

Study of a Subcutaneous Lesion of the Shoulder: The Saint Paolino Tumor

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Summary: In certain small counties in southern Italy, traditional Catholic festivals are observed by erecting tall, large, and weighty wagons referred to as “lilies.” These wagons are borne on the shoulders of several individuals known as “cradles.” This practice has given rise to the emergence of a distinct subcutaneous neof ormation on the shoulder. This study investigates the unique clinical and anatomopathological attributes of “Saint Paolino tumor” (named in honor of the Catholic patron of the widely celebrated lilies festival). This tumor presents as a posttraumatic intermittent chronic lesion occurring on the shoulder, necessitating differential diagnosis from other cutaneous and soft tissue lesions such as spontaneous lipomas, elastofibroma, Madelung disease, and liposarcoma. (*Plast Reconstr Surg Glob Open* 2024; 12:e5946; doi: 10.1097/GOX.0000000000005946; Published online 2 July 2024.)

Lipomas are benign tumors composed of mature fat cells. Most solitary lipomas are superficial and small.¹ Like genetic or familial disorders, these lesions may arise spontaneously or subsequent to acute and chronic trauma.^{2,3} However, the precise role of trauma in lipoma formation remains a subject of debate. Recent years have witnessed associations between acute trauma and lipoma development.^{4,5} The initial instance of a post-traumatic lipoma at a site of chronic trauma was documented by Cairns among wine porters and substantiated by Brenner.^{6,7} Specific towns in southern Italy observe traditional Catholic festivals featuring the assembly of large, heavy wagons termed “lilies” (named after the lily plant’s long stems). These wagons are borne on the shoulders by multiple participants known as “cradles.” The most prominent lily festival takes place annually in Nola, Italy, coinciding with the patronal feast dedicated to Saint Paolino. This study aimed to elucidate the characteristic subcutaneous neof ormation that develops on the cradles’ shoulders due to intermittent, prolonged pressure insults—an

unequivocal manifestation of bodily adaptation to these practices (Figs. 1 and 2). The research delves into the distinctive clinical and pathological attributes of Saint Paolino tumor (SPT), manifesting as a posttraumatic, intermittent, chronic shoulder lesion, while differentiating it from other cutaneous and soft tissue lesions.

THE CASE SERIES

We evaluated 16 cradle patients for subcutaneous neof ormations on the shoulder. (See table, Supplemental Digital Content 1, which shows the 16 cradles evaluated. <http://links.lww.com/PRSGO/D327>.) Comprehensive clinical data, medical histories, ultrasound assessments, and patient photographs were obtained. Surgical intervention was conducted on three patients, with subsequent pathological anatomical evaluations of the excised masses. The remaining cradles have declined undergoing surgical intervention and have opted to postpone it, despite being informed about the risks of not undergoing surgical removal. Moreover, they expressed a particular sentiment regarding the subcutaneous neof ormation on the shoulder, recognizing it as a distinctive emblem to be proudly embraced. The mean age was 44 years (range: 33–58 years), with a mean body mass index of 33 kg per m² (range: 29–40). No patients with genetic or familial histories exhibited subcutaneous fat typical of Madelung disease. Bilateral localization was observed in five patients. Ultrasound examinations yielded uniform characteristics across all cases, revealing multilocular and

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Fig. 1. Presurgery images of the shoulder mass of a cradle patient in frontal view.



Fig. 2. Presurgery images of the shoulder mass of a cradle patient in backward view.

multiseptal organization of subcutaneous hypertrophic fat. Color Doppler imaging indicated heightened vascularity. The three cradle patients underwent surgical excision followed by pathological examination, revealing mature

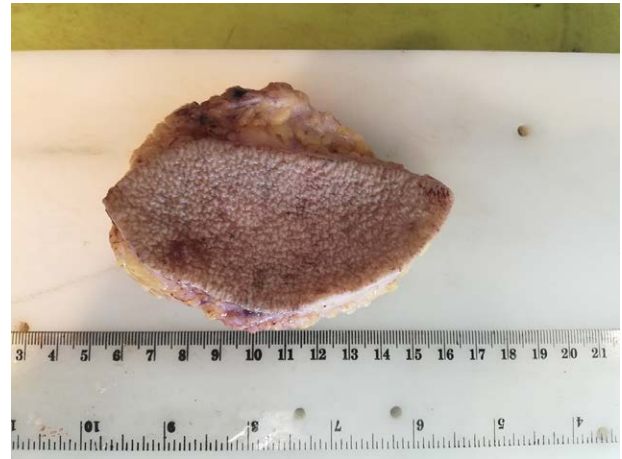


Fig. 3. The macroscopic features of the excised lesion.

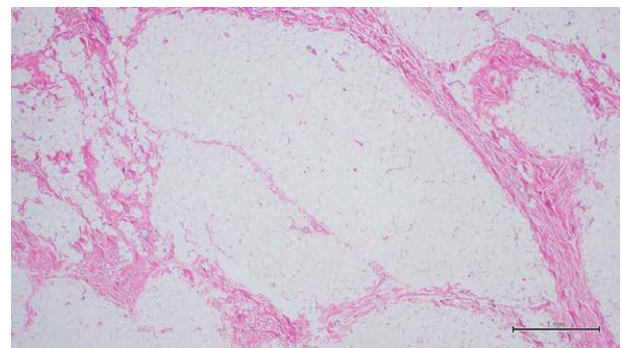


Fig. 4. Frozen section view under microscope, showing the proliferation of mature adipocytes with variable fibrous septa and collagen.

adipocyte proliferation with varying fibrous septa and collagen in all instances. No cytological atypia or necrosis was observed (Figs. 3 and 4). A 1-year follow-up revealed no instances of recurrence.

DISCUSSION

The term SPT denotes a distinct subcutaneous neoplasm on the shoulders of lily bearers—a specific chariot emblematic of a prevalent southern Italian religious festival. Based on gathered clinical histories, this neof ormation develops due to intermittent, long-term pressure on the shoulder. Participants reported carrying loads averaging 50–60kg on their shoulders for roughly 3–4 hours during the festival. This lesion typically manifests around the third repetition of this practice, spaced approximately 6 months apart. In contrast to Cairns' description of wine porters who developed lipomas through sustained daily pressure, SPT's development pattern and dimensions differed. Various neoplastic formations were considered for differential diagnosis, including elastofibroma dorsi, spontaneous lipomas, Madelung disease, and liposarcoma. Liposarcomas are rare malignant tumors with adipocytic differentiation.⁸ Well-differentiated liposarcomas closely resemble mature

adipose tissue but typically exhibit fibrous septation with variable nuclear atypia and enlargement.⁹ Pathological assessment of SPT indicated a lack of cytological atypia, instead revealing proliferated mature adipocytes. Madelung disease, or multiple symmetric lipomatosis, is a rare metabolic disorder characterized by diffuse and symmetric fat tissue accumulation, slow tumor growth, and progressive enlargement, which occurs without trauma.¹⁰ Elastofibroma may manifest as a solid mass due to local mechanical friction, resulting in vascular injury and a hyperplastic scarring response characterized by regenerative hypertrophy of elastic fibers.^{11, 12} However, clinical assessment confirmed SPT as a solid mass situated superficially—more aligned with lipomas, which typically reside in superficial layers. Both lipoma and elastofibroma dorsi exhibit slow growth and can emerge due to trauma. However, pathological evaluations have clarified their nature. SPT seemed akin to a typical lipoma but featured a more fibrotic component akin to a fibrolipoma (Figs. 3–4). The possibility of liponecrosis formation due to prolonged pressure ischemia was considered.¹³ However, further investigations are warranted.

CONCLUSIONS

SPT, a benign neoplasm afflicting adipocytes, results from intermittent pressure trauma. Despite the elongated intervals between traumatic events (up to 6 months), this trauma seems to stimulate neof ormation growth, occasionally attaining significant dimensions (Figs. 1 and 2). SPT warrants inclusion in the differential diagnosis of lipomas, elastofibromas, Madelung disease, and liposarcoma. Although its occurrence aligns with the unique practices of lily cradles, pathological examination provides a definitive diagnosis more closely aligned with lipomas. The growth involves proliferated mature adipocytes, alongside an augmented fibrous and collagenous component, justifying its classification as a fibrolipoma. Such formations are emblematic of those involved in this cultural practice and are carried with pride as a distinct symbol. Consequently, individuals often decline surgical intervention. However, in select cases, individuals may opt to abandon the cultural practice and undergo surgery due to these neoformations attaining considerable dimensions and exerting substantial aesthetic and functional impacts.

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DISCLOSURE

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

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All procedures performed in this study involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and the 1964 Declaration of Helsinki and its later amendments or comparable ethical standards.

REFERENCES

1. Johnson CN, Ha AS, Chen E, et al. Lipomatous soft-tissue tumors. *J Am Acad Orthop Surg*. 2018;26:779–788.
2. Mejía Granados DM, de Baptista MB, Bonadia LC, et al. Clinical and molecular investigation of familial multiple lipomatosis: variants in the HMG A2 gene. *Clin Cosmet Investig Dermatol*. 2020;13:1–10.
3. Caponnetto F, Manini I, Bulfoni M, et al. Human adipose-derived stem cells in Madelung's disease: morphological and functional characterization. *Cells*. 2020;10:44.
4. Tewfik K, Covelli C, Rossini M, et al. Lump on the scalp of a child arising over a previous parietal fracture: growing skull fracture or post-traumatic lipoma? *BMJ Case Rep*. 2022;15:e246283.
5. Bokhari RF, Bangash MH, Ahamed NA, et al. A symptomatic Sylvian fissure lipoma in a post-traumatic patient. *J Radiol Case Rep*. 2014;8:1–7.
6. Cairns RJ. The subcutaneous fat. In: Rook A, Wilkinson DS, Ebling FJG, eds. *Textbook of Dermatology*. 2nd ed. Oxford, England: Blackwell Scientific Publications; 1972:1510.
7. Brenner S. Lipoma on the shoulder in site of chronic trauma. *Arch Dermatol*. 1983;119:450–451.
8. Lee ATJ, Thway K, Huang PH, et al. Clinical and molecular spectrum of liposarcoma. *J Clin Oncol*. 2018;36:151–159.
9. Thway K. Well-differentiated liposarcoma and dedifferentiated liposarcoma: an updated review. *Semin Diagn Pathol*. 2019;36:112–121.
10. Liu Q, Lyu H, Xu B, et al. Madelung disease epidemiology and clinical characteristics: a systemic review. *Aesthetic Plast Surg*. 2021;45:977–986.
11. Nishio J, Nakayama S, Nabeshima K, et al. Current update on the diagnosis, management and pathogenesis of elastofibroma dorsi. *Anticancer Res*. 2021;41:2211–2215.
12. Turna A, Yilmaz MA, Urer N, et al. Bilateral elastofibroma dorsi. *Ann Thorac Surg*. 2002; 73:630–632.
13. Requena L. Normal subcutaneous fat, necrosis of adipocytes and classification of the panniculitides. *Semin Cutan Med Surg*. 2007;26:66–70.