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J Case Rep Images Pathol 2022;8(2):17–21.
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Solitary nodule of cutaneous reticulohistiocytosis: A case report

Jeffrey E Fournier, Tiffany Shao, Snezana Popovic, Salem Alowami

ABSTRACT

Introduction: Solitary cutaneous reticulohistiocytosis represents a rare form of benign monocyte/macrophage proliferation. On routine histology, these lesions are typically described as large cells with cytoplasm showing ground glass appearance and giant cells. They grow up to 1 cm in size with rare cases exceeding this size.

Case Report: This case report of a 28-year-old male demonstrated a nodule of reticulohistiocytosis measuring 2.2 cm in size. Microscopic features showed a well-demarcated nodule in the dermis with large histiocytes with ground-glass eosinophilic cytoplasm, giant cells, and foamy macrophages in a background of mixed inflammatory cells. Immunohistochemical staining showed positive staining for vimentin, CD68, CD31, with focal and patchy positivity for S100, CD43, and CD45 and negative staining for CD1a, langerin, CD21, CD23, CD30, CD34, ERG, D2-40, AE1/AE3, epithelial membrane antigen (EMA), smooth muscle actin (SMA), myogenin, desmin, SOX10, HMB-45, tyrosinase, and MelanA.

Conclusion: The microscopic and immunohistochemical findings are characteristic of this entity but it is important to recognize for proper management and differentiation from other malignant lesions..

Keywords: Cutaneous, Dermal, Reticulohistiocytosis, Solitary

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Received: 21 September 2022
Accepted: 23 November 2022
Published: 31 December 2022

How to cite this article

Fournier JE, Shao T, Popovic S, Alowami S. Solitary nodule of cutaneous reticulohistiocytosis: A case report. J Case Rep Images Pathol 2022;8(2):17–21.

Article ID: 100066Z11JF2022

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doi: 10.5348/100066Z11JF2022CR

INTRODUCTION

Histiocytoses refer to a group of rare disorders that include a proliferation and accumulation of monocyte/macrophage or dendritic cells [1]. Specifically, reticulohistiocytosis (previously referred to as reticulohistiocytoma) refers to solitary and multicentric histiocytic proliferations that have common histological findings of cells with abundant granular eosinophilic cytoplasm (ground glass appearance) mixed with giant cells, with or without systemic involvement respectively [2, 3]. Multiple case reports and few case series have been published on this entity, and it has been shown that the majority measure less than 1 cm with rare cases growing to larger sizes [4–7]. This case report demonstrates microscopic and immunohistochemical findings of a 2.2 cm solitary cutaneous reticulohistiocytosis.

CASE REPORT

A 28-year-old male patient presented to his family physician with a raised lesion in the left anterior

axillary line in August 2021. The lesion was first noticed only months prior in May 2021 and had continued to grow and was reported to intermittently bleed. At the time of presentation, the lesion, a soft and raised nodule, measured 2.2×1.7 cm. No other lesions were identified or noted on clinical examination at the time of resection. When the lesion was resected, the clinical impression was that of a squamous cell carcinoma versus keratoacanthoma.

Microscopic examination demonstrated skin with a well-demarcated nodule involving the dermis and superficial subcutaneous tissue (Figure 1). It was mainly composed of a dense population of large histiocytes with glassy abundant, eosinophilic cytoplasm admixed with occasional giant cells and foamy macrophages (Figure 2). Many of the larger cells demonstrated emperipolesis throughout the lesion. This was in the background of mixed inflammatory cells including many eosinophils and neutrophils. The overlying epithelium appeared to be intact and uninvolved. There was minimal hemosiderin pigmentation consistent with prior hemorrhage.

Immunohistochemical staining was performed and demonstrated positive staining for vimentin, CD68, and CD31 (Figure 3), with focal and patchy positivity for S100, CD43, and CD45 (Figure 4); and negative staining for CD1a, langerin, CD21, CD23, CD30, CD34, ERG, D2-40, AE1/AE3 (Figure 5), EMA, SMA, myogenin, desmin, SOX10, HMB-45, tyrosinase, and MelanA (Figure 6). Molecular studies were performed, including BRAF (exons 11 and 15), Kit (exons 8, 9, 10, 11, 13, 14, 17, and 18), KRAS (exons 2–4), and NRAS 2–4, and the lesion showed no clinically significant sequence variants detected in these regions.

A final diagnosis of "cutaneous histiocytic proliferation, favor reticulohistiocytosis" was rendered. The lesion was completely excised at the time of diagnosis and no further management or treatment was performed.

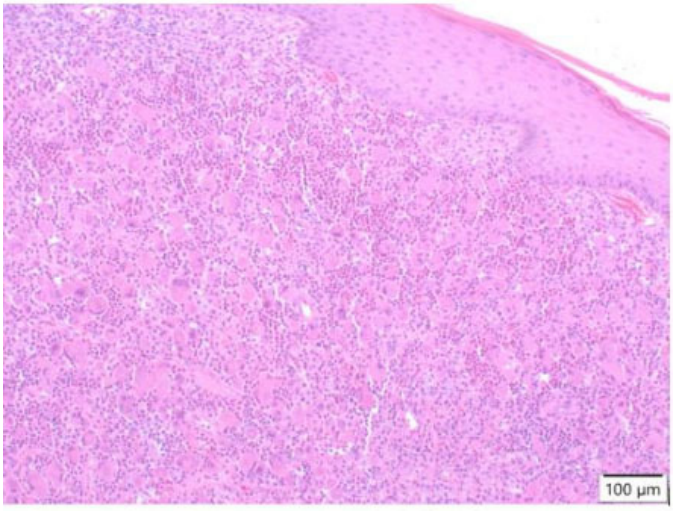

Figure 1: Reticulohistiocytosis viewed at intermediate power magnification (100×; hematoxylin and eosin).

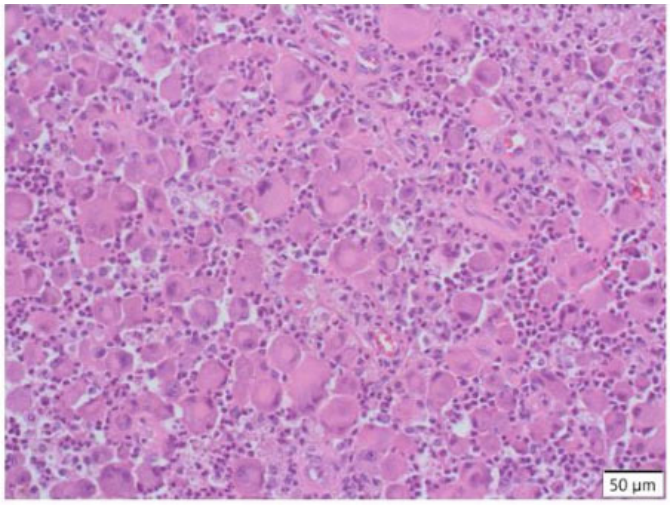

Figure 2: Reticulohistiocytosis viewed at high power magnification (200×; hematoxylin and eosin).

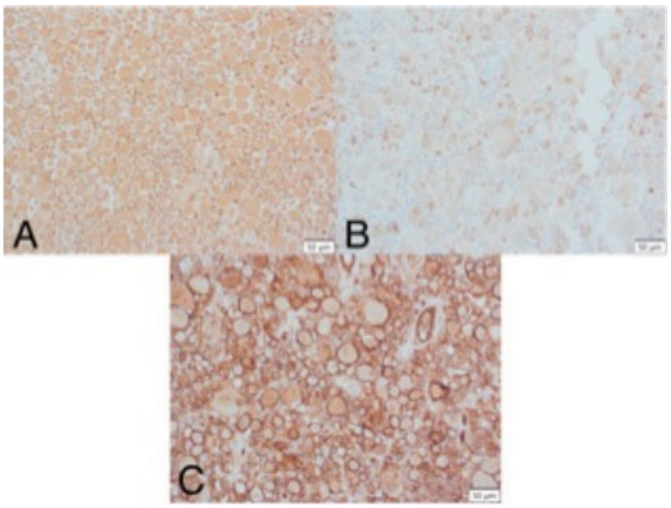

Figure 3: Reticulohistiocytosis with positive (A) vimentin, (B) CD68, and (C) CD31 immunohistochemical staining (200×).

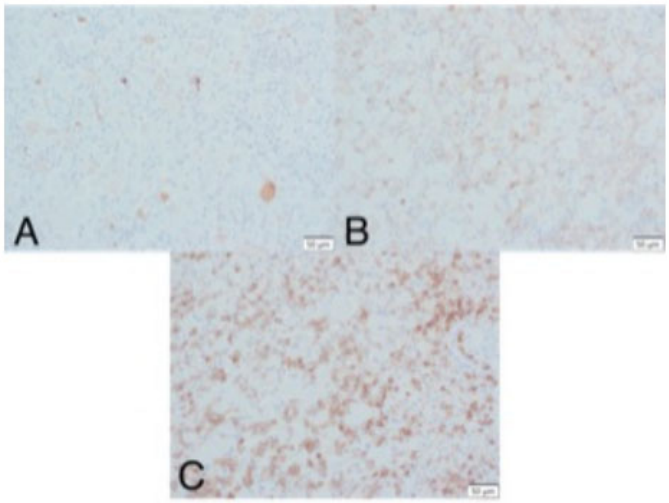

Figure 4: Reticulohistiocytosis with focal and patchy positive (A) S100, (B) CD43, and (C) CD45 immunohistochemical staining (200×).

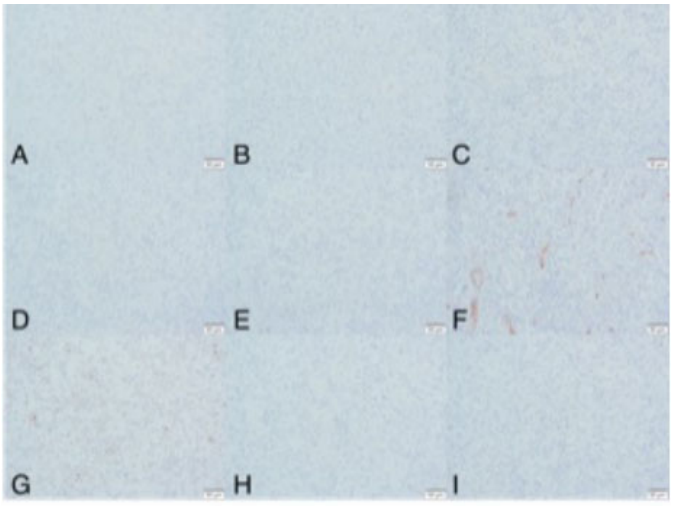

Figure 5: Reticulohistiocytosis with negative (A) CD1a, (B) langerin, (C) CD21, (D) CD23, (E) CD30, (F) CD34, (G) ERG, (H) D2-40, and (I) AE1/AE3 immunohistochemical staining (200×).

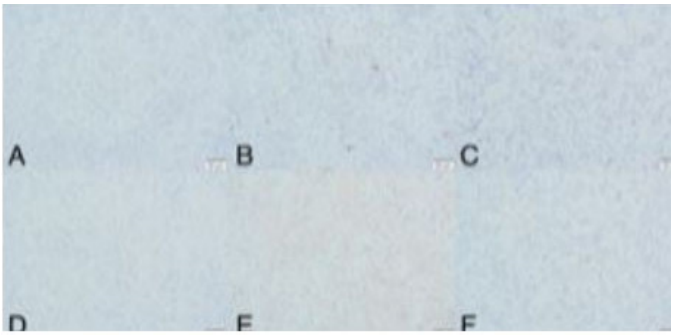

Figure 6: Reticulohistiocytosis with negative (A) EMA, (B) SMA, (C) myogenin, (D) desmin, (E) SOX10, and (F) melanoma cocktail (HMB-45, tyrosinase, and MelanA) immunohistochemical staining (200×).

DISCUSSION

Reticulohistiocytosis is a rare disorder of histiocytes with characteristic findings and three clinical presentations: solitary, diffuse, and multicentric [8]. These most commonly present clinically as multicentric with systemic involvement in the literature [9–12]. Currently, the etiology of solitary cutaneous reticulohistiocytosis remains unclear, but it is believed to represent a reactive process to trauma, inflammatory process, or other unknown causes [2, 13, 14]. The differential diagnosis of these lesions includes other disorders such as Langerhans cell histiocytosis, Rosai–Dorfman disease, juvenile xanthogranuloma, necrobiotic xanthogranuloma, melanocytic lesions, and histiocytic sarcoma [1, 4].

Typically, these lesions present as a yellow to reddish-brown papule or nodule with a smooth surface lacking ulceration [13, 15]. Although the microscopic findings of "ground glass" eosinophilic cytoplasm and giant cells are characteristic, the differential histologically remains quite broad. It is therefore important to utilize immunohistochemical studies to differentiate from other benign and malignant conditions or conditions with genetic implications [16]. Regardless of the type, all forms of reticulohistiocytosis demonstrate the same pattern of immunohistochemical staining [2]. Typical immunohistochemical staining patterns show positivity for CD68, CD163, CD45, vimentin, and negativity for CD1a, langerin, and focal to negative S100 staining [4, 8, 17, 18].

Solitary cutaneous reticulohistiocytosis typically does not exceed 1 cm in diameter; however, rarely larger lesions have been reported [4, 19]. Diagnostic difficulties might arise clinically and histologically as a lesion larger than 1 cm may not be considered likely based on size alone. This pitfall may result in increased pathology reporting turnaround time, increased costs associated with immunohistochemical and other special ancillary tests, and increased patient anxiety while awaiting results of a biopsy or resection.

Once a diagnosis of reticulohistiocytosis is made, a full clinical examination and investigations should be undertaken to work up a patient for systemic involvement or other conditions [2]. As there are three clinical types of reticulohistiocytosis, a pathologist should not sub-classify based on a single biopsy or resection specimen. In such cases, a broad diagnosis of "reticulohistiocytosis" should be made. Solitary reticulohistiocytoses have been known to resolve on their own but may require surgical excision for symptomatic or cosmetic reasons [20–22]. Local recurrence is uncommon after incomplete surgical excision and is especially uncommon after complete surgical resection [4, 23].

CONCLUSION

This represents a case report of a large reticulohistiocytosis with characteristic microscopic and immunohistochemical findings. Although these lesions are benign and rare, with larger lesions being even rarer, it is important to produce a proper diagnosis for patients to receive appropriate management and to limit confusion with other malignant entities.

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Author Contributions

Jeffrey E Fournier – Conception of the work, Design of the work, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Tiffany Shao – Analysis of data, Interpretation of data, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Snezana Popovic – Analysis of data, Interpretation of data, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Salem Alowami – Conception of the work, Interpretation of data, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Guarantor of Submission

The corresponding author is the guarantor of submission.

Source of Support

None.

Consent Statement

Written informed consent was obtained from the patient for publication of this article.

Conflict of Interest

Authors declare no conflict of interest.

Data Availability

All relevant data are within the paper and its Supporting Information files.

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