*Case Report*

# Lingual histiocytic sarcoma in a dog: A case report

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*Abstract*

An 8-year-old spayed Maltese dog was presented for the evaluation of a lingual mass. The dog had excessive drooling and dysphagia. Oral examination revealed a soft, fluctuating mass at the left side of the tongue. Computed tomography showed a 21 × 32 × 66 mm lingual mass and enlarged left medial retropharyngeal lymph node. On cytology of the lingual mass, a round cell tumor was suspected; however, it was not responsive to glucocorticoid therapy. Because of severe complications, glossectomy and lymphadenectomy were performed; unfortunately, the patient died because of dyspnea in the postoperative period. Histopathologic examination and immunohistochemistry revealed lingual histiocytic sarcoma and confirmed lymphatic metastasis. This is a rare case report of a dog with lingual histiocytic sarcoma diagnosed based on histopathology and immunohistochemistry.

**Keywords:** Lingual, Histiocytic sarcoma, Immunohistochemistry, Dog

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## Introduction

Neoplasia confined to the tongue is very rare. In one study, 64% of lingual neoplasm was malignant (Vail *et al*., 2019). The clinical signs of lingual tumors vary depending on the extent of the tumor and are discovered incidentally in approximately 25% of cases (Beck *et al*., 1986). Clinical signs associated with tumors include oral hemorrhage, halitosis, hypersalivation, difficulty breathing or swallowing, and anorexia (Culp *et al*., 2013). Surgical resection is the primary treatment option for lingual neoplasia; however, radiation therapy can be considered for melanoma, inoperable cancer, or regional lymph metastasis (Vail *et al*., 2019).

Canine histiocytic sarcoma (HS) is a rare neoplasia of the lymphoreticular system *(Vail et al*., 2019). HS is common in middle-aged to older dogs and has a high prevalence in several dog breeds, especially in Bernese Mountain Dogs and Flat-Coated Retrievers (Erich *et al*., 2022). HS can be classified into localized HS, in which tumors develop from a single anatomical site, and disseminated HS, which affects multiple organs. Canine HS has an aggressive biological behavior and fatal outcomes (Dervisis *et al*., 2017). HS is diagnosed based on cytologic or histopathologic examination. Additional tests, such as immunohistochemistry (IHC) staining, may be required because neoplastic cells can look similar to other types of cancer (Vail *et al*., 2019). The histopathological characteristics of HS are sometimes indistinguishable from other round-cell tumors, such as lymphoma, poorly differentiated mast cell tumors, and carcinoma (Thongtharb, 2018). IHC helps to make a definitive diagnosis and guide prognostic assessment and available treatment options (Fulmer and Mauldin, 2007).

This case report describes the clinical course of a dog with lingual HS diagnosed based on histopathology and IHC.

## Case description

An 8-year-old spayed Maltese dog was presented to a local with symptoms of excessive drooling. Oral examination revealed a lingual mass, and a fine-needle aspiration was performed. However, the results were undiagnostic because of blood contamination. The dog was treated with prednisolone (2 mg/kg/day) and clindamycin for 2 weeks; however, no clinical response was observed, and the lingual mass became larger.

After 1 month, the dog was referred for definitive diagnosis and treatment. On hospital presentation, the dog showed dysphagia and hypersalivation because of a large lingual mass. Physical examination revealed a 2-cm soft and fluctuant mass at the left side of the tongue (Fig 1.). Findings of complete blood count and serum chemistry were not significant, except for elevated liver enzymes. This was attributed to the steroid administration.

Computed tomography (CT) was performed to evaluate the size and margin of the lingual mass accurately and confirm metastasis. CT revealed a diffuse, relatively ill-marginated, thickened, and heterogeneously hyperattenuating lingual mass (21 × 32 × 66 mm) with heterogeneous contrast enhancement. An enlarged, elongated left medial retropharyngeal lymph node (LN) (16 × 8 × 40 mm) with contrast enhancement was also identified (Fig 2.). No evidence of pulmonary and abdominal organ metastasis was noted. FNA of lingual mass was performed under general anesthesia. On cytological examination of the lingual mass, clustered round cells exhibited mild-to-moderate anisocytosis and anisokaryosis; thus, round cell tumors such as lymphoma, histiocytic tumor, or amelanotic melanoma were suspected (Fig 3.).

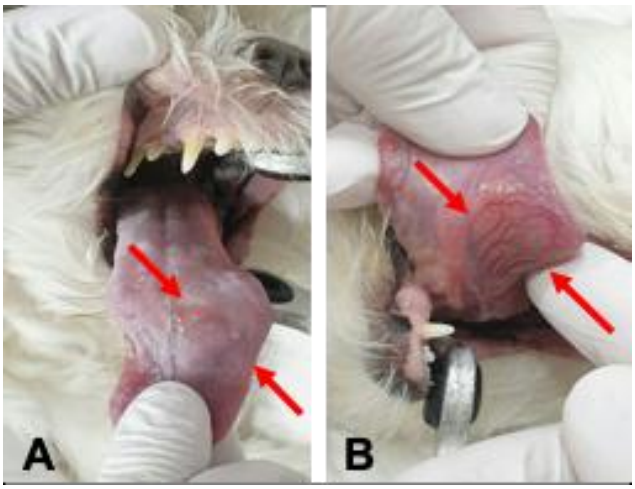


**Figure 1** Gross findings of the lingual mass in a dog with clinical signs of excessive drooling and dysphagia. The mass was soft and fluctuant. Swelling with vascularization was found on the left lateral aspect of the tongue (Red arrows).

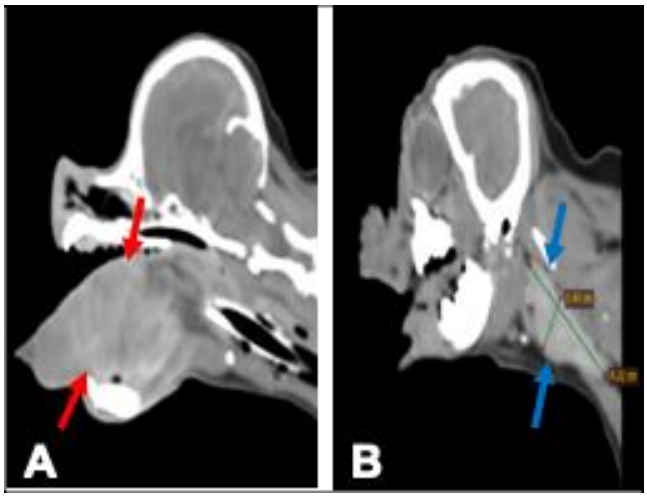


**Figure 2** Sagittal CT images showing a lingual mass (A, red arrows) and left medial retropharyngeal LN (B, blue arrows). (A) Ill-marginated, heterogeneously hyperattenuated, thickened mass is found in the tongue. (B) Enlarged and elongated Lt. medial retropharyngeal LN with contrast enhancement.

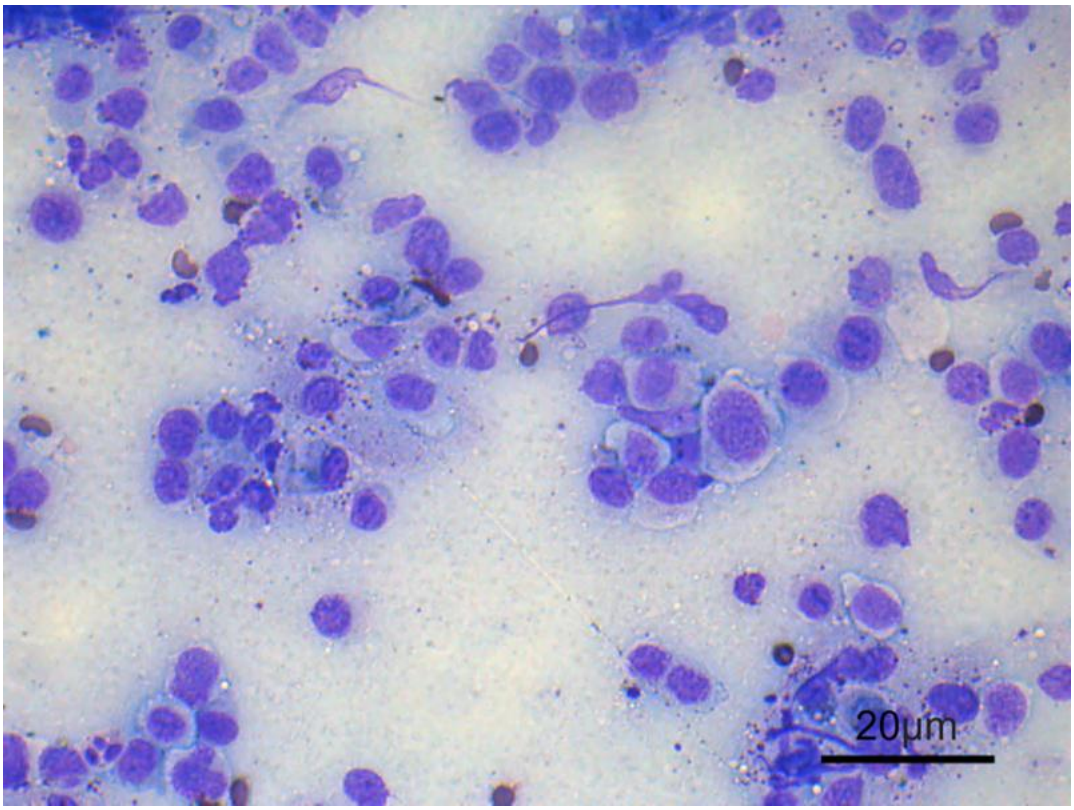


**Figure 3** Cytological examination of the lingual mass. Clustered round cells exhibited anisocytosis and anisokaryosis; thus, round cell tumors such as lymphoma, histiocytic tumor, or amelanotic melanoma were suspected.

Glossectomy and left retropharyngeal lymphadenectomy were performed because the mass caused severe clinical signs and affected the quality of life. The anesthesia was induced with propofol (5 mg/kg intravenously) and maintained with isoflurane. The dog was positioned in dorsal lateral recumbency, and gauze was placed inside the mouth to prevent aspiration pneumonia. Stay sutures (3-0 PDS) were placed at both sides of the tongue, and a glossectomy was performed with electrocautery. The lingual artery, nerve, frenulum, and cutting edge were ligated with 4-0 PDS. After glossectomy, the dog was positioned in right lateral recumbency for left retropharyngeal lymphadenectomy. Incisions of the skin subcutaneous tissue were made, and the retropharyngeal lymph node was separated by blunt dissection. Stay sutures (4-0 PDS) were placed in the lymph node, and lymphadenectomy was performed with electrocautery. On gross examination, the lingual mass was firm, and the cut surface was yellowish-white (Fig 4.). The resected mass and LN were fixed in 10% formalin for histopathological examination.

Postoperative treatment included maintenance intravenous fluid therapy (Plasma Lyte A injection), prednisolone (0.5 mg/kg, subcutaneously), fentanyl continuous rate infusion (2-6 μg/kg/hr), ampicillin (22 mg/kg, intravenously), famotidine (1mg/kg, intravenously), maropitant citrate (1mg/kg,

intravenously). An ice pack was applied periodically on the surgical site to relieve perioperative edema. On the day after the surgery, the dog presented labored breathing. To relieve pain, the rate of fentanyl CRI was increased. Oxygen supply using an oxygen mask and removal of mucous discharge in the oral cavity improved respiratory distress. However, the dog presented open-mouth breathing and orthopnea after 2 hours. Endotracheal was performed, and severe laryngeal edema with serosanguinous discharge was identified during intubation. The patient underwent cardiopulmonary arrest and died.

Histopathological examination of the resected mass and LN confirmed HS (Fig 5.). The mass was mainly composed of neoplastic round cells that replaced the normal architecture of the tongue; cells were arranged in sheets, and some of these cells were arranged in cords. Neoplastic cells were characterized by ovoid- to bean-shaped nuclei, high cellularity, prominent nucleoli, and marked pleomorphism. Mitotic figures were observed very frequently (33 mitoses per 10 HPF). The left retropharyngeal LN was composed of the same neoplastic cells with marked pleomorphism. Mitotic figures were observed frequently as well. Because other round cell tumors and carcinoma, which may differ in treatment and prognosis, are often misdiagnosed as HS, additional toluidine blue and IHC staining procedures were performed for definitive diagnosis. Toluidine blue, Mekan-A, S-100, and pan-cytokeratin IHC stains were performed to distinguish from mast cell tumors, malignant melanoma, and carcinoma, respectively. Neoplastic cells stained negative for toluidine blue, Melan-A, pan-cytokeratin, and S-100 (Fig 6.). Based on the above results, the dog was diagnosed with lingual HS with retropharyngeal LN metastasis.

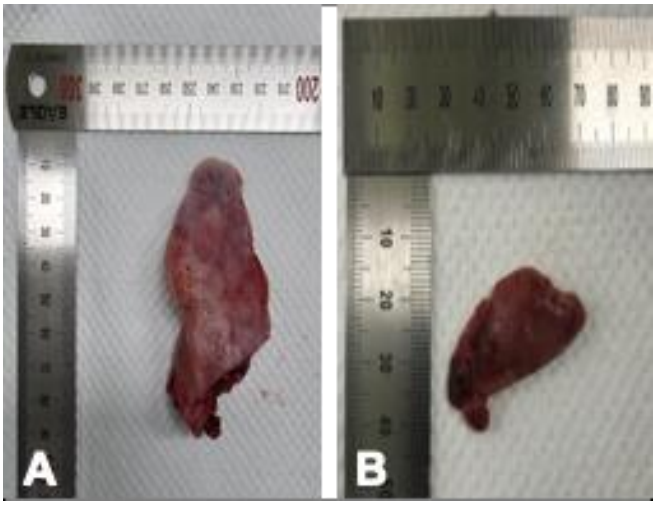


**Figure 4** Surgically resected lingual tumor, 42 x 80 mm in size (A), and left retropharyngeal lymph node, 22 x 41 mm in size (B). The mass was firm, and the cut surface was yellowish-white.

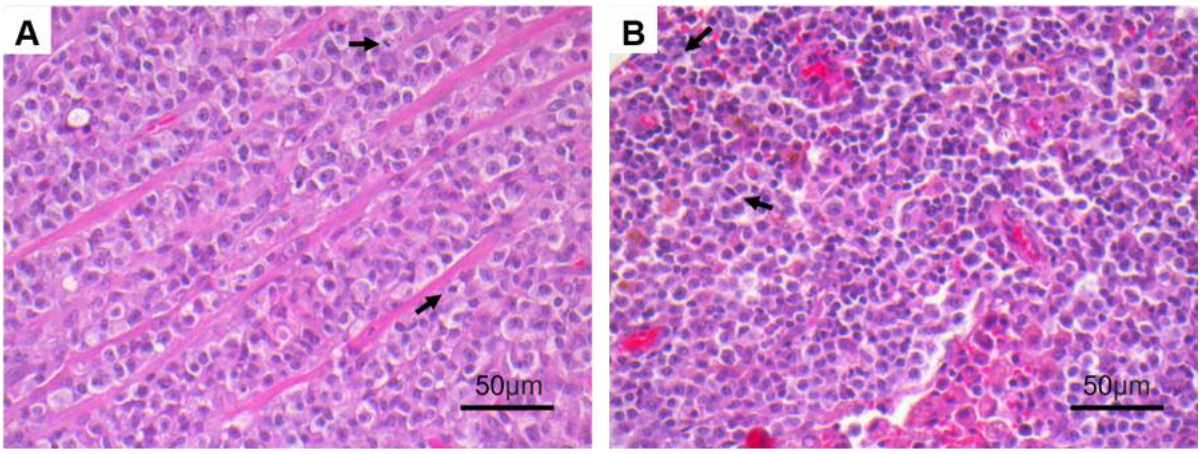


**Figure 5** Histopathological examination of the resected mass (A) and lymph node (B) Neoplastic cells (black arrow) were characterized by ovoid- to bean-shaped nuclei, high cellularity, prominent nucleoli, and marked pleomorphism.

## *Discussion*

HS, a malignant tumor of histiocytic origin, is characterized by neoplastic proliferation of dendritic cells and macrophages (Fulmer and Mauldin, 2007). Although HS has been identified in multiple locations and lingual histiocytic sarcoma was reported in a study, no case report has been reported previously. (Affolter and Moore, 2002)

The clinical signs of HS vary depending on the site of the tumor involvement. Commonly reported signs are nonspecific; weight loss, lethargy, and inappetence are common (Skorupski *et al*., 2007). In this report, the tumor invaded the tongue, and hypersalivation

accompanied by dysphagia for several weeks was the chief complaint.

A complete physical examination, blood test, and diagnostic imaging (radiography, ultrasonography, CT, or magnetic resonance imaging) are performed to evaluate the tumor type and determine the treatment options and prognosis.

HS is diagnosed based on cytologic and histologic examination findings. The cytologic features of HS include pleomorphic, large, discrete mononuclear cells with prominent nucleoli and marked anisokaryosis. The cytoplasm of the neoplastic cells is moderate to abundant, lightly basophilic, and vacuolated. The histopathologic characteristic of HS is marked cellular atypia (Affolter and Moore, 2002). However, these cytologic or histopathologic characteristics are similar to other round cells or carcinoma; thus, the definitive diagnosis of HS can be challenging, particularly in pleomorphic cases. Therefore, additional tests such as immunocytochemistry or IHC are necessary to support the diagnosis of HS (Vail *et al*., 2019).

In this case, histopathologic examination was performed for resected lingual tumor and LN. The mass was mainly composed of neoplastic round cells, and neoplastic cells were characterized by ovoid- to bean-shaped nuclei, prominent nucleoli, and marked pleomorphism. Additional toluidine blue staining and IHC (Melan-A, pan-Cytokeratin, and S-100) were used to distinguish from other poorly differentiated round cell tumors or tumors of epithelial origin. In one study, of the 355 biopsy samples initially diagnosed as suspected HS, only 62.7% had HS confirmed by IHC (Dervisis *et al*., 2017).

Toluidine blue staining is an established method for the identification and quantification of mast cells (Puebla-Osorio *et al*., 2017). Melan-A is a highly specific and low-sensitive melanocytic marker and is useful in distinguishing melanoma from other poorly differentiated carcinoma or sarcoma (Saverino *et al*., 2021). S-100 is a nonspecific marker for melanocytic tumors and peripheral nerve sheath tumors and is positive in all melanoma subtypes (Jungbluth and Busam, 2018). As S-100 negative melanomas are rare, this is one of the most sensitive markers for melanoma. Pan-cytokeratin is an epithelial cell marker, and negative results for pan-cytokeratin help rule out tumors of epithelial origin. In this case, based on the negative results for toluidine blue, Melan-A, S-100, and pan-cytokeratin staining, the dog was diagnosed as primary lingual HS with LN metastasis.

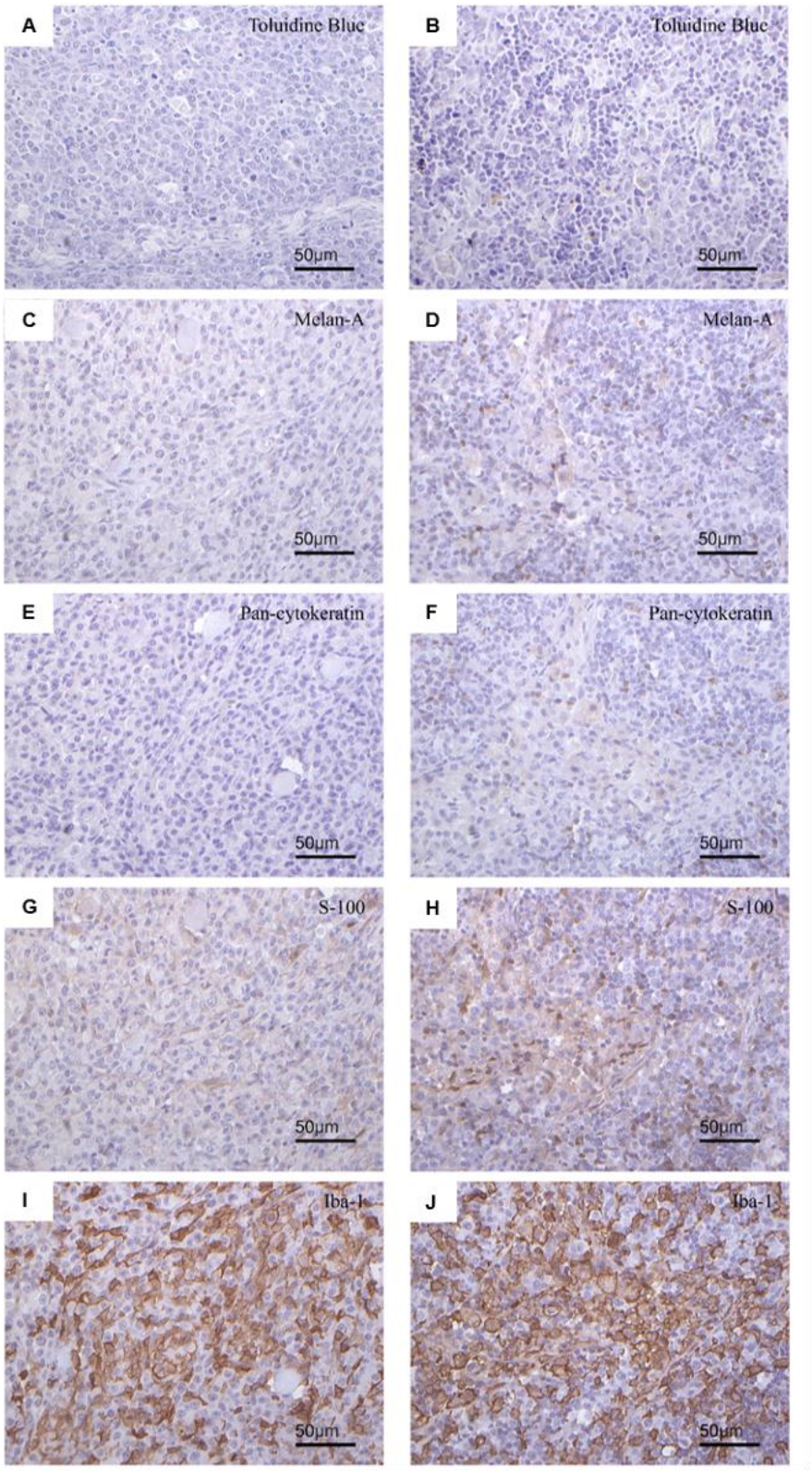


**Figure 6** Additional staining for resected mass (A, C, E, G, and I) and lymph node (B, D, F, H, and J).

The recommended treatment option for localized HS is wide surgical resection. Additional chemotherapy is indicated following surgery for localized HS and primary treatment options for disseminated HS (Cannon *et al*., 2015).

In this case report, the tumor caused severe clinical symptoms that affected the quality of life. To improve the quality of life and determine the tumor type, surgical resection of the lingual tumor and regional LN was performed. Unfortunately, the patient presented dyspnea and exhibited cardiopulmonary arrest the day after the surgery. The cause of death was thought to be severe postoperative edema.

As mentioned above, lingual HS is extremely rare but sufficiently possible. As treatment options vary depending on the type of tumor, accurate diagnosis is important. Therefore, to effectively manage patients with a tongue mass, the clinician should conduct an

accurate physical examination, diagnostic imaging, and histopathologic tests, including immunostaining.

In summary, this report presented a rare case of primary lingual HS in a dog. The dog presented severe drooling and dysphagia caused by the tumor, and CT revealed that the tumor primarily originated from the tongue. Based on the histopathological examination and IHC, the dog was diagnosed with primary lingual HS with local LN metastasis. As both lingual and HS rarely occur, very little is known about their clinical course, treatment, and prognosis. Unfortunately, the dog died during the postoperative period, and the evaluation of response to therapy and progression-free interval was not possible. Further case series studies are needed to define the treatment and prognosis of this tumor.

***Author contributions:*** All authors made the diagnosis and had direct patient contact. DH wrote the manuscript, and SG supervised the study. All authors revised the manuscript.

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