



Case Report

Bilateral Idiopathic Localized Involutional Lipoatrophy in a Child: A Case Report

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ABSTRACT

Idiopathic localized involutional lipoatrophy (ILIL) is a rare form of localized lipoatrophy characterised by focal fat loss without a preceding cause. It commonly affects females and typically presents as unilateral lesions. Childhood cases are exceptionally rare. We report a unique case of bilateral ILIL in a 5-year-old male child with a positive familial occurrence in his 4-year-old maternal cousin, marking a rare presentation. The patient exhibited symmetrical, non-tender, well-demarcated depressed plaques over the gluteal region without systemic involvement. Histopathological findings showed dermal fibrosis and epidermal depigmentation, confirming the diagnosis of ILIL. Tacrolimus 0.1% ointment was prescribed, leading to stabilisation and peripheral repigmentation within 4 weeks. This case highlights the importance of recognising ILIL in unusual demographics, including male children with familial associations and differentiating it from other causes of lipoatrophy.

Keywords: Bilateral lesions, Childhood lipoatrophy, Familial lipoatrophy, Idiopathic lipoatrophy, Idiopathic localized involutional lipoatrophy

INTRODUCTION

Idiopathic localized involutional lipoatrophy (ILIL) is a rare disorder characterised by localised loss of subcutaneous fat without preceding trauma or systemic disease. Most reported cases involve adolescent or adult females with unilateral lesions.^[1] However, recent literature has documented numerous paediatric cases, indicating that ILIL may be more common in children than previously appreciated. Childhood cases are rare, and familial occurrences are exceptional. We present a rare case of bilateral ILIL in a male child with a positive family history, emphasising its clinical and histopathological features.

CASE REPORT

A 5-year-old healthy male presented with bilateral white depressions over the lateral buttocks for 6 months [Figure 1]. The lesions were asymptomatic, insidious in onset and gradually increasing in size. There was no history of trauma, injections, systemic illness or infection.

Clinical examination revealed symmetrical, non-tender, well-demarcated, round depressed depigmented plaques (3 × 3 cm in size) over the bilateral gluteal region. No other body areas were affected. Routine haematological (complete blood count, erythrocyte sedimentation rate, liver and renal function) and biochemical investigations (fasting blood sugar, thyroid function and antinuclear antibodies) were normal.

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Figure 1: (a) Symmetrical, well-demarcated, depressed depigmented plaques over the gluteal region of the patient. Close-up of (b) Left and (c) Right side.



Figure 2: Regression of lesion with peripheral repigmentation at 4 weeks.

A maternal cousin, a 4-year-old female living in the same household, had also developed a similar depressed depigmented plaques unilaterally. However, her plaque showed spontaneous regression with repigmentation but was not clinically examined.

A skin biopsy was performed with differential diagnoses including lupus panniculitis, localized morphea, injection-site lipoatrophy, lichen sclerosus and atrophoderma of pasini and pierini. It revealed basket-weave keratosis, dermal fibrosis and absent inflammation. Adipocytes were not observed in the dermis or upper subcutis, consistent with fat lobule involution. Tacrolimus 0.1% ointment was prescribed, and the lesions stabilised with slight peripheral repigmentation at 4 weeks [Figure 2]. The clinical manifestation and the histopathology helped us in making a diagnosis of ILIL.

DISCUSSION

Localized lipoatrophy encompasses a spectrum of conditions resulting from trauma, injections or autoimmune diseases. Idiopathic lipoatrophy, however, remains a diagnosis of exclusion. Histopathological findings in ILIL typically show fat lobule involution, dermal fibrosis and epidermal atrophy without significant inflammation, as seen in this case.^[2,3]

ILIL predominantly affects females and presents unilaterally. Bilateral involvement in children is rare, with only a few reports in the literature. Sharma *et al.* noted an unexpected male predominance among children with ILIL in a series of 12 cases, contrasting earlier observations.^[2] Our case aligns with such findings and broadens the clinical spectrum of ILIL. Familial cases, such as our patient and his cousin, are exceedingly uncommon, raising questions about potential genetic or environmental factors.^[4] Chang *et al.* have reported such rare familial occurrences, suggesting a possible hereditary or shared environmental predisposition.^[5]

The pathogenesis of ILIL remains unclear. Various hypotheses suggest potential roles of microtrauma, localized hypoxia or subtle immune dysregulation leading to fat involution. Familial cases may indicate a genetic predisposition or shared environmental triggers, such as household exposure to a yet unidentified factor.^[4]

Histological patterns in ILIL are distinctive. The absence of significant inflammation helps distinguish ILIL from autoimmune or connective tissue disorders such as morphea or lupus panniculitis. Differentiation of ILIL from injection-induced lipoatrophy is crucial, as both conditions may appear histologically similar. However, a detailed history and the absence of prior injections or drug exposure support the diagnosis of ILIL.

Therapeutic options for ILIL remain empirical. Spontaneous resolution is often observed, with some cases requiring intervention during the progressive phase. Topical anti-inflammatory agents, including calcineurin inhibitors like tacrolimus, have shown efficacy in halting lesion progression and facilitating repigmentation, as observed in our case.^[6]

CONCLUSION

This case contributes to evidence that ILIL may have diverse presentations beyond the typical demographic of adolescent females. Bilateral and familial occurrences, though rare, highlight the need for clinicians to maintain a broad differential when evaluating cases of localized lipoatrophy. Further research is warranted to elucidate the pathogenesis and optimise therapeutic strategies for this rare entity.

Ethical approval: Institutional Review Board approval is not required.

Declaration of patient consent: The authors certify that they have obtained all appropriate patient consent forms. In the form, the patients have given their consent for their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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