RARE C-GROUP HISTIOCYTOSES ASSOCIATED WITH MYELOID NEOPLASMS

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INTRODUCTION

Histiocytoses are rare proliferative disorders of mononuclear-phagocyte system cells arising, as far as we know, from bone marrow progenitors. The association between histiocytic disorders and myeloid neoplasms (MN) is well-known to the scientific community [1, 2]. Nevertheless, the biological features of cases are still poorly investigated and there are no specific predictive and prognostic markers as well as no standard therapeutical approach. Most report describe cases of Langerhans Cell Histiocytosis, Xanthogranuloma or Erdheim-Chester Disease (the two latter characterized by foamy-cell infiltrates) associated with MN. Otherwise, just a few papers focus on more rare histiocytic disorders. Therefore, we investigate the frequency and characteristics of the association between rare non-foamy C-group Non-Langerhans Cell Histiocytoses (NF-C-NLCH) and MN from the scientific and medical literature.

METHODS

We performed a literature search of published papers, with last query in May 2018, using four retrieval systems: PubMed, Scopus, Web of Science and Open Grey. The search included each combination of MN related terms and names of the NF-C-NLCH (Generalized Eruptive Histiocytosis, Reticulohistiocytosis and Benign Cephalic Histiocytosis). The obtained papers (526) were deduplicated via Mendeley (246), selected by title and abstract and finally filtered according to clinical-pathological descriptions. We gathered up 12 case reports: 5 cases diagnosed with Generalized Eruptive Histiocytosis and 7 with Reticulohistiocytosis (both multicentric and non-multicentric). No cases of Benign Cephalic Histiocytosis were found. Just one patient presents multisystem disease.

RESULTS

Most patients were male, with a median age of 59 years. Most patients present with a generalized papular skin eruption (Figure1), displaying an epithelioid histiocytic infiltrate CD68+, CD1a- with ground-glass cytoplasm (Figure 2). The associated myeloid neoplasms was Acute Myeloid Leukemia in 4 cases (Figure 3), Chronic Myelomonocytic Leukemia in 3 cases, Myelodysplastic syndrome with ring sideroblasts in 2 cases and Myeloproliferative neoplasms in 3 cases. A clonal relation between histiocytosis and MN was proved in two reports [12, 14].

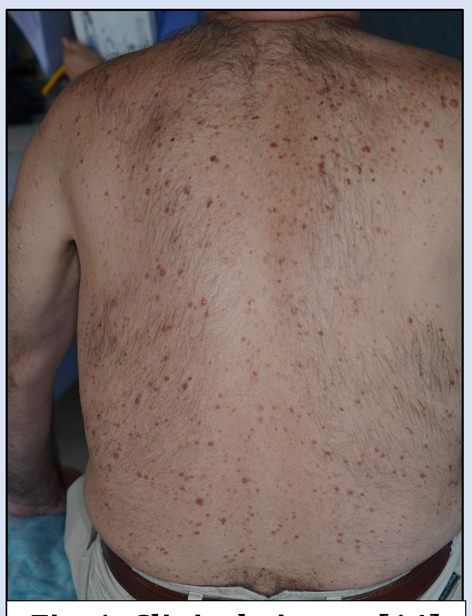

Fig. 1: Clinical picture [14]

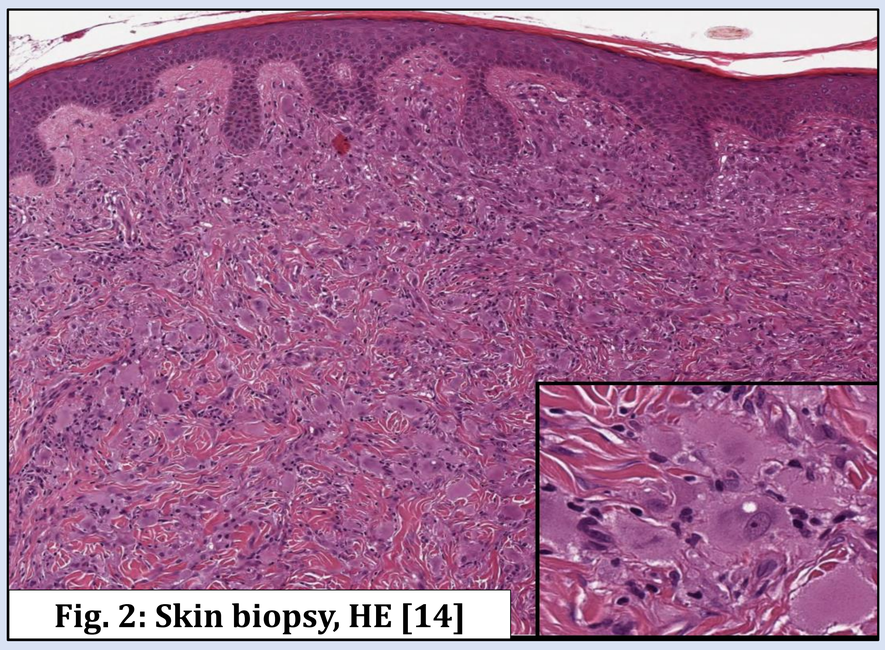

Fig. 2: Skin biopsy, HE [14]

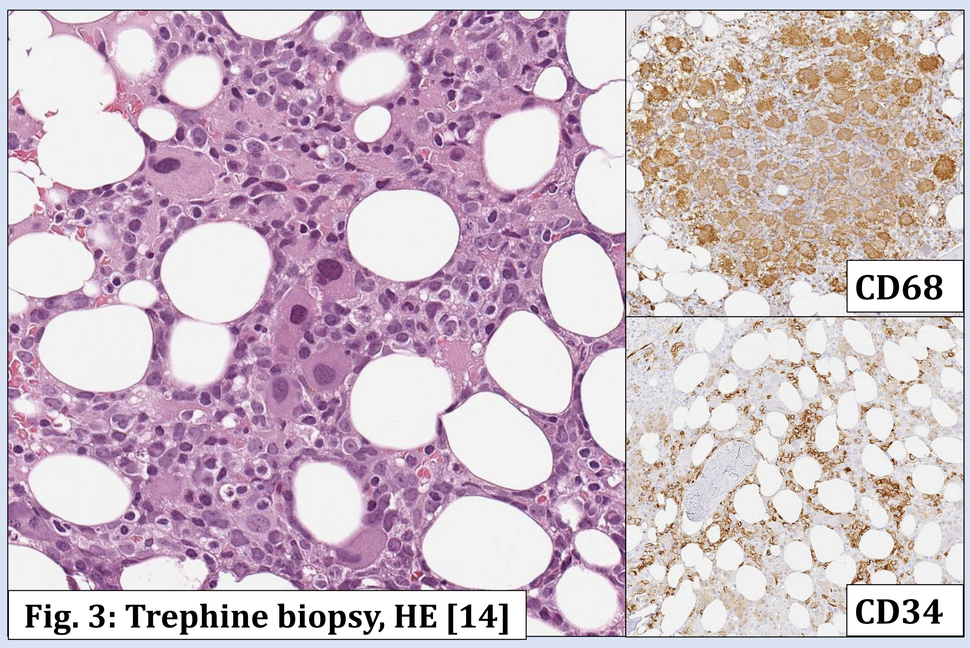

Fig. 3: Trephine biopsy, HE [14]

ref.	NLCH	MN	Age	Sex	Clonal relation	Therapy	Follow up
[8]	GEH	AMoL	59	M	n.d.	Chemotherapy, HSCT	Alive
[10]	GEH	MDS	60	F	no	n.d.	n.d.
[11]	GEH	CMML	80	M	n.d.	none	Alive
[12]	GEH	CMML	84	M	-Y	EPO	DOD
[13]	GEH	CEoL	20	M	no	Imatinib	AWD
[9]	RHC	AML	42	M	n.d.	n.d.	n.d.
[14]	RHC	AML + SM	59	M	der(1;9)(q10;p10)	Chemotherapy, HSCT	Alive
[4]	RHC	AML	65	M	n.d.	Chemotherapy	n.d.
[3]	RHC	CMML	57	M	n.d.	n.d.	n.d.
[5]	RHC	PMF	22	M	n.d.	n.d.	n.d.
[6]	RHC-M	MDS	59	M	n.d.	Chemotherapy	Alive CR
[7]	RHC-M	PMF	57	M	no	Chemotherapy, Radiotherapy	DOD

CONCLUSIONS

Despite their exceeding rarity, NF-C-NLCH result often associated and sometimes clonally related to MN. For some instance, NF-C-NLCH skin eruption may also be expression of a systemic myeloid dyscrasia more than of a simple histiocytic dermatosis. Therefore, a complete hematological evaluation is mandatory for all histiocytosis patients, even in single-system conditions and bone marrow biopsy highly recommended at the onset of hematological abnormalities.

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