Journal of Oral and Maxillofacial Surgery, Medicine, and Pathology 32 (2020) 49–52

Contents lists available at ScienceDirect

Journal of Oral and Maxillofacial Surgery, Medicine, and Pathology

journal homepage: www.elsevier.com/locate/jomsmp

Case Report

A case of spindle cell/pleomorphic lipoma on inferior border of the mandible

Kanako Munakata[a,b], Seiji Asoda[a,*], Shin Usuda[c], Hidetaka Miyashita[a], Tomoya Soma[a], Toshihiro Hasegawa[a,b], Chiharu Ogawa[b], Sadao Ohyama[b], Tomohiro Yamauchi[b], Miho Kawaida[d], Kaori Kameyama[d], Taneaki Nakagawa[a], Hiromasa Kawana[a]

[a] Division of Oral and Maxillofacial Surgery, Departmant of Dentistry and Oral Surgery, School of Medicine, Keio Universitiy, Tokyo, Japan
[b] Department of Dentistry & Oral surgery, Tokyo Metropolitan Cancer and Infectious Diseases Center Komagome Hospital, Japan
[c] Department of Dentistry & Oral surgery, Federation of National Public Service Personnel Mutual Aid Associations Tachikawa Hospital, Japan
[d] Department of Pathology, Keio University School of Medicine & Division of Diagnostic Pathology, Keio University Hospital, Japan

ARTICLE INFO

Keywords:
Spindle cell/pleomorphic lipoma
Spindle cell
Mandible

ABSTRACT

Spindle cell lipomas and pleomorphic lipomas are uncommon and distinct variants of benign lipomas. Although spindle cell lipomas and pleomorphic lipomas were originally considered different entities, the recent WHO classification defines them as spindle cell/pleomorphic lipoma (SCL/PL). This is because of their similar clinical features and the existence of intermediate histopathological features between them. A 77-year-old male presented to our clinic with a painless swelling on the left inferior border of the mandible. Computed tomography revealed a clearly defined mass in the subcutaneous lesion. With a histopathological diagnosis of myxolipoma, demonstrated by biopsy, excisional surgery was performed under general anesthesia. The histopathology revealed SCL/PL with the presence of spindle cells and mature adipocytes. No tumor recurrence had been noted at two year after surgery.

1. Introduction

Spindle cell/pleomorphic lipoma (SCL/PL) is a rare and histological variant of lipoma. It typically occurs in the posterior neck, back, and shoulders of elderly males. Histopathologically, SCL/PL shows a mixture of mature adipocytes and spindle cells, which are features of spindle cell lipomas (SCL), and it also shows floret-like giant cells, a feature of pleomorphic lipomas (PL). Although SCL and PL had originally been considered different entities, this intermediate mature of morphological features between them has resulted in the term SCL/PL [1]. To the best of our knowledge, only 3 reports have described SCL/PL in the maxillofacial region, whereas there have been over 50 case reports of SCL in the oral cavity [2]. Herein, we report a rare case of SCL/PL involving the maxillofacial region.

2. Case report

A 77-year-old man was admitted to our clinic for the investigation of an asymptomatic mass on his left inferior border of the mandible that had persisted for 10 years without any investigation. The mass was painless and mobile on palpation (Fig. 1). Computed tomography (CT) revealed a 30-mm in diameter, well-circumscribed mass. The mass was present over the platysma. It showed a high density compared with the surrounding fat tissue (Fig. 2). Magnetic resonance imaging (MRI) showed a heterogeneous low-signal intensity on T1-weighted images and heterogeneous intermediate to high-signal intensity on T2-weighted images (Fig. 3A, B). Contrast-enhanced MRI indicated heterogeneous enhancement that corresponded to a low-signal intensity on T1-weighted images (Fig. 3C). We could not identify the disease from the study of images, which led to a tissue biopsy. The mass underwent incisional biopsy, and the pathological diagnosis was myxolipoma. Based on the pathological diagnosis, surgical excision was planned.

The mass was easily removed from the surrounding tissue with an approach directly over the biopsy scar. The specimen was 15 × 30 mm, brownish-yellow, and well-encapsulated (Fig. 4A). The cut surface of the specimen was smooth, which is the feature of myxoid area (Fig. 4B). On histopathological examination, with a low-magnified view, the specimen was composed of two areas: adipose area and low adipose

* Corresponding author at: Division of Oral and Maxillofacial Surgery, Departmant of Dentistry and Oral Surgery, School of medicine, Keio University, 35 Shinanomachi, Shinjyuku-ku, Tokyo, 160-0016, Japan.
E-mail address: asoda@keio.jp (S. Asoda).

https://doi.org/10.1016/j.ajoms.2019.09.005
Received 15 March 2018; Received in revised form 18 July 2019; Accepted 11 September 2019
Available online 03 October 2019

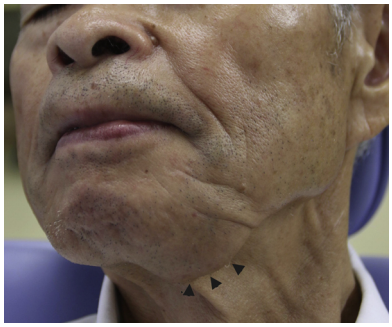

Fig. 1. Facial observation. Arrowhead: the position of the tumor.

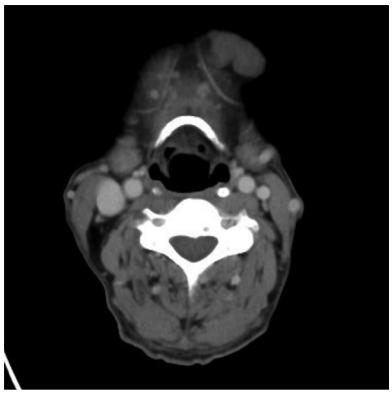

Fig. 2. Axial computed tomography image.

area (Fig. 5). With a high-magnified view, the specimen consisted of mature adipocytes and fibrous connective tissue along with myxoid area. Also, proliferation of spindle-shaped cells and small thick-rope-like collagen bundles was observed (Fig. 6A). Immunohistology for CD34 was positive in spindle cells (Fig. 6B). The definitive diagnosis was SCL/PL based on pathological evaluation.

3. Discussion

SCL and PL were first described by Enzinger et al. as specific types of lipoma. Although SCL and PL had been considered different entities, they are now thought to exist within the same spectrum. Both lipomas share the same clinical, morphological, immunohistochemical, and cytogenetic features. Thus, the recent WHO classification defines them as spindle cell/pleomorphic lipoma (SCL/PL). SCL/PL accounts for 1.5% of all adipocytic neoplasms [3]. It is common in elderly males, with a median age of 55 years old. It predominantly occurs in the posterior neck, back, and shoulders mostly with asymptomatic long-standing subcutaneous masses [4]. On searching the database Pubmed (National Center for Biotechnology Information, USA) using the keyword “spindle cell/pleomorphic lipoma”, we identified 5 case reports and 2 reviews [1–9]. The number of cases involving the head and neck area was 46, by adding case reports and reviews, and also including our case. No cases involving the inferior border of the mandible were found. The patients comprised 33 males and 13 females, with ages ranging from 20 to 91 years (mean age: 58.8). The most common sites in the oral and maxillofacial region were the nose (11 cases), cheek (9 cases), and forehead (6 cases). Only one patient showed recurrence among all cases.

Macroscopically, SCL/PL is a yellowish to greyish-white mass

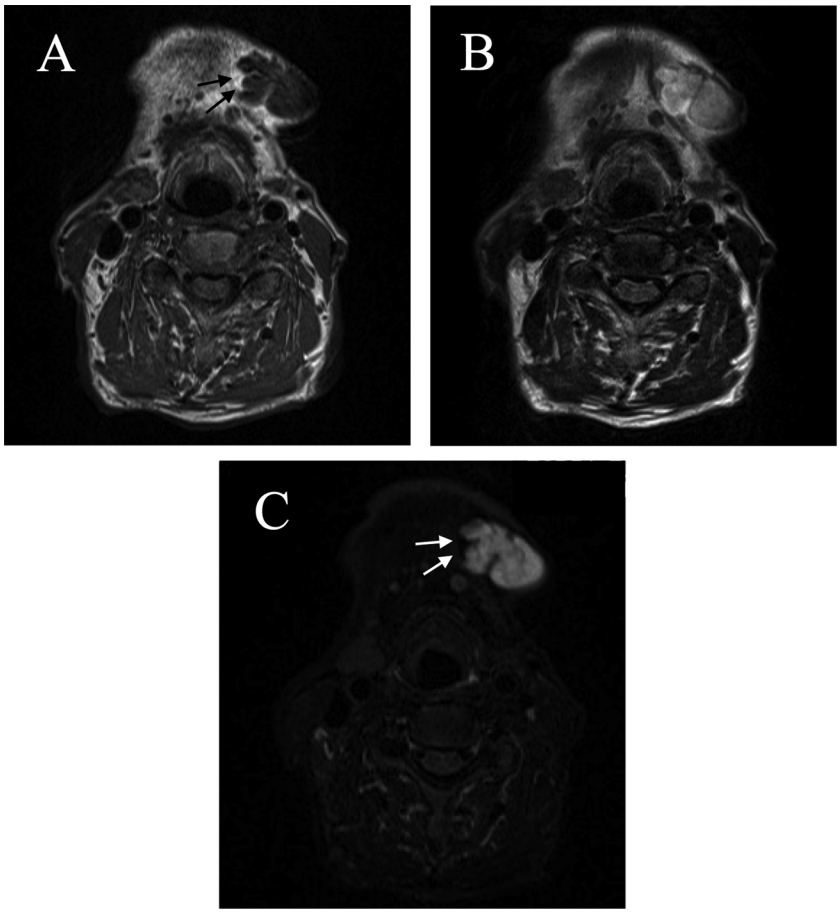

Fig. 3. Axial magnetic resonance imaging. (A) T1-weighted images show a heterogeneous low-signal intensity occupying a large space. The arrow shows a small area of high-signal intensity. (B) T2-weighted images show a heterogeneous intermediate to high-signal intensity. (C) Contrast-enhanced MRI shows that heterogeneous enhancement corresponded to low-signal intensity on T1-weighted images. The arrow shows a small area of non-enhancing lesions.

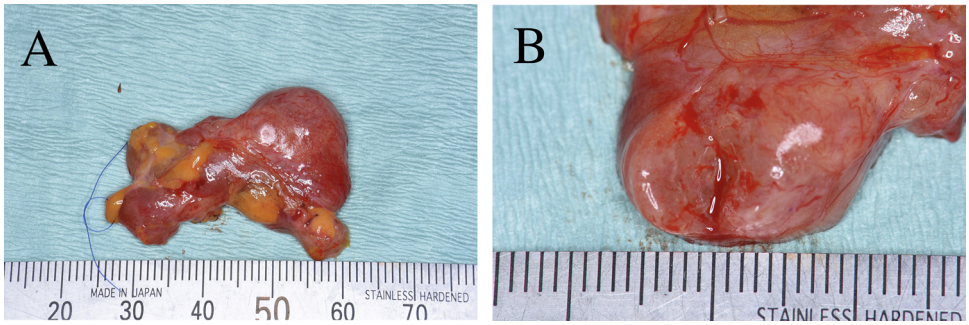

Fig. 4. Resected specimen. (A) Resected specimen. (B) Cut surface of specimen.

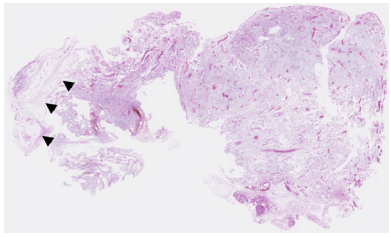

Fig. 5. Low-powered magnified view. The arrowhead shows the adipose area.

depending on the extent of fatty and spindle cell components. It is firmer than typical lipoma [4]. It is histopathologically characterized by an admixture of spindle cells and mature fat cells, with thick ropy collagen and a myxoid stroma in the matrix. Immunohistochemically, spindle cells are typically positive for CD34. In rare cases, some are positive for s-100 protein and desmin. In our case, spindle cells and mature adipocytes were observed, and CD34 was positive in spindle cells.

It is important to exclude the possibility of an atypical lipomatous tumor/ well-differentiated liposarcoma (ALT/WD liposarcoma) in the diagnosis of SCL/PL. ALT/WD liposarcoma grows asymptomatically in subcutaneous lesions, with a similar morphological pattern to mature adipocytic proliferation. The spindle cell variant of ALT/WD liposarcoma, one of the subtypes of ALT/WD liposarcoma, is composed of a fairly bland neural-like spindle cell proliferation with a fibrous and/or myxoid background. The predilection sites of ALT/WD liposarcoma are deep soft tissues of the limbs and retroperitoneum, being rare in oral and maxillofacial areas. Although ALT/WD liposarcoma exhibits mature adipocytic proliferation, it shows significant variation in cell size with hyperchromatic stromal cells and lipoblasts, which can aid in differentiation. Also, MDM2 and CDK4 immunostaining are said to be useful to separate ALT/WD liposarcoma from other soft tissue tumor [10]. Furthermore, there are several reports recently telling the combination with p16 immunostaing is more sensitive to ALT/WD liposarcoma, which could be helpful in distinguishing ALT/WD liposarcoma [11,12].

Imaging findings of SCL/PL have been reported to be variable because of the ratio of adipose and non-adipose components. Lee et al. reported images similar to those we obtained using T1WI and T2WI: heterogeneously hypointense signal on T1, heterogeneously hyperintense on T2 [3]. Also, their contrast-enhanced MRI showed a heterogeneously enhanced mass, which was consistent with our case. In our case, contrast enhancement was observed, which was similar to the low-signal region of T1WI, and this region occupied a large proportion of the tumor (Fig. 3A, C). Such MRI findings were consistent with the low-magnified histological view. The contrast-enhanced area was a low-adipose area, that is, a spindle cell-composed area, and the remaining area was an adipose area (Figs. 3C and 5).

In summary, we herein report a rare case of SCL/PL of the mandible. As the characteristics vary among cases, it may be difficult to make a diagnosis solely based on imaging findings such as from CT and MRI, or even with biopsy before excision. Furthermore, with the important differential diagnosis of ALT/WD liposarcoma, the diagnosis should be determined immunohistopathologically. SCL/PL rarely shows recurrence, and a two years have elapsed since excision with no recurrence in our case.

Declaration of Competing Interest

None of the authors have any conflicts of interest to declare.

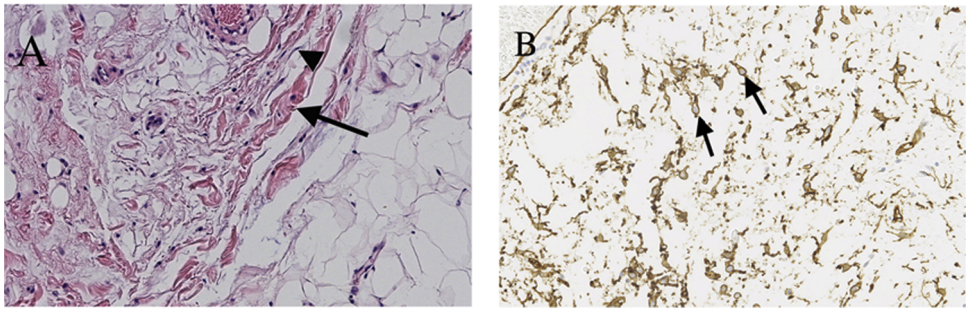

Fig. 6. High-powered magnified view. (A) Hematoxylin-eosin stain. The arrowhead shows spindle cells. The arrow shows a collagen bundle. (B) CD34 immunohistochemical stain. Arrows indicate the CD34-stained cells.

References

[1] Enzinger FM, Weiss SW. Benign lipomatous tumor. In: Enzinger FM, Weiss SW, editors. Soft tissue tumors. 5th ed.St Louis: Mosby; 2008. p. 431–52.

[2] Giada V, Paolo A, Rosario C, Giuseppe C, Salvatore L, Gaetano M. Spindle Cell/Pleomorphic lipoma of the oral cavity. J Craniofac Surg 2009;20:1992–4.

[3] Hyun KL, Seung BH, Gyung HC, Hwan H, Kyu YJ. Retropharyngeal spindle cell/pleomorphic lipoma. Korean J Radiol 2013;14(3):493–6.

[4] Miettinen MM, Mandahl N. Spindle cell/pleomorphic lipoma. In: Flethcer CD, Unni K, Mertens F, editors. WHO classification of tumors of soft tissue and bone. IARC Press; 2013. p. 29–30.

[5] Chritopher AF, Thomas M, Heinz K, Christopher DMF. Intradermal spindle Cell/Pleomorphic lipoma a distinct subset. Am J Dermatopathol 2000;22(6):496–502.

[6] Jorge SRF, Fernanda M, Marioa FS, Jose FN, Fernando CS. Intradermal spindle call/pleomorphic lipoma of the vulva: case repot and review of the literature. J Cutan Pathol 2001;29:59–62.

[7] Duygu G, Aydanur K, Banu L. Pedunculated cutaneous spindle cell/pleomorphic lipoma. J Cutan Pathol 2010;37:57–9.

[8] Mark L, Michael R. Uncommon Tumor, Uncommon Location: A Dermal-Based Spindle Cell/Pleomorphic Lipoma. Am J Dermatopathol 2016;38:122–4.

[9] Alison C, Steven B, John G, Jason H, Nasir U, Brian R. Spindle cell/pleomorphic lipomas of the face: an under-recongnized diagnosis. Histopathology 2015;66(3):430–7.

[10] Binh MB, Sastre-Garau X, Guillou L, et al. MDM2 and CDK4 immunostainings are useful adjuncts in diagnosing well-differ- entiated and dedifferentiated liposarcoma subtypes: a comparative analysis of 559 soft tissue neoplasms with genetic data. Am J Surg Pathol 2005;29:1340–7.

[11] He M, Aisner S, Benevenia J, et al. p16 immu-nohistochemistry as an alternative marker to distinguish atypical lipomatous tumor from deep-seated lipoma. Appl lrnmunohistochem Mol Morphol. 2009;17:51–6.

[12] Thway K, Flora R, Shah C, et al. Diagnostic utility of p16, CDK4, and MDM2 as an immunohistochemical panel in distinguishing well-differentiated and dedifferentiated liposarcomas from other adipocytic tumors. Am J Surg Pathol 2012;36:462–9.