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Case report

Ancient schwannoma of the tongue: A case-report and literature review

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Abstract. Schwannomas are benign tumors arising from Schwann cells of the nerve. Although they commonly appear in the soft tissues of the head and neck (25%), the intraoral origin (1%) and the ancient histological subtypes represent the rarest variant. Specific diagnosis is difficult and often delayed. Radiological examination can be helpful, but complete excision remains the treatment of choice to obtain histological information and reduce the risk of recurrence. We report a rare case of intramural schwannoma of the tongue in a 80-year-old male patient, excised completely via a transoral approach. Histophatological examination and immunohistochemistry confirmed diagnosis and the patient had no postoperative complications. We performed a literature review with a PubMed search about schwannomas of the mobile tongue about the last 30 years. We found 585 articles, of which 54 met the inclusion criteria with a total of 90 cases of schwannoma of the tongue. We finally described the most relevant features of these tumors and their management. (www.actabiomedica.it)

Key words: tongue schwannoma, intramural schwannoma, tongue surgery, schwann cells, s-100

Introduction

Schwannomas are benign tumors arising from Schwann cells of the nerve. Also known as neurilemmomas or neurinomas, they commonly appear in the soft tissues of the head and neck (25%-45%). However, only 1% of these lesions shows an intraoral origin (1–3) where they may involve in order of frequency: tongue, buccal mucosa, intramedullary bone of maxilla or mandible, floor of mouth, palate, gingiva, lips, and vestibular mucosa (4).

Six histologic subtypes schwannoma have been described: classic, cellular, epithelioid, plexiform, reticular/microcystic, and ancient (representing the rarest) (5). Although rare in the pediatric population, these tumors can occur at all ages, and males and females are equally affected. Schwannomas usually appear as a solitary, slow-growing, circumscribed and encapsulated mass; however, they can be multiple when associated with neurofibromatosis (NF) (2,6). Radiologic examination by contrast-enhanced Computed Tomography (CT) and Magnetic Resonance Imaging (MRI) can be helpful in assessing the pattern and size of the lesions.

The rarity of the site of origin makes specific diagnosis difficult and often delayed; therefore, complete excision remains the treatment of choice to obtain histological information and reduce the risk of recurrence (7).

In this paper, we describe the management of a rare case of ancient intra-lingual schwannoma and reviewing the current English Literature.

Case report

An 80-year-old man presented to the emergency department of our hospital with a painless mass reported for 20 years on the ventral surface of the tongue

that had increased significantly in size in recent days. He complained of difficulty swallowing and speech impediment due to the size and the resulting reduction in anterior tongue movements. Gustatory sensation was not impaired. The patient's medical history included type II diabetes mellitus, chronic inflammatory demyelinating polyneuropathy (CIPD), monoclonal gammopathy of undetermined significance (MGUS), hypertension, gastroesophageal reflux disease (GERD) and hepatic steatosis. Surgical removal of multiple basal cell carcinomas on the face has also been reported.

Oral examination revealed a large solitary, oval mass in the anterior third of the ventral tongue with a tense-elastic consistency (Figure 1). The lesion was completely submucosal; the mucosal surface appeared healthy with no signs of injury or hemorrhage. An exploratory puncture was performed, but no fluid was drained.

Contrast-enhanced CT showed an oval, well-defined, capsulated mass measuring 3.7 cm cranio-caudal (CC), 4.0 cm antero-posterior (AP) and 3.0 cm medial-lateral (ML). The mass was hypodense with inhomogeneous superfluid density; some vessels were noted inside.

On gadolinium-enhanced MRI, the lesion was hypointense in T1-weighted images and showed heterogeneous hyperintensity in T2-weighted images (Figure 2). Some areas of restriction were observed in the diffusion-weighted images (DWI). The lesion involved the entire thickness of the body of the anterior third of the tongue. The enhancement was homogeneous and diffuse, except for some central areas that show no contrast. After discussion in our multidisciplinary board and review of the radiological images, transoral excision of the lesion through a ventral incision of the tongue was planned.

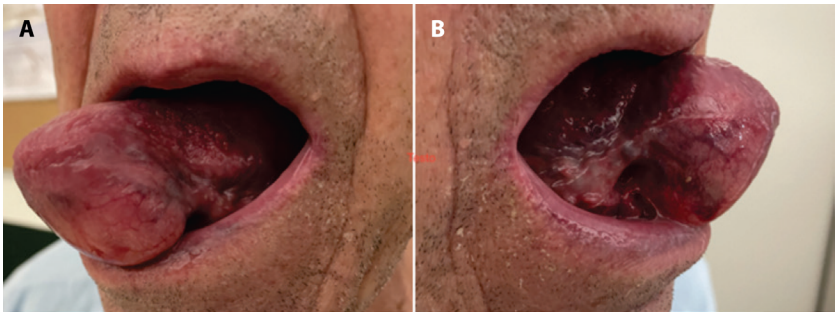

Figure 1. Preoperative appearance of the ventral tongue submucosal mass observed from left (a) and right side (b).

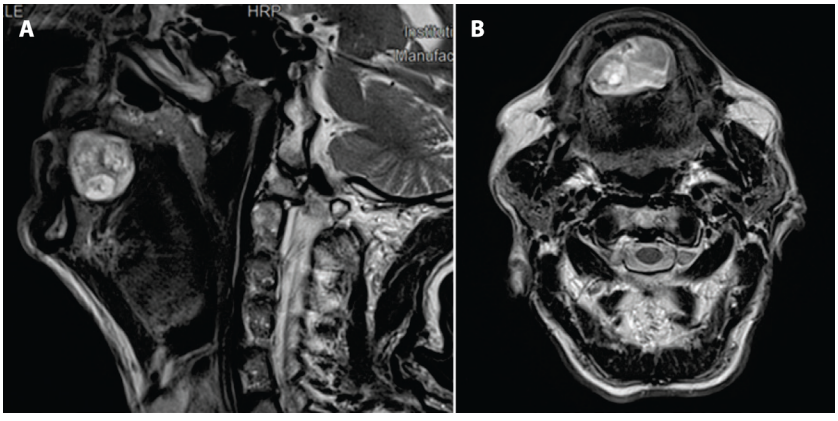

Figure 2. MRI T2-weighted images show a heterogeneous hyperintense capsulated mass of the anterior third of the tongue in both sagittal (a) and axial (b) plane.

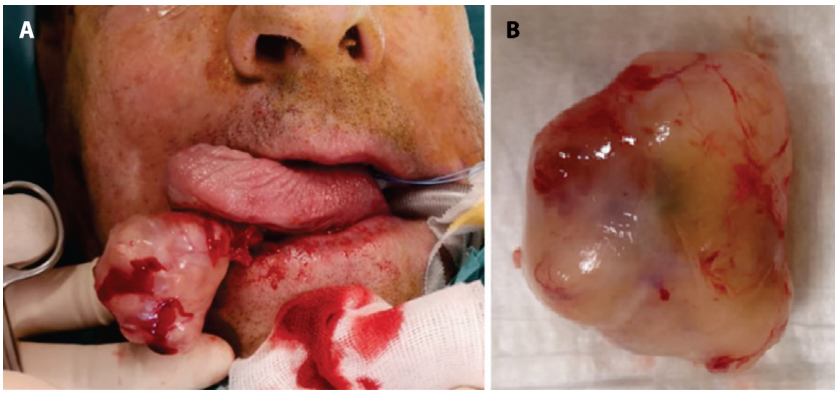

Figure 3. Intraoperative appearance of the tumor at the end of dissection (a); a close-up view of the specimen is shown on the right panel (b).

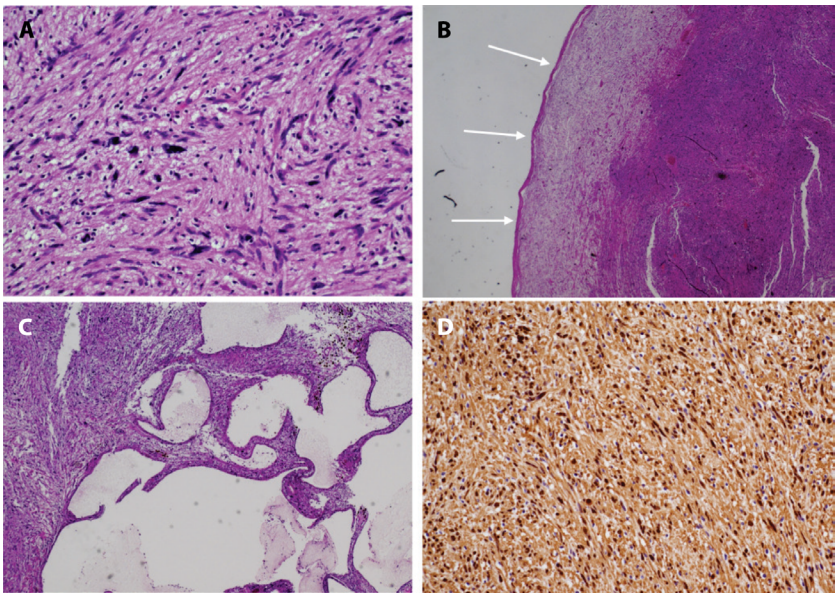

Figure 4. Histological characteristics of the tumor. Considerable nuclear pleomorphism (a) is apparent as well as an area of diffuse edema (white arrows) (b) and pseudocysts (c). Strong positivity at S-100 immunostaining is also shown (d).

The lesion appeared intraoperatively as a yellowish encapsulated mass. It was excised "en-bloc" from within the musculature (Figure 3).

The final histopathologic report revealed an "ancient schwannoma" characterized by the presence of pseudocyst, nuclear pleomorphism, area of diffuse edema, involutional changes, and reactive cytologic changes. Finally, at immunohistochemistry the sample showed strong positivity for S-100 protein, which is the most widely used marker for peripheral nerve sheath tumors (Figure 4).

The patient had no postoperative complications. Mild hypoesthesia at the tip of the tongue was reported in the first month. Speech, taste, and tongue movements were preserved at the 1-year follow-up visit.

Literature review

We performed a PubMed search of the terms "tongue AND schwannoma," "lingual AND schwannoma," "tongue AND neurilemmoma," and "lingual AND neurilemmoma" with the date range of 1990-2022. The search was limited to English language case reports. We included cases with a confirmed diagnosis of schwannoma and described the characteristics of tumors involving the mobile tongue, including its ventral portion. Lesion involving the floor of the mouth and the base of the tongue were excluded. We found 585 articles, of which 54 met the inclusion criteria with a total of 90 cases of schwannoma of the tongue reported (Figure 5).

The following items were extracted from the case reports for data analysis: gender, age, maximum size, tumor site, signs, symptoms, presence or absence of nerve deficit, radiological examinations, treatment, and follow-up (Table 1).

Fifty-three patients were males (58.9%) and 37 females (41.1%), with a male-to-female ratio of almost 3:2., unlike previously reported data (2,6). This finding may be explained by the variability of anatomical sub-sites involved in other reviews, especially in those that included the base of tongue (8,25,51,64).

The median age at diagnosis was 25 years (range 7-82), in agreement with other recent reviews (8). Six out of 90 cases (6.7%) had mucosal ulceration,

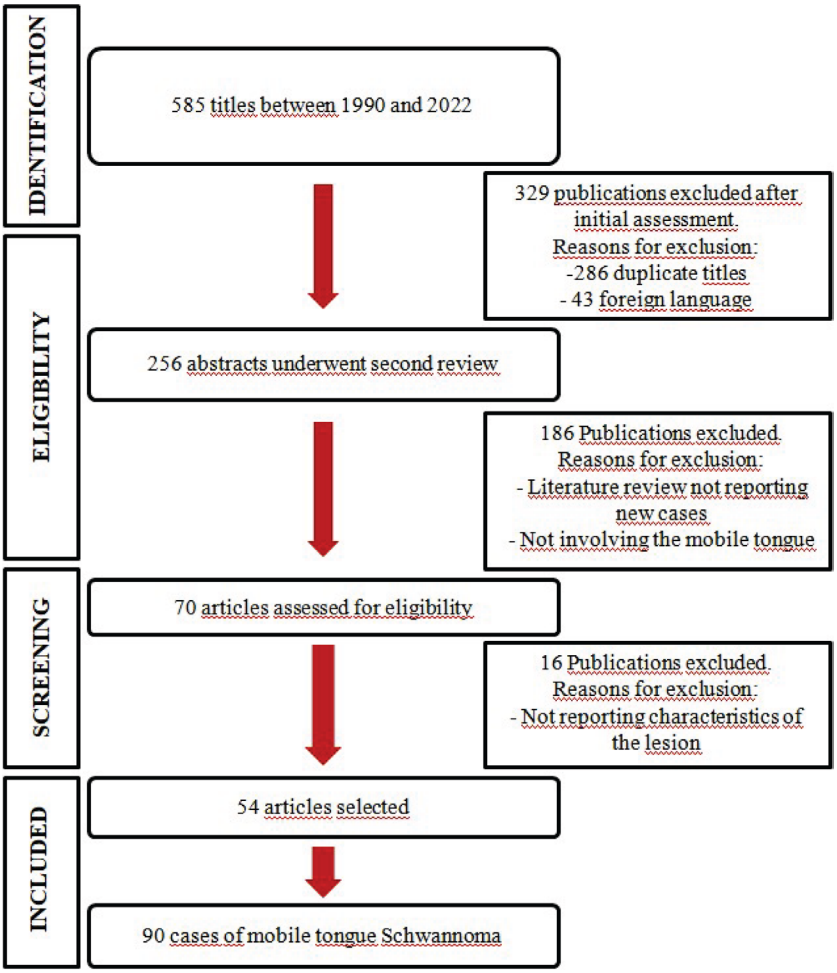

Figure 5. This flowchart illustrates the process we used to select articles for the review.

Table 1. Demographic and clinical characteristics of 90 patients with ancient schwannoma described in the English literature during 1990-2022.

Author	Year	N. of cases	Sex	Age	Dimension	Site	Nerve deficit	Approach	Follow-up
Fechner et al.[9]	1991	1	F	75	1,5	Posterolateral border	No	Transoral	N.S.
Gallesio et al.[10]	1992	1	F	21	1,9	Tip	Yes	Transoral	1 yr
Williams et al.[11]	1993	2	M	28	0,5	Dorsal	N.S.	N.S.	N.S.
			M	58	1	Lateral border	N.S.	N.S.	N.S.
Nakayama et al.[12]	1996	1	F	40	5,5	Ventral	No	Transoral	2 yr
Pfeifle et al.[13]	2001	2	F	30	1	Tip	No	Transoral	N.S.
			M	18	N.S.	Ventral	No	Transoral	N.S.
Go et al.[14]	2002	2	F	39	0,8	Lateral border	No	Transoral	N.S.
			M	30	3	Dorsal	No	Transoral	6 mo
Pahwa et al.[15]	2003	2	M	65	1,5	Lateral border	No	Transoral	N.S.
			M	35	1,5	Tip and dorsal	No	Transoral	N.S.
Cinar et al.[16]	2004	1	M	7	1	Tip	No	Transoral	N.S.
Hwang et al.[17]	2005	1	M	23	2,8	Tip	Yes	Transoral	6 mo
Vafiadis et al.[18]	2005	1	M	18	3,1	Tip	No	Transoral	N.S.
Hsu et al.[19]	2006	7	3 F, 4 M	9-45	1,34	5 Lateral border, 2 Tip	N.S.	Transoral	1 yr-16 yr
Enoz et al.[20]	2006	1	M	7	2,5	Tip	No	Transoral	5 yr
Jethanamest et al.[21]	2006	1	F	19	5,4	Ventrolateral tongue	No	Transoral	N.S.
Patnayak et al.[22]	2007	1	F	45	2	Posterolateral border	No	Transoral	N.S.
Pereira et al.[23]	2008	1	M	12	1,5	Posterior third	No	Transoral	1 yr
Bonan et al.[24]	2008	1	F	46	N.S.	Tip	No	Transoral	1 yr
Cohen et al.[25]	2009	2	F	19	1.8	Lateral border	No	Transoral	N.S.
			M	77	0,7	Lateral border	No	Transoral	N.S.
Gupta et al.[26]	2009	1	F	18	1	Dorsal	No	N.S.	N.S.
Jeffcoat et al.[27]	2010	1	M	68	1,5	Lateral border	No	Transoral	No
Naidu et al[28]	2010	1	M	12	2	Ventral	No	Transoral	3mo
Karaca et al.[3]	2010	1	M	13	2	Anterior third	No	Transoral	1 yr
Catalfamo et al.[29]	2011	1	M	28	0,8	Dorsal	N.S.	Transoral	N.S.
Lukšić et al.[30]	2011	1	M	10	1,8	Lateral border	No	Transoral	5 yr
Ferrari et al.[31]	2011	1	M	24	4	Ventral tongue	No	Transoral	N.S.
Husain et al.[32]	2011	1	F	10	5	Lateral border	No	Transoral	1 yr
Al-mahdi et al.[33]	2012	1	M	27	3	Lateral margin extending in the submandibular space	N.S.	Transoral and submandibular	N.S.
Manna et al.[34]	2012	1	M	15	1,7	Posterior third	No	Transoral	6 mo
Lira et al.[35]	2013	1	F	26	2,5	Posterior third	Yes	Transoral	No

Table 1 (*Continued*)

Author	Year	N. of cases	Sex	Age	Dimension	Site	Nerve deficit	Approach	Follow-up
Ferreira et al.[36]	2013	1	F	18	0,5	Tip	Yes	Transoral	1 yr
Moreno-García et al.[7]	2014	1	F	13	2	Ventral		Transoral	1 yr
Bhola et al.[37]	2014	1	F	14	1,5	Anterolateral border	No	Transoral	1 yr
Feltes-Ochoa et al.[38]	2015	1	M	52	0,2	Dorsal	No	Transoral	6 mo
Kavcic et al.[39]	2016	1	F	20	1.5	Tip	No	Transoral	1mo
Badar et al.[40]	2016	1	F	24	N.S.	Posterior third	No	Transoral	N.S.
Sugawara et al.[41]	2016	1	M	16	1,5	Dorsal	No	Transoral	N.S.
Medhi et al.[42]	2016	1	M	22	5	Posterior third	No	Right paramedian mandibulotomy	N.S.
Qayoom et al.[43]	2016	1	M	25	0,9	Anterior third	No	Transoral	1mo
Lee et al.[44]	2017	1	F	71	3	Ventral	Yes	Transoral	1 yr
Abreu et al.[45]	2017	1	M	20	1,9	Lateral border	No	Transoral	N.S.
Amer et al.[46]	2018	1	F	13	1,7	Tip	No	Transoral	N.S.
Diplan et al.[47]	2018	1	F	40	6	Dorsal and ventral	No	Transoral	N.S.
Sharma et al.[48]	2019	1	M	25	2.5	N.S.	No	N.S.	N.S.
Shashikumar et al.[49]	2019	1	F	34	5	Dorsal	No	Transoral	2mo
Thompson et al.[50]	2020	19	6 F, 13 M	12-82	1,07	13 Anterior two-thirds 6 Posterior third	No	N.S.	N.E.D
Barca et al. [51]	2020	1	M	40	1,8	Ventral	No	Transoral	2 yr
Kumar et al.[52]	2020	1	M	28	3	Anterior third	No	N.S.	N.S.
Chen et al.[53]	2020	1	M	53	4	Ventral	No	Transoral	15mo
Ahmed et al.[54]	2020	1	F	14	3	Dorsal	No	Transoral	2 yr
Ohta et al.[55]	2021	1	M	17	1.4	Lateral border	No	Transoral	10 mo
Yun et al.[56]	2021	1	M	8	1.7	Anterolateral border	No	Transoral	1 yr
Chi et al.[57]	2021	5	4 F, 1 M	25-62	N.S.	2 Lateral border, 2 Dorsal	N.S.	Transoral	N.S.
Agha Hosseini et al.[58]	2021	1	M	15	1	Lateral border	No	Transoral	1 yr
Alrohaimi et al.[59]	2021	1	M	12	2,3	Ventral	No	Transoral	3 yr
Phulware et al.[60]	2022	4	2 F, 2 M	15-39	1,5	Anterior two-thirds	N.S.	Transoral	N.E.D.

while the rest of the cases were described with normal mucosa (81%). The median size was 1.85 cm (range 0.2 cm – 6.0 cm). The description of the tongue subsites involved was not univocal and with exception of 2 cases where the subsite was not stated, all the remainder were characterized as: lateral border (n=22, 25%); anterior two-thirds (n=20, 22.7%); tip (n=13, 14.8%); dorsal (n=11, 12.5%); posterior third (n=11, 12.5%);

ventral (n=10, 11.4%); lateral border extending to submandibular space (n=1, 1.1%).

According to our analysis, half of the patients were asymptomatic (50 percent), while those who were symptomatic (50 percent) complained of: pain (n=13, 28.9%), dysphagia (n=10, 22.2%), difficulty in articulating words (n=9, 20%), paresthesia (n=4, 8.9 %), difficulty in chewing (n=4, 8,9%), bleeding (n=2, 4.4%), tongue distortion (n=1, 2.2%), ageusia (n=1, 2.2%), pressure sensation (n=1, 2.2%). In general, the mucosa appeared normal; bleeding and erosion were rare, as described by Naidu et al. (28) and Abreu et al. (45).

All patients presented with a single lesion, except for one patient who presented with multiple lesions, although he did not have NF2 (53).

Fine-needle aspiration cytology (FNAC) was performed in 9.3% of cases, but only 12.8% of these cases were diagnostic for a schwannoma.

Discussion

Schwannoma is a benign tumor arising from Schwann cells of the nerve sheath surrounding any cranial, peripheral, or autonomic nerve. Although the etiology is still unknown, it appears to originate from the proliferation of Schwann cells within the perineurium, which leads to displacement and compression of the surrounding nerve. (19,61) Schwannomas are usually large and long-lasting tumors, are largely located in deep structures such as the retroperitoneum, and rarely occur in the oral and maxillofacial region.(62,63). The head and neck district is involved in 25-45%; within the extracranial localizations intraoral presentations constitute only 1%. (2,3)

Definitive diagnosis is obtained by histopathologic examination. Based on histologic features and growth pattern, several types of Schwannomas are identified: classic schwannoma, ancient schwannoma, cellular schwannoma, epithelioid schwannoma, plexiform schwannoma and reticular/microcystic schwannoma. (5)

Classical schwannoma is the most common; it is histologically characterized by alternating hypercellular and hypocellular areas (7,62,65). In contrast, ancient schwannoma is the rarest (66) and is characterized by nuclear atypia of “degenerative type” and other degenerative changes, including cystic stromal changes, hemorrhage, calcifications, and diffuse stromal hyalinization (5). The peculiarity of the lesion with these degenerative changes is that it can be misdiagnosed as sarcoma or other forms of soft tissue neoplasms (62,63).

Immunohistostaining commonly reveals positivity for S-100, Leu-7 antigen, vimentin, and glial fibrillary acidic protein supporting the Schwann cell origin of these tumors (67). Although usually benign, cases of malignant Schwannoma of the head and neck region have been reported, including one case of malignant schwannoma of the tongue (68).

Regarding radiological investigations reported, 17.4% of the patients underwent MRI, and in all cases the tumor appeared isointense in T1WI and heterogeneously hyperintense in T2WI. In contrast, 7.0% of the patients underwent CT scan which revealed a heterogeneously hypodense mass. The characteristics of Schwannoma on CT are as follows: low to intermediate attenuation, intense contrast enhancement (small tumors typically show homogeneous enhancement, whereas larger tumors may show heterogeneous enhancement) and remodeling of adjacent bone with smooth, corticated edges (69,70).

Radiological heterogeneity is attributed to regions of compactly arranged cells (Antoni A type) intermixed with regions of loosely arranged cells (Antoni B type), with variable cellularity and water content (71). In addition, heterogeneous enhancement, cystic spaces, and foci of hemosiderin due to internal hemorrhage, may be present in larger lesions (70).

Surgery is the treatment of choice, as confirmed by our review. All patients underwent complete surgical excision, and the surgical approach depends on the site and extent of the lesion. Specifically: 88 out of 90 cases (97.8%) underwent transoral resection; 1 case (1.1%) was approached with a paramedian mandibulotomy (72) and 1 case (1.1%) received a combined transoral and submandibular approach.

The clinical case described by us and those reported in the literature show that surgical treatment is radical and curative in most of cases, especially if the lesion is removed completely without residue. No long-term nerve deficit was observed in the cases analyzed, and no recurrence of the disease was found at follow-up.

Conclusion

Although rare, Schwannoma should be considered in the differential diagnosis of a smooth, slow-growing mass of the tongue. It is a benign lesion, but in rare cases it can have malignant degeneration. The ancient subtype is the rarest and, because of degenerative changes, may be misdiagnosed as sarcoma or other forms of soft tissue neoplasms. Radiologic imaging, particularly MRI, can be helpful in characterizing the lesion and planning treatment.

Radical surgical excision is the gold standard for obtaining the diagnosis and performing comprehensive treatment to prevent recurrence of the disease.

Declaration of Consent: Informed consent was acquired from the patient and that the patient consented to the publication of all images, clinical data, and other data included in the manuscript.

Ethic Committee: This analysis did not need ethics committee approval as it did not constitute biomedical research.

Conflict of Interest: The authors declare that there is no conflict of interests regarding the publication of this paper.

Authors Contribution: DC, MSL and DS have designed and developed the article. Author AS has conducted the literature review and written the case report. Author MSL has supervised and lead the review and revised the case report. Author CC has performed the histopathological examination, created the histological images and contributed to the article with her knowledge and explanations on the rarity of this tumor. All authors (AS, MSL, DS, CC, DC) read and approved the final manuscript.

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