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Benign Cephalic Histiocytosis

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Ayala F, Balato N, Iandoli R, Donofrio V, De Rosa G. Benign cephalic histiocytosis. Acta Derm Venereol (Stockh) 1988; 68: 264–266.

A case of benign cephalic histiocytosis in a 18-month-old male child is reported. Characteristic distribution of the maculo-papular lesions on the cheeks, forehead and earlobes, in addition to light and electron microscopical findings permitted the diagnosis of this rare disorder. Ultrastructurally, worm-like and comma-shaped bodies were seen in the cytoplasm of the histiocytes infiltrating the upper dermis. *Key word: Pediatric dermatology.*
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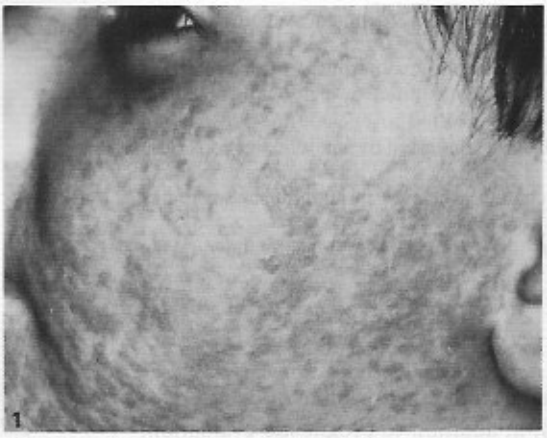
Benign cephalic histiocytosis (BCH) was first described by Gianotti et al. (1) in 1971 as "Juvenile histiocytosis with intracytoplasmic worm-like particles". Subsequently, other authors reported several cases of this rare entity (2–6), that is actually known as BCH



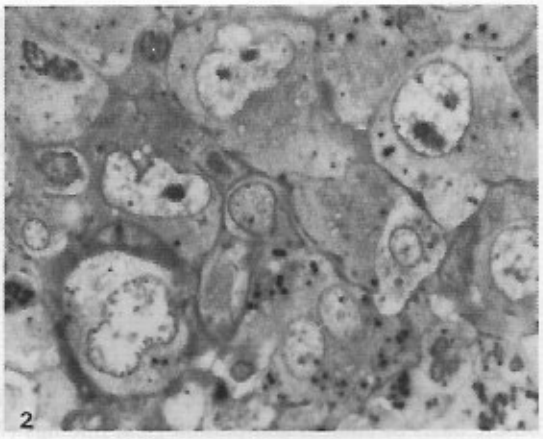


Fig. 1. Papular eruption involving whole face.

Fig. 2. Semithin section showing the histiocytic nature of most cells in the dermal infiltrate.

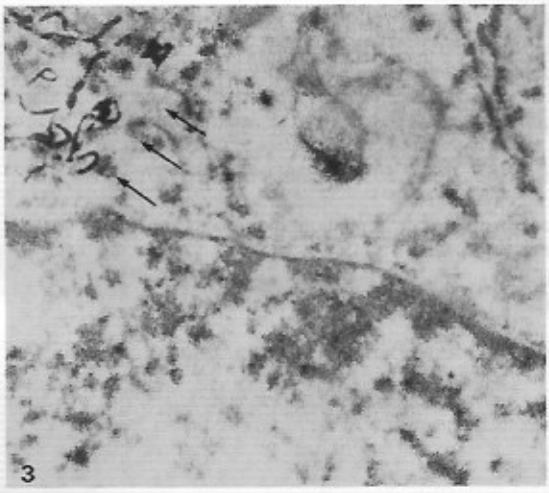
because of its benign course and peculiar distribution of the lesions. As a rule, spontaneous regression occurs in all cases of this cutaneous, non-X, non lipid histiocytic proliferation (2).

We recently had the opportunity to study an additional case, that is herein described.

CASE REPORT

An 18-month-old male child was referred to the Dermatological Clinic for a yellowish, asymptomatic papular eruption involving the cheeks, forehead and earlobes (Fig. 1), whose onset dated about one year before. The family history was negative for skin diseases.

Clinical examination revealed numerous discrete papular lesions, sometimes arranged in plaques. The individual lesions varied in size from 1 to 3 mm; a reticulated pattern was present in some areas. There was no mucous membrane involvement. No other pathological findings were observed on



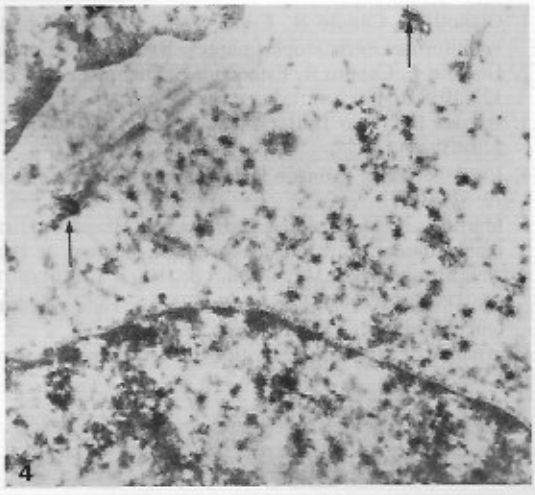


Fig. 3. Histiocyte containing worm-like bodies (*arrows*).

Fig. 4. Arrows indicate comma-, ring-shaped and worm-like bodies in a histiocyte.

physical examination. Routine laboratory tests were within normal limits; roentgenographic investigations did not show any remarkable changes.

Biopsy of a papular lesion revealed a normal epidermis. The superficial dermis was characterized by a both sparse and circumscribed cellular infiltrate, mainly constituted by cells with abundant cytoplasm and round or spindle nuclei. Toluidine-blue stain permitted to exclude the presence of mast-cells in the infiltrate. The histiocytic nature of the cells was confirmed by semithin sections (Fig. 2).

Ultrastructural examination showed the presence of "coated vesicles", in addition to worm-like bodies. Comma, S, arc, ring and rod-shaped bodies were also observed (Figs. 3, 4).

No treatment was started, due to the benign nature of the disorder. At one-year follow-up the lesions were slightly improved.

DISCUSSION

Cutaneous histiocytoses in children may be classified as either self-healing or progressive on the basis of clinical and pathological findings (7), in addition to prognostic evaluation.

BCH is one of the self-healing clinical forms; it is characterized by a distinct papular eruption predominantly involving the head, neck and shoulders, that generally begins in the second half-year of life. In rare cases, the occurrence of lesions on the upper arms, trunk and lower extremities has been reported.

Histologically, light microscopy reveals no peculiar findings, unlike other forms of cutaneous histiocytosis; this seems to be the explanation for the misdiagnosis of some BCH cases as atypical forms of juvenile xanthogranuloma.

Under the electron microscope, the most characteristic feature of BCH is the presence of the intracytoplasmic worm-like or comma-shaped bodies, whose origin and function have not yet been fully elucidated. A particular modification of the endoplasmic reticulum has recently been proposed (7), in order to interpret the origin of these peculiar structures. The comma-shaped bodies were originally considered the peculiar ultrastructural hallmark of BCH; nevertheless, they have been subsequently seen in lymphoma as well as in some other forms of histiocytosis (7). On the other hand, the latter can be excluded on the basis of different clinical and evolutive patterns.

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