

CASE REPORT

Parenchymatous glossitis: A case report

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Key Clinical Message

We present a case of parenchymatous glossitis with unilateral severe inflammation of the hyoglossus muscle, resulting in laryngeal edema. The route of inflammation was unique. Contrast-enhanced CT was useful for diagnosis. Administration of sensitive antibiotics led to improvement. Creatine phosphokinase may be an indicator of diagnosis and treatment response.

KEYWORDS

creatine phosphokinase, hyoglossus muscle, lingual abscess, lingual cellulitis, parenchymatous glossitis

1 | INTRODUCTION

Parenchymatous glossitis is a diffuse pyogenic inflammation of the tongue parenchyma without pus accumulation, whereas lingual abscess is a pyogenic inflammation with pus accumulation in the tongue parenchyma. As both diseases are rare, they are rarely encountered by clinicians. Typical classical parenchymatous glossitis causes rapid entire tongue swelling, resulting in an inability to articulate and swallow, and often results in inflammatory edema that spreads to the larynx and causes asphyxia.¹⁻³ Even in recent years, lingual abscesses have been reported. On the other hand, parenchymatous glossitis has not been reported.⁴⁻⁸ The pathogenesis, the route of inflammation, and blood tests for parenchymatous glossitis have rarely been discussed in the previous literature. In this report, we present a case of hemi-parenchymatous glossitis with severe inflammation of the hyoglossus muscle. The findings of this case provide a foundation for constructive discussion of these issues.

2 | CASE HISTORY

A 78-year-old male presented with fever, dysphagia, swelling of the right side of the neck, and hoarseness and had developed fever for 3 days, dysphagia for 2 days, and right cervical swelling and hoarseness for 1 day before being referred to our department. He had nephrotic syndrome and was taking prednisolone (2.5 mg/day). Additionally, he had a history of glottic carcinoma after radiotherapy and diabetes mellitus (DM). He had visited another hospital 2 days before the referral and received blood tests and 2.0 g/day of ceftazidime hydrate for 2 days.

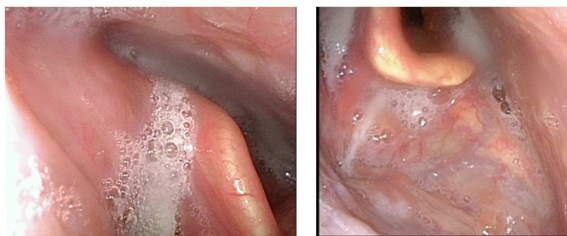
3 | INVESTIGATIONS AND TREATMENT

The swelling was observed on the right side of the tongue, floor of the mouth, and submandibular area. Laryngoscopy revealed redness and swelling of the right

pharyngoepiglottic fold and edema of the arytenoid and right supraglottic regions (Figure 1A). No trauma to the tongue was observed. A dental examination ruled out odontogenic infection.

The blood test 2 days before referral showed a white blood cell (WBC) count of $4.97 \times 10^3/\mu\text{L}$ (77.1% neutrophil), creatine phosphokinase (CPK) 758 U/L, C-reactive protein (CRP) 1.51 mg/dL, and amylase 107 U/L. The

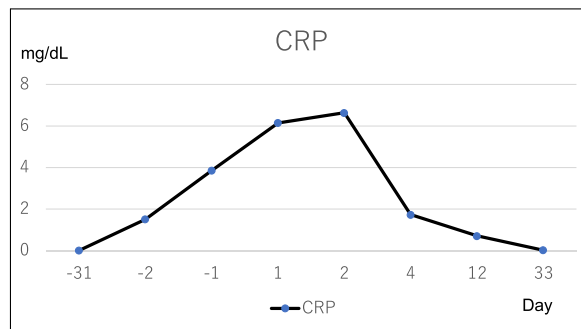
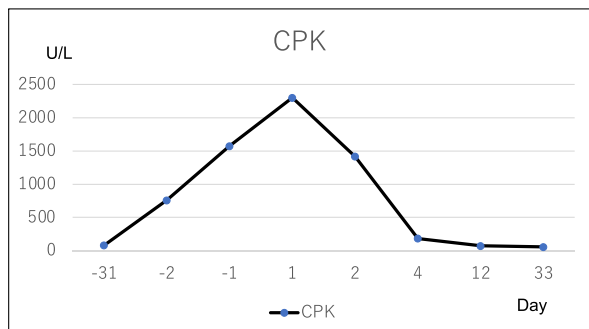
(A) Larynx and pharynx findings on admission



(B) Tongue, larynx, and pharynx findings on the 4th day of hospitalization



FIGURE 1 (A) Larynx and pharynx findings on admission show redness, swelling, and edema of the right pharyngoepiglottic fold, arytenoid, and right supraglottic regions. (B) Tongue, larynx, and pharynx findings on the 4th day of hospitalization show a concentration of tongue inflammation and improvement of laryngeal edema.



	Day 31 before admission	Day 2 before admission	Day 1 before admission	Admission Day 1	Day 2	Day 4	Day 12	Day 33	Normal level
CPK	82	758	1573	2301	1417	183	73	58	59-248 (U/L)
CRP	0.01	1.51	3.86	6.15	6.64	1.74	0.71	0.03	0-0.14 (mg/dL)
WBC	3.25	4.97	4.80	4.76	3.22	4.46	4.37	2.81	$3.90-9.80 (\times 10^3/\mu\text{L})$
Neutrophil	59.5	77.1	NA	80.5	83.2	NA	NA	57.0	40.0-74.0 (%)
Amylase	NA	107	77	NA	NA	NA	NA	NA	39-134 (U/L)

NA: not applicable Red: high level

FIGURE 2 Results of blood test. At onset, blood tests show a worsening trend in CPK and CRP and normal WBC counts. After therapy, CPK and CRP tend to decrease, and CPK returns to normal levels faster than CRP. creatine phosphokinase, CPK; C-reactive protein, CRP; white blood cell, WBC; NA, not applicable. Red shows high levels.

blood test on the day of the visit showed a WBC count of $4.76 \times 10^3/\mu\text{L}$ (80.5% neutrophil), CPK 2301 U/L, CRP 6.15 mg/dL, and a worsening trend in CPK and CRP (Figure 2). Blood cultures were negative. The contrast-enhanced CT findings were as follows. (1) The right side of the tongue parenchyma, excluding the anterior part, was diffusely inflamed and partially extended beyond the lingual septum. (2) The right hyoglossus muscle was significantly thickened along its entire length from the sublingual space to the hyoid bone. (3) Inflammation spread to the submandibular and visceral spaces, resulting in laryngeal edema (Figures 3 and 4).

Based on the contrast-enhanced CT findings, exclusion of odontogenic infection, and blood test results, we diagnosed hemi-parenchymatous glossitis with laryngeal edema. He was admitted to the hospital on the day of the visit and received 1.0 g/day of meropenem hydrate for 6 days and 6.6 mg/day of dexamethasone sodium phosphate for 2 days.

4 | OUTCOME AND FOLLOW-UP

The findings showed an improvement in conditions, and on the 4th day of hospitalization, the tongue inflammation was concentrated on the right lingual margin, where the hyoglossus muscle ends, and the laryngeal edema improved. (Figure 1B). He was discharged on the 7th day of hospitalization because blood tests and subjective symptoms revealed an improvement. No recurrence was observed at 12 months.

5 | DISCUSSION

The tongue is not naturally susceptible to infection. The reasons are (1) the muscular tissue that makes up most of the parenchyma and its rich vascular supply, (2) the rich lymphatic drainage, (3) the regularity of the tongue, which allows saliva to cleanse it continuously, (4) the thick covering of keratinous mucous membranes, and (5) the immunological properties of saliva.⁴ Pyogenic inflammation of the tongue parenchyma, such as parenchymatous glossitis and lingual abscess, is rare. When host defense mechanisms are compromised, these diseases are more likely to occur following trauma, foreign bodies, and infection.³⁻⁷ This patient had DM, nephrotic syndrome, was taking prednisolone, and was thought to be susceptible to infection.

We searched for articles using the keywords “parenchymatous glossitis,” “profound glossitis,” “phlegmonous glossitis,” “tongue phlegmon,” “deep glossitis,” “interstitial glossitis,” and “lingual cellulitis”¹⁻³ through PubMed and Google Scholar from 1943, when penicillin became widely available, to 2023. Consequently, two articles were published in English.^{7,8} Typical classical parenchymatous glossitis is characterized by rapid swelling of the entire tongue protruding between the teeth, accompanied by severe pain. The disease progresses to suppuration and lingual abscess. The suppurative process, if not drained early, may extend to the pharynx or larynx.¹⁻³ However, the last case of such typical classical parenchymatous glossitis progressing to lingual abscess has not been reported since 1965.⁸

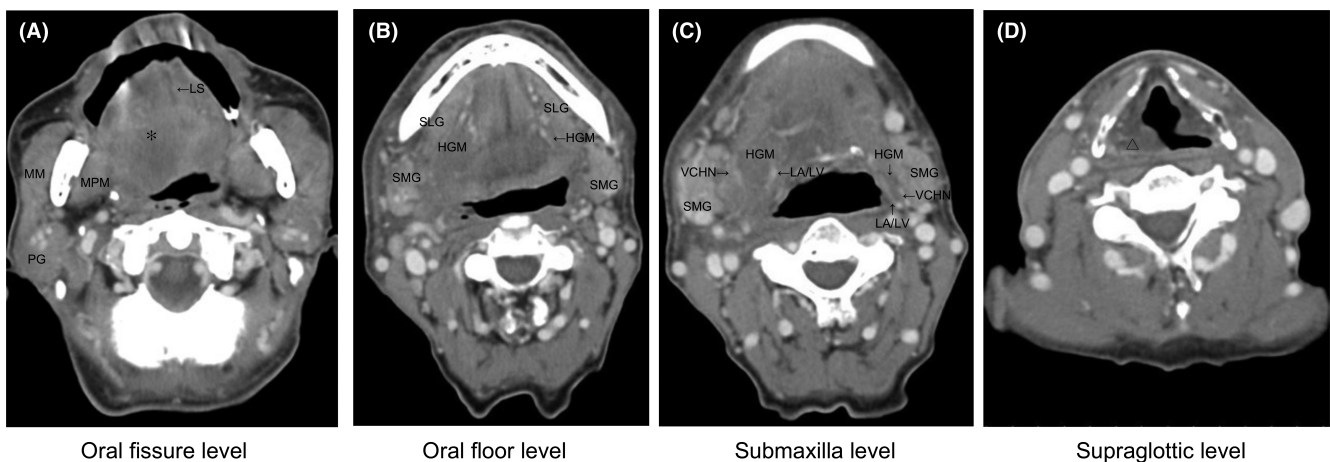


FIGURE 3 Axial CT images. (A) Oral fissure level. (B) Oral floor level. (C) Submaxilla level. (D) Supraglottic level. The CT images reveal the following findings: The right tongue parenchyma is diffusely inflamed. The right hyoglossus muscle is significantly thickened along its entire length. Inflammation spreads to the submandibular and visceral spaces. HGM, hyoglossus muscle; MM, masseter muscle; MPM, medial pterygoid muscle; LS, lingual septum; PG, parotid gland; SMG, submandibular gland; SLG, sublingual gland; LA, lingual artery; LV, lingual vein; VCHN, vena comitans of hypoglossal nerve. Asterisk reveals diffuse inflammation of the tongue parenchyma. Triangle reveals laryngeal edema.

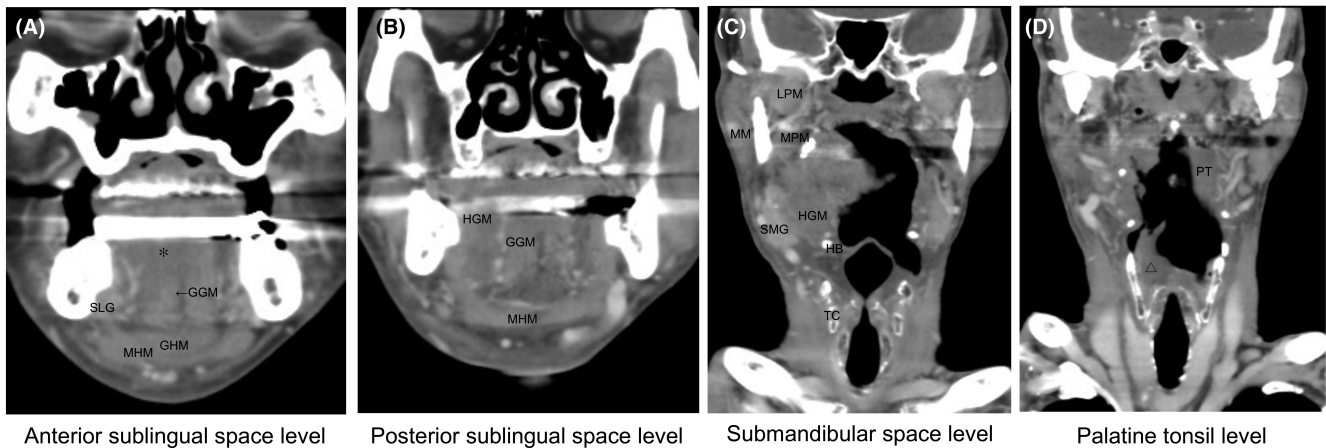


FIGURE 4 Coronal CT images. (A) Anterior sublingual space level. (B) Posterior sublingual space level. (C) Submandibular space level. (D) Palatine tonsil level. GGM, genioglossus muscle; GHM, geniohyoid muscle; HGM, hyoglossus muscle; MHM, mylohyoid muscle; MM, masseter muscle; MPM, medial pterygoid muscle; LPM, lateral pterygoid muscle; SMG, submandibular gland; SLG, sublingual gland; PT, palatine tonsil; HG, hyoid bone; TC, thyroid cartilage. Asterisk reveals diffuse inflammation of the tongue parenchyma. Triangle reveals laryngeal edema.

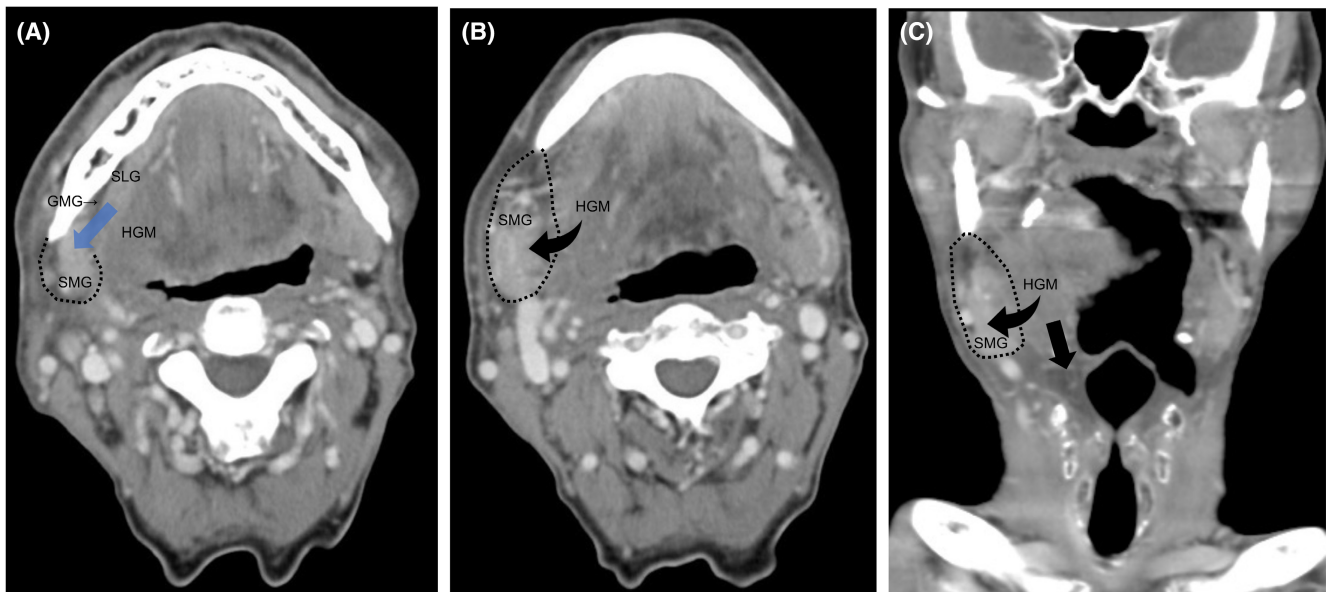


FIGURE 5 Route of inflammatory spread. (A) The blue arrow reveals that the route of inflammation of the sublingual space spreads to the submandibular space through the glosso-mylohyoid gap. (B, C) The black arrows reveal that the route of inflammation of the hyoglossus muscle spreads to the submandibular and visceral spaces. GHM, geniohyoid muscle; HGM, hyoglossus muscle; SMG, submandibular gland; SLG, sublingual gland; Dot black line reveals the superficial layer of the deep cervical fascia that forms the submandibular space.

The following three important clinical issues were identified: (1) Hemi-parenchymatous glossitis with severe inflammation of the hyoglossus muscle is a type of parenchymatous glossitis. (2) Inflammation of the hyoglossus muscle can spread to the adjacent submandibular and visceral spaces. Inflammation spreading to the visceral space results in laryngeal edema. (3) CPK may be an indicator of diagnosis and treatment response in parenchymatous glossitis.

Hemi-parenchymatous glossitis with severe inflammation of the hyoglossus muscle is a type of parenchymatous glossitis. In 2003, Antoniadis et al. reviewed 50 cases of lingual abscesses published in the English literature over the past 30 years.⁵ Even after that, lingual abscesses have been reported as an uncommon disease.⁶ On the other hand, there have been no reports of parenchymatous glossitis before lingual abscess formation in the past 50 years. This case may indicate a condition in which

parenchymatous glossitis occurs before lingual abscess formation and a type of parenchymatous glossitis.

Inflammation of the hyoglossus muscle can spread to the adjacent submandibular and visceral spaces. Inflammation spreading to the visceral space results in laryngeal edema. The hyoglossus muscle is one of four extrinsic muscles of the tongue. It originates from the lateral region of the anterior aspect of the body of the hyoid bone and ends at the lingual margin. It is located deep in the submandibular space.^{9,10} In this case, the submandibular space was swollen. This could be caused by inflammation of the hyoglossus muscle, odontogenic infection, and sialadenitis (submandibular adenitis or sublingual adenitis). Since odontogenic infection was ruled out and amylase was not elevated at onset, we diagnosed inflammation of the hyoglossus muscle as the cause. In addition, we believe that inflammation of the hyoglossus muscle spread to the adjacent visceral space on the caudal side of the hyoid bone, resulting in laryngeal edema. There is no fascia between the posterior part of the sublingual space and the submandibular space, and this area is called the glosso-mylohyoid gap.^{10–12} Inflammation of the sublingual space caused by odontogenic infection and sublingual adenitis spreads to the submandibular space through the glosso-mylohyoid gap and further to the parapharyngeal space.^{11,12} In this case of parenchymatous glossitis, inflammation spreads from the hyoglossus muscle to the submandibular and visceral spaces, resulting in laryngeal edema. This route differs from the glosso-mylohyoid gap route (Figure 5). The hyoglossus muscle may occupy a crucial position in the route of inflammatory spread of pyogenic inflammation of the tongue parenchyma.

CPK may serve as an indicator of diagnosis and treatment response in parenchymatous glossitis. CPK is an enzyme present in large amounts in muscles (striated, cardiac, and smooth muscles), and when muscle damage occurs, CPK is released from the muscle cells into the blood, resulting in high levels.¹³ The muscles of the tongue are composed of striated muscles. CPK levels are expectantly high in cases of parenchymatous glossitis and lingual abscess, which cause inflammation of the tongue muscles. However, reports of CPK in parenchymatous glossitis or lingual abscess are not found. In this case, CPK and CRP levels were high at onset. CPK tended to decrease the following day, but CRP remained high. In addition, CPK returned to normal levels faster than CRP. (Figure 2). A few cases with high CPK levels are reported in patients with pyomyositis.¹⁴ CPK is an indicator of therapeutic efficacy in polymyositis and dermatomyositis, which are typical inflammatory muscle diseases.¹⁵ Based on the changes in CPK in this case, we suggest that CPK may serve as an indicator of diagnosis and treatment response. On the other hand, WBC count is not expected to be helpful in the diagnosis of pyogenic inflammation of the tongue

parenchyma. In this case, the WBC count was normal at onset, and it has been reported that most patients with lingual abscesses usually have a normal WBC count or mild leukocytosis.⁴

Contrast-enhanced CT was useful in the diagnosis of parenchymatous glossitis. Differential diagnosis between parenchymal glossitis, oral floor cellulitis, and lingual abscess is difficult based on the course and local findings. In this case, parenchymatous glossitis was diagnosed based on contrast-enhanced CT findings. MRI is also useful in the differential diagnosis. However, MRI is time-consuming, and contrast-enhanced CT is safer in dangerous cases with rapid airway obstruction.⁴

6 | CONCLUSION

This case report highlights hemi-parenchymatous glossitis with severe inflammation of the hyoglossus muscle. The involvement of the hyoglossus muscle can lead to unique complications such as laryngeal edema, underscoring the importance of recognizing this variant in clinical practice. In addition, CPK may serve as a potential indicator of diagnosis and treatment response in parenchymatous glossitis, although further research is needed to validate this potential diagnostic utility.

AUTHOR CONTRIBUTIONS

Masami Suzuki: Project administration; writing – original draft. **Kohtaro Eguchi:** Writing – review and editing. **Naohiro Yoshida:** Project administration; writing – review and editing.

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None.

CONFLICT OF INTEREST STATEMENT

The authors declare that there are no conflicts of interest.




DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available from the corresponding author upon reasonable request.

CONSENT

Written informed consent was obtained from the patient to publish this report in accordance with the journal's patient consent policy.

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