

Comprehensive Review of Reported Nonclassical Spindle Cell Lipoma Presentations and a Unique Case Report

Abigail J. Engwall-Gill, MD*
Mulin Xiong, MD†
Stephanie M. C. Bray, MD, MS*

Introduction: Spindle cell lipoma (SCL) is a rare subset of benign lipomatous neoplasms, making up approximately 1.5% of all adipocytic neoplasms. Since SCLs were first described in 1975, numerous nonclassical cases have been reported in the literature, with variation in location, cytologic composition, patient demographic, and progression. Although some previous reports review related cases of specific rare presentations or institution-based summaries, no comprehensive summary of published nonclassical SCL case reports has been done.

Methods: PubMed was queried for nonclassical presentations of SCL from 1978 to 2018. The nonclassical characteristics were analyzed and described. Those with multiple nonclassical features were studied for common characteristics. The Fisher exact test was used, and a *P* value of 0.05 was determined to be statistically significant.

Results: We present the first case of a patient with six nonclassical findings: a fast-growing, infiltrating into skeletal muscle, located in two positions on the chin of an 18-year-old African American woman. In this review of the existing nonclassical SCL, we evaluate 125 cases for demographics, location, size, erosion/infiltration, multiple lesions in one individual, and the number of nonclassical findings in one individual. Women and younger than 40-year-old patients significantly present with more than one nonclassical finding.

Conclusions: Thorough characterization of the rare nonclassical cases of this benign condition could guide diagnostic decision-making and identify trends in disease presentation over time as well as alert the clinician to the increased risk of rapid regrowth or invasion in an individual with nonclassical findings especially young age and female gender. (*Plast Reconstr Surg Glob Open* 2022;10:e4462; doi: 10.1097/GOX.0000000000004462; Published online 18 August 2022.)

INTRODUCTION

Lipomas are the most common soft tissue neoplasms. Of this large group, spindle cell lipoma (SCL) is an uncommon subdivision of benign fatty tumors, making up only 1.5% of all adipose tumors.^{1–3} Pathology diagnosis requires uniform small spindle cells and mature adipocytes. These slow-growing benign soft tissue tumors

typically present in the subcutaneous layer of the back, shoulder, or neck of White men between the ages of 40–70.^{2,4} Pathological differentiation from liposarcoma, malignant lesions, myxoid tumors, and other spindle cells is important. When pathology determines an SCL, prognosis is consistently good; thus, definitive treatment is considered to be wide local excision.^{2,3} Lesions present classically and are characterized as solitary, painless, 1–13 cm, benign, and slow-growing nodules in the subcutaneous or dermal tissue. Lipomas have been associated with various syndromes, such as Gardner syndrome, hereditary multiple lipomatosis, Dercum's disease, and Madelung's disease, as well as associated with trauma.

From the *Department of Surgery, Michigan State University, Lansing, Mich.; and †Michigan State University, Lansing, Mich. Received for publication February 4, 2022; accepted May 24, 2022. Presented at Mid Michigan Research Day (GMEI), Lansing, MI, in April of 2019.

Copyright © 2022 The Authors. Published by Wolters Kluwer Health, Inc. on behalf of The American Society of Plastic Surgeons. This is an open-access article distributed under the terms of the [Creative Commons Attribution-Non Commercial-No Derivatives License 4.0 \(CCBY-NC-ND\)](#), where it is permissible to download and share the work provided it is properly cited. The work cannot be changed in any way or used commercially without permission from the journal. DOI: 10.1097/GOX.0000000000004462

Disclosure: The authors have no financial interest to declare in relation to the content of this article.

Related Digital Media are available in the full-text version of the article on www.PRSGlobalOpen.com.

Possible differential diagnoses are ordinary lipoma, atypical lipomatous tumors (usually deeper tissues than SCL), myofibroblastoma, neurofibromas, dermatofibrosarcoma protuberans, and liposarcoma.⁵ Because Enzinger and Harvey⁶ first described SCL in 1975, there have been copious nonclassical cases reported in literature with diversity in location, cytologic composition, progression, or even patient demographics. Although some previous reports review related cases of specific rare presentations or institution-based summaries, no comprehensive summary of published nonclassical SCL case reports has been done. This article will review the current literature regarding nonclassical SCLs and discuss the demographics of nonclassical SCL: gender, age, race, location, size, unique histology, invasion, and multiple lesions. Additionally, we evaluated those cases with multiple nonclassical findings in a single individual as well as the demographics of these patients who likely will require a wider local excision so as to avoid possible recurrences and subsequent surgeries in highly sensitive cosmetic areas. We will also present a new case to the literature encountered by our department of a unique fast-growing SCL infiltrating into skeletal muscle, located in two positions on the chin of an 18-year-old African American woman.

METHODS

A decision was made to perform a review of the literature via a query of PubMed online database to capture published nonclassical presentations of SCL from 1978 to 2018. The Medical Subject Headings terms of “SCL,” “lipomatous tumor,” “atypical,” “soft tissue tumor,” and “adipocytic neoplasm” were searched. The primary outcome of interest was to characterize nonclassical SCL. We identified applicable case reports, and these cases were analyzed for differentiating characteristics, such as patient demographic, cytologic composition, abnormal progression, and lesion location. We explored any barrier to characterization. These results were further analyzed as far as gender, ethnicity, age [defined as young (<40 years of age) and old (>40 years of age)], location, size distribution with a subset of larger than 13 cm, nonclassical cytology, bone erosion or infiltrating muscle, multiple locations, and finally, multiple nonclassical features present in each case. Those with multiple nonclassical features were studied for common characteristics. All statistics were performed with Excel (Microsoft, 2018) using the Fisher exact test for all categorical data, and a *P* value of 0.05 was determined to be statistically significant.

CASE REPORT

An 18-year-old African American woman with no medical history, including family cancers, presented with a several-month history of bilateral chin masses, stating the mass on the left was painful. The left chin mass was 3.5×3 cm, mobile with no associated skin changes, and the right chin mass was only discreetly palpable; however, the patient desired to have both removed. A computed tomography (CT) scan was obtained to evaluate the underlying tissues and possible involvement, which did

Takeaways

Question: We examined nonclassical spindle cell lipoma (SCL) for patterns.

Findings: A new case of a unique fast growing SCL infiltrating into skeletal muscle, located in two positions on the chin of an 18-year-old African American woman. Females and young patients commonly present with multiple nonclassical SCL findings.

Meaning: Nonclassical SCL can alert the clinician to the increased risk of rapid regrowth or invasion specifically in young and female patients.

not show a discrete mass on either side, but only diffuse subcutaneous soft tissue fat stranding (Fig. 1).

The bilateral masses were excised cosmetically with a skin-sparing technique and submandibular incisions (Fig. 2). Macroscopically, the specimens were pink-tan rubbery tissue 1.6×1.0 cm on the left and 0.7×0.5 cm on the right (Fig. 3).

Less than 2 months after the first excision, she presented to the clinic with recurrent painful chin masses. These masses were confirmed on ultrasound and found to be larger, firmer, and more fixed than before.

Intraoperatively, due to skin involvement with the scar from the previous excision, a more radical reexcision of masses and surrounding soft tissue was done from an infra-mandibular approach widely over the mentum to ensure complete excision of the masses with advancement flaps to allow for placement of the final scar closure on the inferior aspect of the mandibular border. Pathology again was consistent both macroscopically and microscopically with

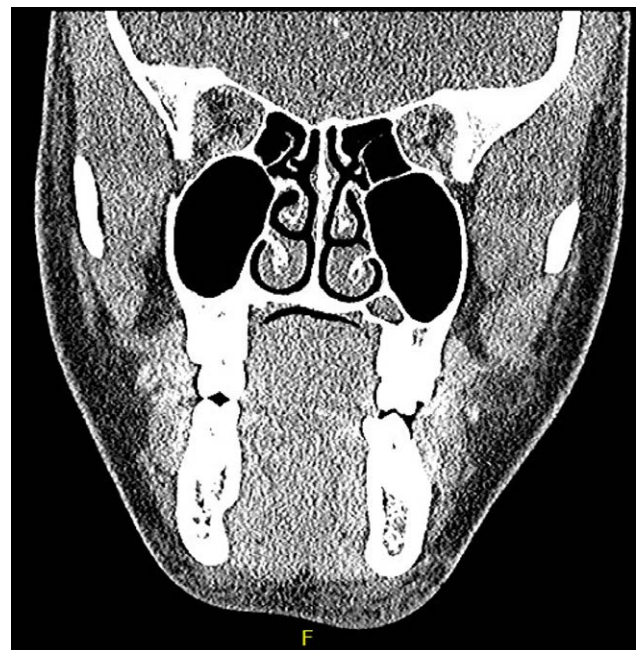


Fig. 1. This is an 18-year-old African American woman who presents with painful, fast-growing submandibular masses. CT findings of nondescript soft tissue fat stranding shown in the image.



Fig. 2. Intraoperatively, the bilateral palpable submandibular masses were marked for excision.

SCL with reactive changes. Postoperatively, the drain was removed without complication. She has been compliant with a chin garment and scar massage and presented to her 3-month follow-up without any complications (Fig. 4). After this unique presentation, informed written consent was obtained from the patient to share information and images.

RESULTS

During the 40-year time frame of 1978–2018, a total of 12 reported cases of nonclassical SCL were published in the literature. (See appendix, Supplemental Digital Content 1, which displays the reference lists, <http://links.lww.com/PRSGO/C130>.) Demographic analysis of these case reports demonstrated that a majority of nonclassical patients were men (71%) and older than 40 (89%). Only 22 of the 125 cases explicitly stated race or ethnicity, while 13 cases included a patient image that allowed for inference of such information (Table 1). The most common location was classical: upper back, shoulder, or posterior neck. Of the 14 cases under 40, seven were located in the



Fig. 3. Macroscopic appearance of SCL.



Fig. 4. The patient presents for her 3-month follow-up (of her second excision) with no signs of recurrence.

face, oropharynx, and orbit regions. Only five cases were larger than average size SCL with a range from 17 to 42 cm and average size of 24 cm. Only 17 (13.6%) of the nonclassical SCL cases showed unique histology. Furthermore, of the 125 cases, two (1.6%) were noted to have bone erosion, and six (4.8%) had an intramuscular component. There were only three (2.4%) nonclassical SCL cases with multiple lesions. There was one patient with five lesions, another with four lesions, and another with three. One of the patients had two nonclassical findings with a lesion on the tongue, and another patient had three nonclassical findings as a young patient with multiple lesions on extremities.

The final nonclassical criteria our study evaluated were the presence of multiple nonclassical findings (Table 2). The majority of the patients had only one nonclassical finding (67, 53.6%).

Forty nonclassical SCL cases presented with two nonclassical findings (32%). Of those 18 cases with three or more nonclassical findings: 72% were young (under 40), 66% were women, and 55% were a non-White minority. There were 15 patients with three nonclassical findings (12%), two patients with four findings (1.6%), and only one had five nonclassical findings (0.8%). The patient who had five nonclassical findings was a young Indian woman with unique cytology on her thumb.

Further analysis was performed to look at the data among certain demographics. Of note, we specifically

Table 1. Demographics and Characteristics

Characteristics	N	Percent
Gender, n		
Men	89	71.2
Women	36	28.8
Median age, (range) y	55 (14 mo–82 y)	
Classical age (>40 y), n	111	88.8
Nonclassical (<40 y), n	14	11.2
Race information available, n	35	28
Asian	17	48.6
White	14	40
African American	3	8.6
Location, n		
Classical	29	23.2
Oral	20	16
Extremities	11	8.8
Pharynx/larynx	10	8.0
Face	9	7.2
Ocular	9	7.2
Breast	7	5.6
Tongue	7	5.6
Scalp	4	3.2
Nasal cavity	4	3.2
Genitourinary	4	3.2
Other organs	3	2.4
Gastrointestinal tract	2	1.7
Spermatic cord	2	1.7
Anterior neck	1	0.8
Perianal	1	0.8
Supraclavicular	1	0.8
Buttock	1	0.8
Size mentioned, n	107	85.6
Large size (>13 cm)	5	4.7
Median large size (range), cm	20 (17–42)	
Unique histology, n	17	13.6
Pseudoangiomatous	8	47.1
Fibrous tissue	2	11.8
Verocay	1	5.9
Dermal plexiform	1	5.9
Low fat variant	1	5.9
Pedunculated pleomorphic	1	5.9
Dendritic fibromyxolipoma	1	5.9
Extramedullary hematopoiesis	1	5.9
Collagen rosettes	1	5.9
Bone Erosion, n	2	1.6
Intramuscular, n	6	4.8
Multiple lesions, n	3	2.4

Indentation indicates subgroup and percentage of that group.

sought to look at age and gender with respect to the number of nonclassical findings present, as these characteristics are what we believed distinguished our patient

Table 2. Multiple Nonclassical Findings in One Individual

Characteristics	N	Percent	P
One nonclassical finding	67	53.6	
Women	0	0	<0.001
Age (<40)	0	0	<0.001
Two nonclassical findings	40	32	
Women	24	60	
Age (<40)	7	17.5	
Non-White	5	12.5	
Three nonclassical findings	15	12	
Age (<40)	10	66.7	
Woman	9	60	
Non-White	7	46.7	
Four nonclassical findings	2	1.6	
Women	2	100	
Age (<40)	2	100	
Non-White	2	100	
Five nonclassical findings	1	0.8	
Women	1	100	
Age (<40)	1	100	
Non-White (Indian)	1	100	

Indentation indicates subgroup and percentage of that group, and race was not noted in majority of one nonclassical finding group.

from other reports. When comparing men to women among all categories, it was noted that there were no women with one isolated nonclassical finding, which was statistically significant ($P < 0.001$). In examining age, for those younger than 40, there were no reports of a young patient with only one nonclassical finding as all 19 patients under 40 had more than one nonclassical finding ($P < 0.001$). When comparing three or more nonclassical findings to only two nonclassical findings, there was also a statistically significant higher predominance of more than three findings in the young group ($P < 0.001$).

DISCUSSION

Since 1975, when SCL was first described, there has not been a comprehensive review of the nonclassical cases. While SCL in and of itself is a rare finding, there have been reviews in the literature of classical SCL as well as a few smaller reviews of nonclassical SCL. Briefly, D’Antonio et al⁷ reviewed two cases with locations in the face and larynx, Zamecnik⁸ evaluated three cases of the pseudoangiomatous variant of SCL, Said-Al-Naief et al⁹ described two cases of SCL located in the oral cavity, and Diaz-Cascajo et al¹⁰ looked at three cases with unique cytologies of fibrous and pleomorphic SCL. There were a few larger series, most notably Ko et al,¹¹ which looked at women as a nonclassical feature of 395 cases of classical SCL. Another larger study was Ud Din et al,¹² which looked at atypical locations as a nonclassical feature of 439 classical cases. However, all of these reviews either examine only one nonclassical feature or are limited in the number of cases and scope. No reviews to date have comprehensively categorized all the different reported features of nonclassical SCL as we have sought to do with this review.

There have been 125 cases of nonclassical SCL reported in literature since 1978. Nonclassical SCL has similar characteristics to classical SCL with the majority presenting as a single lesion on the back, shoulder, or posterior neck area (23%) of older (88.8%) men (71.2%), despite their other nonclassical findings. Interestingly, when examining women in these reports, they are significantly more likely to present with more than one nonclassical finding ($P < 0.001$). Additionally, young patients (under 40 years of age) also demonstrated a significant predisposition to having more than one nonclassical finding ($P < 0.001$). These young patients were also more likely to have three or more nonclassical findings than just two nonclassical findings ($P < 0.001$).

Our patient is a young African American woman who presented with fast-growing, multiple lesions on her face. When compared with the 125 other nonclassical cases, she is the only case with six nonclassical findings: a young (11.2%), African American (3%), woman (29%) who presents with multiple lesions (2.4%) with intramuscular invasion (4%) on her face (7.2%). Her quick recurrences are an important reason for reporting this case as a warning and evaluating nonclassical SCL, especially those with multiple nonclassical findings. The return of her SCL might have been related to an incomplete resection versus an atypical growth pattern that has yet to be characterized. Realistically, this is likely a combination of both, as we see

that increased nonclassical findings result in potential for bone erosion (both cases were in those with three nonclassical findings), large size, or multiple locations.

Bhat et al¹³ presented the only other case with more than four multiple nonclassical findings. Their unique patient had five nonclassical findings and was similar to our case as she was also a young, minority (Indian), woman.

There were three patients with the ethnicity of African American listed, but 72% had no ethnicity identified. We do appreciate that it is a limitation of this review in that it is unclear how many nonclassical cases were truly minority ethnicities. One African American case was a man with a large lipoma in the classical location.¹⁴ Another was a man with a lipoma in the oral cavity.⁹ The final African American case was similar to ours with four nonclassical findings in a young African American woman with a lesion in her oral cavity.¹⁵ Of the six that showed intramuscular invasion, only one case had more than one nonclassical finding.¹⁶

Classical SCL has a firmer, more rubbery consistency on physical examination than standard lipomas. Regarding treatment, standard of care advocates for wide local excision. However, this can pose some concerns among the nonclassical groups, where locations, such as face/neck/oropharynx/orbit, can make wide local excision result in less than ideal outcomes for cosmesis and function. It was noted among the nonclassical cases, and 50% of these cases were located in face/neck/oropharynx/orbital; furthermore, 37% of the lesions occur in these regions of patients under 40.

We believe that knowing this predominance for the head and neck region in this subset of young female patients can help aid diagnosis and treatment planning. If a firmer, more rubbery consistency mass is found on examination, which does not seem to be a classic lipoma in presentation, we feel the surgeon should keep the possibility of a nonclassical SCL as a possible differential diagnosis. Furthermore, the surgeon should consider a woman who is under 40 to be likely to have more nonclassical findings: possibly invasion, unique cytology, or other characteristics, which could, as in our case, result in rapid recurrence. Considering these findings, the surgeon may plan for the need for a wider local excision as well as cosmetic closure. This might encourage the intervention of a surgeon who is able to create potential flaps or reconstructive coverage in an emotionally sensitive area such as the face of a young individual.

CONCLUSIONS

Our patient is the only unique report of six nonclassical findings: multiple locations, infiltrative, fast-growing SCLs, which presented on the chin of a young African American woman. Despite predominance of typical SCL

presentation, a relative abundance of nonclassical cases have been reported in the literature. Thorough characterization of the rare nonclassical cases of this benign condition could guide diagnostic decision-making and identify trends in disease presentation over time as well as alert the clinician to the increased risk of rapid regrowth or invasion in an individual with nonclassical findings, especially in young and female patients.

Abigail J. Engwall-Gill, MD
Department of Surgery
Sparrow Hospital
1215 East Michigan ave
Lansing, MI 48912
E-mail: engwalla@msu.edu

PATIENT CONSENT

The patient provided written consent for the use of her image.

REFERENCES

1. Seo BF, Kang IS, Oh DY. Spindle cell lipoma: a rare, misunderstood entity. *Arch Craniofac Surg*. 2014;15:102–104.
2. Domanski HA, Carlén B, Jonsson K, et al. Distinct cytologic features of spindle cell lipoma. A cytologic-histologic study with clinical, radiologic, electron microscopic, and cytogenetic correlations. *Cancer*. 2001;93:381–389.
3. Thompson LDR. Spindle-cell lipoma. *Ear Nose Throat J*. 2009;88:992–993.
4. Forcucci JA, Sugianto JZ, Wolff DJ, et al. “Low-fat” pseudoangiomatous spindle cell lipoma: a rare variant with loss of 13q14 region. *Am J Dermatopathol*. 2015;37:920–923.
5. Fletcher CDM, Martin-Bates E. Spindle cell lipoma: a clinicopathological study with some original observations. *Histopathology*. 1987;11:803–817.
6. Enzinger FM, Harvey DA. Spindle cell lipoma. *Cancer*. 1975;36:1852–1859.
7. D’Antonio A, Mottola G, Caleo A, et al. Spindle cell lipoma of the larynx. *Ear Nose Throat J*. 2013;92:E9.
8. Zamecnik M. Pseudoangiomatous spindle cell lipoma with “true” angiomatous features. *Virchows Arch*. 2005;447:781–783.
9. Said-Al-Naief N, Zahurullah FR, Sciubba JJ. Oral spindle cell lipoma. *Ann Diagn Pathol*. 2001;5:207–215.
10. Diaz-Cascajo C, Borghi S, Weyers W. Fibrous spindle cell lipoma: report of a new variant. *Am J Dermatopathol*. 2001;23:112–115.
11. Ko JS, Daniels B, Emanuel PO, et al. Spindle cell lipomas in women: a report of 53 cases. *Am J Surg Pathol*. 2017;41:1267–1274.
12. Ud Din N, Zhang P, Sukov WR, et al. Spindle cell lipomas arising at atypical locations. *Am J Clin Pathol*. 2016;146:487–495.
13. Bhat A, Vijaya C, Rao SB. Pseudoangiomatous variant of spindle cell lipoma: report of a rare case. *Indian J Pathol Microbiol*. 2016;59:376–378.
14. Ghazanfari A, Oppenheimer R. Massive spindle cell lipoma. *Otolaryngol Head Neck Surg*. 2006;134:164–165.
15. McDaniel RK, Newland JR, Chiles DG. Intraoral spindle cell lipoma: case report with correlated light and electron microscopy. *Oral Surg Oral Med Oral Pathol*. 1984;57:52–57.
16. Sund S, Hordvik M, Maehle B, et al. Large intramuscular spindle-cell lipoma. With review of the literature. A case report. *APMIS*. 1988;96:347–351.