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Case Report

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Langerhans cell histiocytosis in a four-month-old male infant

Abstract

Langerhans cell histiocytosis (LCH), a condition that mainly affects the skin and bones, is a rare disorder caused by the accumulation of histiocytes in different tissues. The resultant cutaneous lesions can be highly variable in appearance, potentially leading to misdiagnosis as other common dermatoses. In this report, we describe the case of a 4-month-old boy who presented with a generalized vesiculopustular itchy skin rash that had persisted for 3 months. In addition, the patient had a wide spectrum of cutaneous lesions, the pattern of which, to the best of our knowledge, has not been previously reported in the literature for LCH. On the basis of the typical clinical findings, the patient was ultimately diagnosed with LCH. This case highlights the importance of considering LCH in the differential diagnosis, even during the neonatal period, and of distinguishing this condition from other common neonatal dermatoses.

Keywords: dermatology, pediatric dermatosis, histiocyte, cutaneous lesion, langerhans cells

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Abbreviations: LCH, Langerhans cell histiocytosis

Introduction

Langerhans cell histiocytosis (LCH) is a rare disorder caused by the accumulation of Langerhans histiocytes in different tissues.[1] Although primarily affecting the skin and bones, other organs may be involved to a lesser extent. The estimated incidence is 1 to 2 cases per million per year in adults and 2 to 9 per million per year in children.[2] Although individuals of all ages and both sexes can be affected, the peak incidence occurs in children between 1 and 3 years, with a preponderance among white males. The extent of LCH can be divided into two major categories, single-system and multisystem. "Single-system" LCH (typically in older children and adults), involves a single organs/system, which, depending on the number of sites involved, can be further subdivided into unifocal and multifocal. "Multisystem" LCH (typically in children aged < 2 years), is characterized by the involvement of two or more organs, with or without involvement/dysfunction of "risk organs" (hematopoietic system, liver, and/or spleen).[2] In pediatric cases, the most frequently affected sites are bones (80%), followed by skin (40%) and the lymphatic ganglia.[3]

Case report

A 4-month-old boy presented with a generalized vesiculopustular itchy skin rash that had persisted for 3 months (Figure 1a - Figure 1d). In addition, the patient was characterized with a wide spectrum of cutaneous lesions, occurring concurrently in novel patterns (Figure 2a - Figure 2d). Moreover, the boy's body temperature was elevated, but not indicative of serious illness. Given the observation of generalized vesicular lesions on the palms, soles (Figure 2a), and scalp, the initial provisional diagnosis was varicella (Figure 1d). However, the relatively long duration of symptoms (3 months) rendered this diagnosis less likely. Given the typical light-colored pearly appearance of the umbilicated papules over the patient's face and trunk (Figure 1c, Figure 2b), molluscum contagiosum was also considered. Molluscum contagiosum was ruled out by the mosaic pattern of other cutaneous lesions, particularly the pink, purpuric papules on his lower abdomen and buttocks (Figure 1b, Figure 1c). The yellow-crusted lesions on the patient's scalp (Figure 2c, Figure 2d) and the macerated plaques on his groin and neck (Figure 1b) suggested cradle cap, their association with the other skin lesions made this less likely. Blueberry muffin nevus was included in the differential diagnosis, due to the purpuric papules on the lower abdomen and buttocks (Figure 1b, Figure 1c). On the basis of the typical clinical findings, we ultimately arrived at a diagnosis of Langerhans cell histiocytosis (LCH).

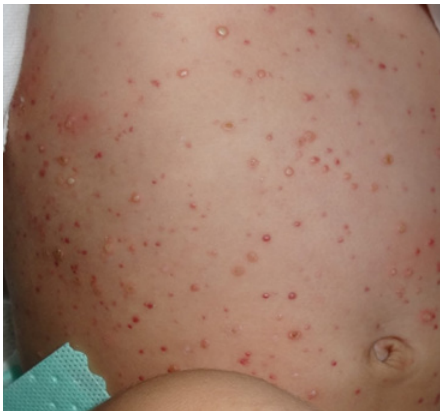

Figure 1a An exceedingly polymorphic spectrum of skin lesions, including papules, vesicles, pustules, crusts, purpuric papules, brown, and hypopigmented macules.

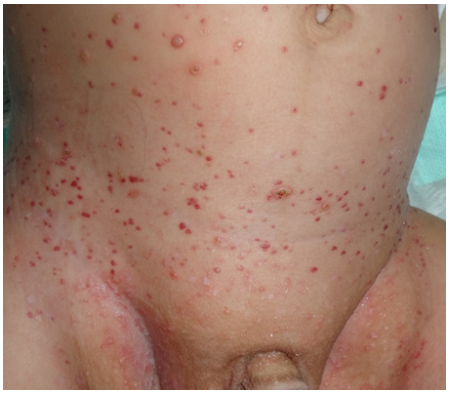

Figure 1b Scattered purpuric lesions on the patient's lower abdomen and groin, and macerated plaques over both inguinal folds.

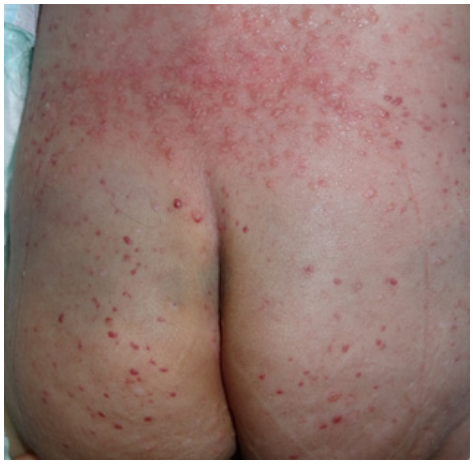

Figure 1c Koebner phenomenon presenting as clustered, linear, and skin-colored to red papules on the patient's waist area, some exhibiting central umbilication resembling *Molluscum contagiosum*. Red papules on both buttocks resemble mulberry nevi.

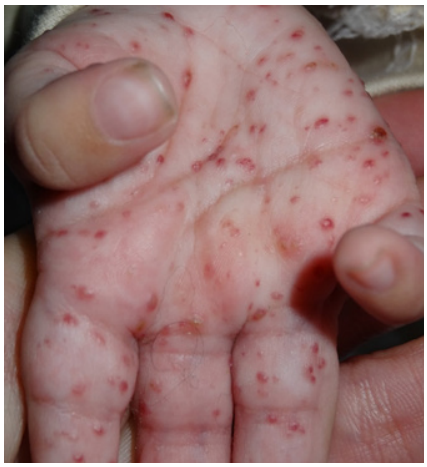

Figure 1d Palmar involvement, with scattered papules, vesicles, and pustules, ranging from skin-colored to dark red.

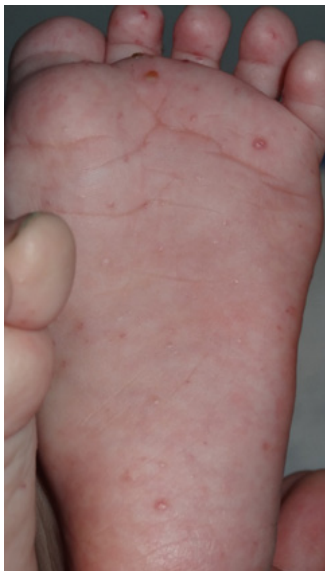

Figure 2a Plantar involvement, few scattered vesicles, pustules, and hypopigmented scars.

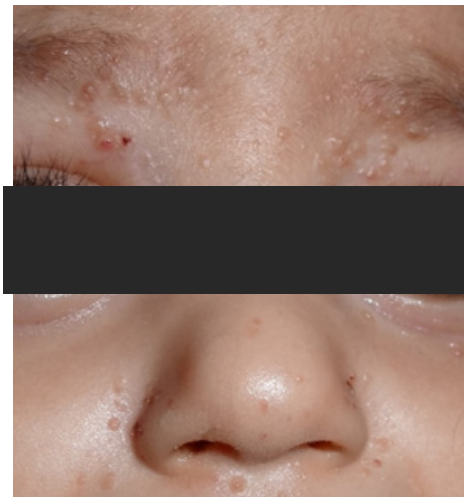

Figure 2b Involvement of the eyelids and nasolabial folds. Molluscum-like symmetrical, bilaterally periocular, and perinasal pearly umbilicated papules.

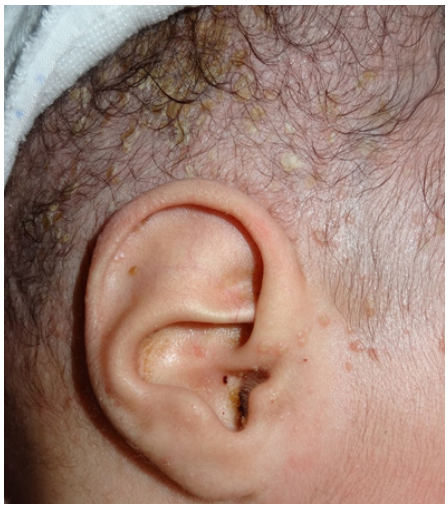

Figure 2c Involvement of the external ear and scalp.

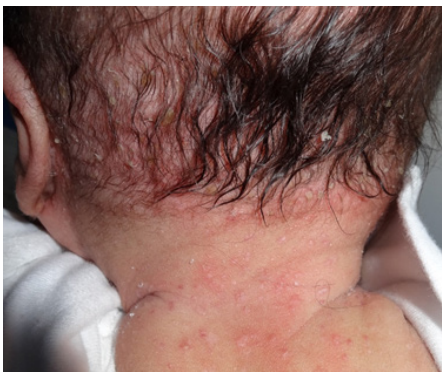

Figure 2d Seborrheic dermatitis-like lesions on the occipital and retroauricular areas.

Discussion

Skin involvement is observed in 40% of children and 20% of adults with LCH. Cutaneous LCH is characterized by a spectrum of lesions that can mimic several other types of skin disease.[2] The most commonly involved sites are the scalp, trunk, and intertriginous areas, with the face, palms, and soles being less commonly affected. The

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findings in the present case are similar to those reported by Johno et al., who described a varicelliform eruption.[1] To the best of our knowledge, involvement of the palms and soles has been reported only 10 times (Table 1).

Table 1 Reported LCH cases with Palmoplantar involvement

Publication Year	1st Author	Title	Journal	Country	Age & Sex	Number
1990	Herman LE	Congenital self-healing reticulohistiocytosis: A new entity in the differential diagnosis of neonatal papulovesicular eruptions	Arch Dermatol		1DF	1
2001	Stein SL	Langerhans cell histiocytosis presenting in the neonatal period: A retrospective case series	Arch Pediatr Adolesc Med	USA	6F, 13M	19
2010	Li Z	Two case report studies of Langerhans cell histiocytosis with an analysis of 918 patients of Langerhans cell histiocytosis in literatures published in China	Int J Dermatol	China	6MF, 2YM	2
2010	Battistella M	Neonatal and early infantile cutaneous langerhans cell histiocytosis: Comparison of self-regressive and non-self-regressive forms	Arch Dermatol	France	10M, 11F	21
2016	Morren MA	Diverse Cutaneous Presentations of Langerhans Cell Histiocytosis in Children: A Retrospective Cohort Study	Pediatr Blood Cancer	Belgium		32
2018	Kalpana S	Systemic Congenital Langerhans Cell Histiocytosis Masquerading as Diffuse Neonatal Hemangiomatosis	Indian Pediatr	India	3MF	1
2021	Lv X	Langerhans cell histiocytosis misdiagnosed as cow protein allergy: A case report	BMC Pediatr	China	2MM	1
2022	Han D	Multisystem Langerhans Cell Histiocytosis in Younger Infants First Presenting in Skin: A Case Series	J Pers Med	China	6,5MM, 4MF, 3.5YF, 3YF	4
2023	Utiyama TO	Langerhans cell histiocytosis: A rare case of the multisystemic form in an infant	An Bras Dermatol	Brazil	2MM	1
2024	Afonso C	Cutaneous Manifestations of Langerhans Cell Histiocytosis in Pediatric Age: A Case Report	Cureus	Portugal	8MF	1

Diffuse hemorrhagic vesiculopustular and umbilicated papulovesicles involving the trunk, palms, and soles were reported by Morren et al.[2] The involvement of the palms and soles with hemorrhagic papules and vesicles has been reported on five occasions, including the case of a 2-month-old Chinese infant described by Lv et al.[4] (Table 2).

Table 2 Reported LCH cases with hemorrhagic skin lesions

Publication Year	1st Author	Title	Journal	Country	Age & Sex	Number
2003	Inuzuka M	Congenital self-healing reticulohistiocytosis presenting with hemorrhagic bullae	J Am Acad Dermatol	Japan	1DM	1
2018	Kalpana S	Systemic Congenital Langerhans Cell Histiocytosis Masquerading as Diffuse Neonatal Hemangiomatosis	Indian Pediatr	India	3MF	1
2020	Widodo I	Case series of cutaneous Langerhans cell histiocytosis in Indonesian children: The clinicopathological spectrum	Dermatol Rep	Indonesia	4F, 6M	10
2021	Lv X	Langerhans cell histiocytosis misdiagnosed as cow protein allergy: A case report	BMC Pediatr	China	2MM	1
2023	Utiyama TO	Langerhans cell histiocytosis: A rare case of the multisystemic form in an infant	An Bras Dermatol	Brazil	2MM	1

In contrast, hypopigmented macules accompanied by vesiculopustules on the palms and soles have been reported 11 times, including by Battistella et al. (Table 3).[2,5,6,7]

Involvement of the eyelids has only been reported 10 times, including cases described by El Hindy et al., Ramzan et al., Huang et al., and Gupta et al., in which the patients had single, unilateral lesions on either the upper or lower eyelids (Table 4).[7–9]

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Table 3 Reported LCH cases with Hypopigmented lesions

Publication Year	1st Author	Title	Journal	Country	Age & Sex	Number
2010	Battistella M	Neonatal and early infantile cutaneous langerhans cell histiocytosis: Comparison of self-regressive and non-self-regressive forms	Arch Dermatol	France	10M, 11F	21
2010	Mehta B	Langerhans cell histiocytosis presenting as hypopigmented papules	Pediatr Dermatol	India	1.5YM	1
2014	Uaratanawong R	Congenital self-healing reticulohistiocytosis presented with multiple hypopigmented flat-topped papules: A case report and review of literatures	J Med Assoc Thai	Thailand	2MF	1
2016	Kassardjian M	Congenital selfhealing reticulohistiocytosis: An underreported entity	Cutis	USA	6MM	1
2018	Mori S	Cutaneous Langerhans cell histiocytosis presenting with hypopigmented lesions: Report of two cases and review of literature	Pediatr Dermatol	USA	5WM, 10MF	2
2018	Bishnoi A	Hypopigmented and acneiform lesions: An unusual initial presentation of adult-onset multisystem Langerhans cell histiocytosis	Indian J Dermatol Venereol Leprol	India	35YM, 21YM	2
2019	Wu X	Congenital self-healing reticulohistiocytosis with BRAF V600E mutation in an infant	Clin Exp Dermatol	China	5MF	1
2020	Dhar S	Langerhans cell histiocytosis in children: A retrospective case series of 126 cases	Pediatr Dermatol	India		126
2023	Utiyama TO	Langerhans cell histiocytosis: A rare case of the multisystemic form in an infant	An Bras Dermatol	Brazil	2MM	1
2024	Afonso C	Cutaneous Manifestations of Langerhans Cell Histiocytosis in Pediatric Age: A Case Report	Cureus	Portugal	8MF	1
2024	Xiao A	Hypopigmented Cutaneous Langerhans Cell Histiocytosis in a Hispanic Infant	Cutis	USA	7MM	1
2008	Feroze K	Langerhans cell histiocytosis presenting with hypopigmented macules	Indian J Dermatol Venereol Leprol	India	3YM	1

Table 4 Reported LCH cases with eyelid involvement

Publication Year	1st Author	Title	Journal	Country	Age & Sex	Number
1989	Tosaka Y	A case of localised histiocytosis X of the eyelid	Nippon Ganka Gakkai Zasshi	Japan	33YM	1
1992	Miller ML	Diffuse histiocytosis X involving the eyelid of a 65-year-old woman	Am J Ophthalmol	USA	65YF	1
1994	Weissgold DJ	Eosinophilic granuloma of the eyelid	Ophthalmic Plast Reconstr Surg	USA	15YF	1
1995	Daras C	Langerhans' cell histiocytosis of the eyelid	Br J Ophthalmol	South Africa	3YF	1
1998	Chikama T	Langerhans cell histiocytosis localized to the eyelid	Arch Ophthalmol	Japan	46YM	1
2009	Oono S	Langerhans cell histiocytosis limited to the eyelid margin	Jpn J Ophthalmol	Japan	52YM	1
2011	El Hindy N	Langerhans cell histiocytosis of the eyelid	J Paediatr Child Health		7YM	1
2012	Ramzan M	Eyelid nodule: A rare presentation of Langerhans cell histiocytosis	J Pediatr Hematol Oncol	India	5YM	1
2012	Huang JT	Langerhans cell histiocytosis mimicking molluscum contagiosum	J Am Acad Dermatol		2YF	1
2014	Gupta R	Langerhans cell disease of the eyelids masquerading as blepharochalasis	Pediatr Dermatol	India	4YF	1

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In the present case, we observed multiple, symmetrical bilateral lesions that were distributed on both the upper and lower eyelids, similar to the observations reported by Gupta et al.[7] Although involvement of the nasolabial folds has rarely been reported, the Koebner phenomenon, observed in the present case as a belt on the waist area, has previously been described by Akay et al. in a Turkish girl.[9]

In this case it is characterized by the clustering of lesions on the sacral area. In a case described by Dhar et al., it was assumed that the lesions were attributable to friction.[10,11] Additionally, involvement of the paranasal and preauricular areas, as well as the scalp was reported by Akay et al. in a Turkish girl.[10] Minute lichen nitidus-like papules have been reported by both Dhar et al., and Lozano Masdemont et al.[11,12] In the present case, we detected two minute hypopigmented, lichen nitidus-like papules on the patient's right hand. Furthermore, molluscum-like pearly umbilicated papules have been reported by Huang et al.,[12] in a 2-year-old girl who had pink friable papules on the margin of her eyelid.[4] (Table 5)

Table 5 Reported cases of LCH presenting with Molluscum like lesions

Publication Year	1st Author	Title	Journal	Country	Age & Sex	Number
2010	Mehta B	Langerhans cell histiocytosis presenting as hypopigmented papules	Pediatr Dermatol	India	1.5YM	1
2011	Edwards AN	Molluscumlike papules in a 4-month-old boy--quiz case: Langerhans cell histiocytosis (LCH)–congenital self-healing reticulohistiocytosis	Arch Dermatol	USA	4MM	1
2012	Huang JT	Langerhans cell histiocytosis mimicking molluscum contagiosum	J Am Acad Dermatol	USA	2YF	1
2015	Hatter AD	Langerhans Cell Hyperplasia from Molluscum Contagiosum	Am J Dermatopathol		14YM	1
2017	Karpman MS	Molluscum Contagiosum-Like Presentation of Langerhans Cell Histiocytosis: A Case and Review	Pediatr Dermatol	Canada	5.5MF	1
2018	Fernández Armenteros JM	Langerhans cell histiocytosis mimicking molluscum contagiosum: A case series	Pediatr Blood Cancer	Spain	5MF, 13MM, 24YF	3
2022	Sanke S	Molluscum-Like Lesions in a Child with T Cell Lymphoma	Indian J Dermatol	India	3YF	1
2022	Katayama S	An Uncommon Pediatric Rash: Langerhans Cell Histiocytosis	J Pediatr	Japan	5MF	1
2019	Campos-Filho MM	Molluscum contagiosum-like presentation of Langerhans cell histiocytosis: A case report	WDC	Brazil	3YM	1

This contrasts with the present case, in which more typical, skin-colored umbilicated lesions were symmetrically and bilaterally distributed on the patient's periocular and perinasal areas. These are broadly consistent with the polymorphic cutaneous lesions described by Katayama et al.[13]

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None.

Conflict of interests

The authors declare there is no conflict of interest.

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