

Spindle cell lipoma in the gingiva of a dog: a case report

H.J. KIM¹, E.J. CHOI¹, H.R. LEE¹, G.J. PARK², E.S. YUN¹, J.H. KIM¹, S.H. DO¹

¹College of Veterinary Medicine, Konkuk University, Seoul, Republic of Korea

²Woo-Sin Animal Hospital, Seoul, Republic of Korea

ABSTRACT: Spindle cell lipoma is a benign lipomatous tumour that mainly occurs in the subcutis of the head, neck, and shoulder in elderly men. Oral spindle cell lipoma is extremely rare in all species, and no cases have been reported in veterinary medicine. A 10-year-old male Maltese dog was presented for evaluation of a white-to cream-coloured gingival mass. Microscopically, the mass was composed of mature fat cells and spindle cells containing thick fibrocollagenous bundles. The neoplastic cells showed positive immunoreactivity to vimentin and CD34. Based on the clinical and histopathological findings, spindle cell lipoma of the gingiva was diagnosed. Further immunohistochemical analysis revealed that both adipocytes and spindle cells exhibited positive reactions to oestrogen receptors α and β . These findings demonstrate the clinical, morphological, and immunohistochemical characteristics of an oral spindle cell tumour. The potential role of sex steroid hormones in the pathogenesis of this tumour is also discussed.

Keywords: oral cavity; spindle cell lipoma; CD34; vimentin; oestrogen receptor

Benign tumours of adipose tissue, known as lipomas, are the most common soft tissue tumours in all species (Meuten 2002; Al Sheddi et al. 2014). Lipomatous tumours mainly affect the subcutis of the back, shoulder, head, and neck (Said-Al-Naief et al. 2001). However, intra-oral lipoma is relatively rare, with an overall incidence reported to be 1% to 4% of all benign oral tumours in humans (Furlong et al. 2004; Allon et al. 2011). Oral lipomas can occur on the tongue, hard palate, floor of the mouth, cheek (buccal mucosa), lip, and gingiva (Ayasaka et al. 1993). In animals, only a few cases of oral lipoma/liposarcomas have been reported, including in the salivary gland (Clark et al. 2013), epiglottis (Carpenter 2012), larynx (Brunnberg et al. 2013), and tongue (Piseddu et al. 2011).

Lipomas are classified as conventional lipoma (simple lipoma), fibrolipoma, angiolipoma, myolipoma, pleomorphic lipoma, and spindle cell lipoma, according to their specific histological features (Fletcher et al. 2002; Gross et al. 2005). Spindle cell lipoma is a rare variant of lipoma that is characterised by admixture of collagen-forming spindle cells with mature adipose tissues. Spindle cell lipoma frequently occurs in the subcutis of the posterior neck and back; how-

ever, cases have been reported at the hip, elbow, ventral abdomen, inguinal region, and on the forehead (Enzinger and Harvey 1975; Al Sheddi et al. 2014). Oral spindle cell lipoma is rare in all species, and about 40 cases have been reported in humans. To the best of our knowledge, spindle cell lipoma of the gingiva has never been reported in veterinary medicine.

Clinically, spindle cell lipomas appear similar to simple lipomas; they are slow-growing, well-circumscribed, oval- or discoid-shaped, mobile masses that do not cause pain (Lombardi and Odell 1994; Darling et al. 2002). The differential diagnosis of spindle cell lipoma includes simple lipoma, fibrolipoma, myxolipoma, myxoid solitary fibrous tumour, atypical lipomatous tumours, schwannoma, and neurofibroma (Piattelli et al. 1999). Here, we report a case of spindle cell lipoma of the gingiva in a 10-year-old male Maltese dog. The clinical, histological, and immunohistochemical findings of this case are presented.

Case description

A 10-year-old male Maltese dog was presented to a private veterinary clinic for evaluation of a

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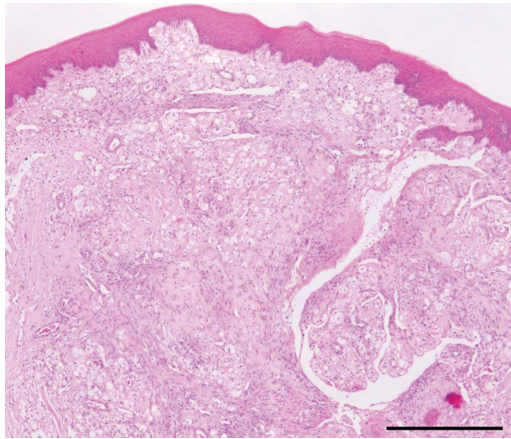


Figure 1. Histopathological features of the gingival specimen under low magnification. The majority of the dermal collagenous stroma was replaced by mature adipocytes and spindle cells with thick fibrocollagenous stroma (haematoxylin and eosin, bars = 500 μ m)

localised swelling on the gingival mucosa of the premolar teeth. Clinical examination revealed a yellowish-white round mass 5 mm in diameter, covered by intact oral mucosa. Findings of routine blood tests and radiography were unremarkable.

The excised specimen was immediately fixed in 10% neutral buffered formalin to enable histopathological evaluation. The specimen was embedded in paraffin and sliced into 4- μ m sections. Haematoxylin and eosin and Masson's trichrome stain were used to identify extracellular components. For immunohistochemical staining, the following primary antibodies were used: anti-vimentin (DakoCytomation, Glostrup, Denmark, 1:100), anti-S100 (Abcam, Cambridge, UK, 1:100), anti-oestrogen receptor α (Abcam, Cambridge,

UK, 1:100), anti-oestrogen receptor β (Santa Cruz Biotechnology, CA, USA, 1:100), and anti-CD34 (Abcam, Cambridge, UK, 1:100). The antibody-labelled sections were incubated with an avidin-biotin-peroxidase complex (ABC) solution using an avidin-biotin-peroxidase complex kit (Invitrogen, Carlsbad, CA, USA). 3,3'-diaminobenzidine (Zymed Laboratories, CA, USA) was used for visualisation, and the sections were counterstained with Mayer's haematoxylin.

Microscopic examination revealed the gingival mass to exhibit mild to moderate epidermal thickening with rete pegs formation. In the superficial dermis, spindle cells were admixed with mature adipocytes such that most of the lipocytes were separated by spindle cells containing eosinophilic fibrotic bundles. These components were distributed evenly, and neovasculture was also prominent. Most of the dermal collagenous stroma was replaced with mature adipocytes containing lipid globules which varied in size. Small uniform spindle cells were aligned around the lipomatous lesion (Figure 1).

At higher magnification, newly formed vacuolated cells could be seen in the border between mature lipocytes and fibrous tissues. These adipocytes had paracentric to centric, round to ovoid nuclei and were located within the fibrocollagenous background. Most of the adipocytes had clear cytoplasm which varied in size; lipoblasts were not seen. Spindle cells within the highly eosinophilic fibrous stroma exhibited single ovoid-to-elongated fusiform nuclei with poorly defined eosinophilic cytoplasm. Most of the eosinophilic stroma stained blue with Masson's trichrome, which ruled out proliferation of muscular components (Figure 2).

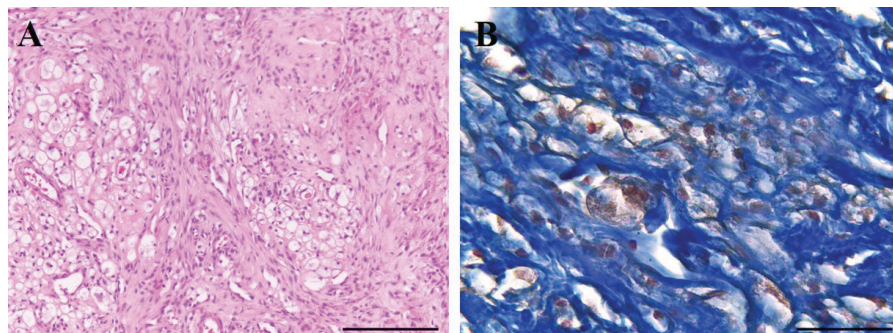


Figure 2. Histopathological features of the gingival specimen under higher magnification. Variably sized lipid globules containing mature adipocytes were surrounded with small uniform spindle cells with eosinophilic backgrounds; haematoxylin and eosin, bars = 200 μ m (A). Most of the eosinophilic connective tissue showed a positive response to collagen fibre staining; Masson's trichrome staining, bars = 100 μ m (B)

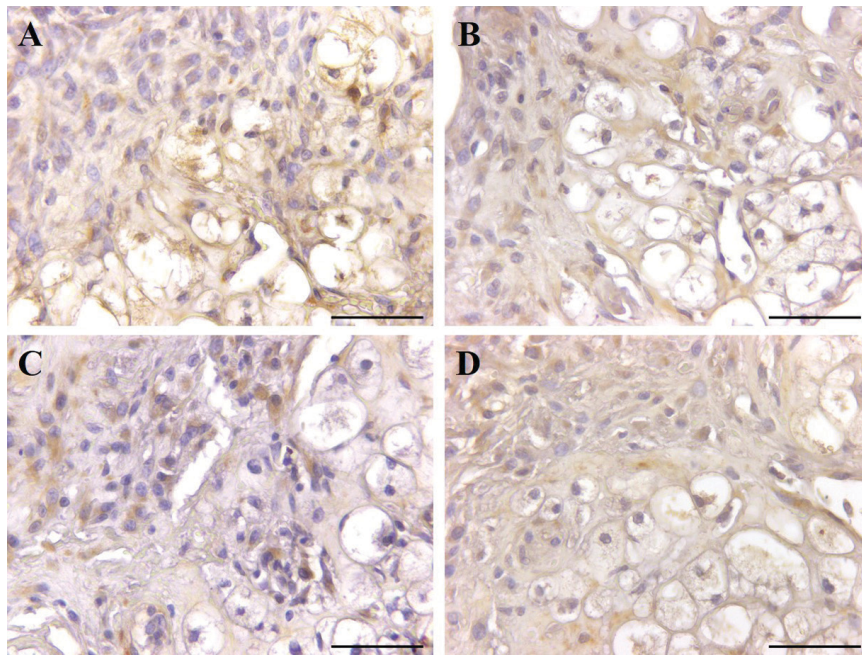


Figure 3. Immunohistochemical characterisation of tumour cells. Immunohistochemistry revealed a strong positive reaction for vimentin in the cytoplasm of tumour cells (A). The tumour cells also showed a positive immunoreaction to CD34 (B). Both spindle cells and mature adipocytes showed reactivity to oestrogen receptor alpha (C) and beta (D). A–D = immunohistochemistry, bars = 50 µm

The neoplastic lesion was further examined by immunohistochemistry (IHC) to confirm the characteristics of the adipocytes and spindle cells. Most of the spindle cells and vacuolated cells were positive for vimentin and CD34 (Figure 3A, B). On the other hand, the cells did not react with S100 (data not shown). Further immunohistochemistry revealed that both spindle cells and lipocytes exhibited positive reactions for oestrogen receptors α and β (Figure 3C, D). According to these histopathological findings, the final diagnosis was spindle cell lipoma of the gingiva consisting of mature adipocytes admixed with spindle cells.

DISCUSSION AND CONCLUSIONS

Spindle cell lipoma, first reported by Enzinger and Harvey in 1975, accounts for approximately 1.5% of all lipoma cases. Spindle cell lipoma predominantly occurs as a solitary, asymptomatic, slow-growing subcutaneous mass in the shoulder and posterior neck of elderly males without pain (Al Sheddi 2014). Lipomas represent 0.1 to 5% of all benign neoplasms of the oral cavity and have been reported at other anatomical locations such

as the buccal mucosa, tongue, floor of the mouth, and hard palate (Fregnani et al. 2003; Manor et al. 2011). Spindle cell lipomas, especially in the intra-oral region, occur so rarely that only a few cases have been reported in any species. Here, we present the first case of spindle cell lipoma of the gingiva reported in a dog.

Microscopically, spindle cell lipoma is characterised by the presence of mature fat cells admixed with spindle cells. However, the proportion of mature adipocytes to spindle cells can vary. Because of this, spindle cell lipoma is often misdiagnosed as simple lipoma (if predominantly composed of mature adipocytes with widely scattered spindle cells) or spindle cell tumour (if composed of numerous spindle cells with only a small number of mature adipocytes). Due to this histopathological heterogeneity, spindle cell lipoma must be distinguished from classical lipoma, myolipoma, fibrolipoma, and myxoid solitary fibrous tumour (Said-Al-Naief et al. 2001).

Spindle cells are usually small uniform cells arranged in parallel arrays. However, previous articles have reported some cases in which spindle cells have formed a palisading pattern or a “fibrosarcoma-like” arrangement, with pronounced nuclear

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pleomorphism and hyperchromatism (Piattelli et al. 2005). For this reason, spindle cell lipomas must also be distinguished from malignant tumours including well-differentiated liposarcoma, myxoid fibrosarcoma, and atypical lipomatous tumours (Darling et al. 2002).

The previously reported immunohistochemical profile of spindle cell lipoma has generally included positive staining for CD34 and vimentin and negative staining for S100 (Lombardi et al. 1994; Magro et al. 2001). In the present study, tumour cells exhibited immunoreactivity to CD34 and vimentin but not to S100. The exact pathogenesis and origin of spindle cell components are still unknown. Various spindle cell origins, including spindle-shaped mesenchymal cells with features of fibroblasts and differentiated adipocytes, immature mesenchymal cells, and CD34-positive dendritic interstitial cells, have been proposed (Al Sheddi et al. 2014).

The role of sex steroid hormones in the pathogenesis of oral spindle cell lipoma is unclear. According to previous reports, spindle cell lipoma in the oral region has a slight predominance in males, whereas in other locations, spindle cell lipoma more commonly affects females (Darling et al. 2002). Furthermore, Syed et al. (2008) reported that spindle cells exhibited strong immunoreactivity to androgen receptors. However, Vecchio et al. (2009) reported that spindle cells showed no immunoreactivity to any of the sex steroid hormones. In our case, spindle cell lipoma of the gingiva was diagnosed in an elderly male dog, and adipogenic spindle cells showed positive immunoreactivity to oestrogen receptors α and β . Although this is a single case, this finding may clarify the pathogenic role of oestrogen in the development of oral spindle cell lipoma in male dogs.

Intra-oral spindle cell lipoma is thought to be a benign entity, usually curable with local excision in humans (Darling et al. 2002; Piattelli et al. 2005). Preoperative diagnosis is difficult because of the heterogeneous imaging features produced by fat and spindle cells. Imaging modalities such as computed tomography and magnetic resonance imaging would be helpful in defining a confluence of fat and fibrous densities. For a definitive diagnosis, the specimen should be histopathologically examined only if an admixture of spindle cells and abnormal growth of adipocytes are present.

In conclusion, ours is the first case of spindle cell lipoma of the gingiva reported in a dog. Clinical,

histological, and immunohistochemical features of the tumour were compared to those in previous studies (Lombardi et al. 1994; Darling et al. 2002; Piattelli et al. 2005). Preoperative diagnosis of spindle cell lipoma of the gingiva is difficult, as it must be differentiated from other malignant soft tissue tumours. The diagnosis of spindle cell lipoma can be made by observing distinguishing histological features including spindle cells that stain positive for vimentin and CD34, admixed with mature adipocytes. Additionally, the possible oestrogenic effects on abnormal development of oral spindle cell lipoma are suggested. However, further studies are required to demonstrate the relationship between sex steroid hormones and the pathogenesis of oral spindle cell lipoma.

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Corresponding Author:

Professor Sun Hee Do., D.V.M., Ph.D., Konkuk University, College of Veterinary Medicine,
Department of Veterinary Clinical Pathology, Seoul 143-701, Republic of Korea
E-mail: shdo@konkuk.ac.kr
