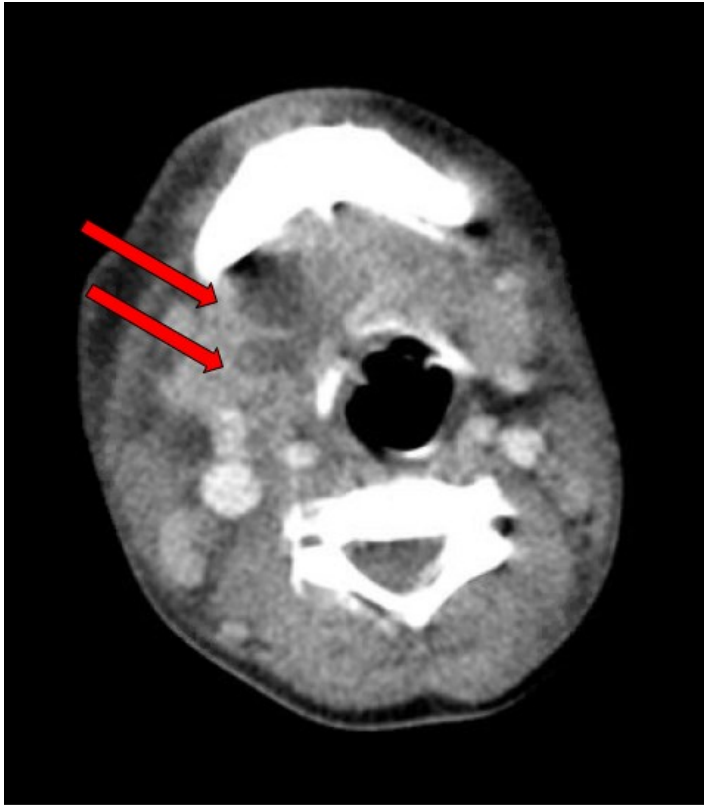


Figure 2: Axial neck CT image demonstrating rim-enhancing collection involving the right parapharyngeal, peritonsillar, and sublingual spaces with mass effect, without significant narrowing of the airway.

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