



Spontaneous Regression of Atypical Fibroxanthoma of the Hand

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Authors' contributions

This work was carried out in collaboration among all authors. Author MAS designed the study and approved of the final paper. Author SB wrote the first draft of the manuscript and managed the literature search. Author RM verified and approved of the final paper. All authors read and approved the final manuscript.

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Case study

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ABSTRACT

Atypical fibroxanthoma is a rare fibrohistiocytic tumour with intermediate malignancy that generally affects photoexposed areas of the elderly. The diagnosis is anatomopathological and the management is surgical.

We report the case of a spontaneous regression of an atypical fibroxanthoma of the hand. This phenomenon is a very rare situation related to a probable immunological response.

Keywords: *Atypical fibroxanthoma; biopsy; regression.*

1. INTRODUCTION

Atypical fibroxanthoma is a rare fibrohistiocytic tumour with intermediate malignancy [1] that generally affects photoexposed areas of the elderly. The diagnosis is anatomopathological

and the management is surgical. Spontaneous regression is an exceptional phenomenon rarely reported in literature and which physiopathological and immunological mechanisms are still unknown. We report a case

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of a spontaneous regression of this tumour after percutaneous biopsy.

The aim of this work is to study through this case the clinical, anatomopathological, therapeutic and evolutionary aspects of this tumor. A review of literature is present to discuss causes of this regression.

2. PRESENTATION OF CASE

A 78 year-old woman, with a pathological history of diabetes and hypertension, came to our consultation for an ulcerobudding lesion of the dorsal side of the left hand evolving for 2 weeks without a notion of trauma or inoculation by a foreign body.

On clinical examination, we have objectified a painless rumpled ulcerobudding lesion of the dorsal side of the left hand characterized by : a diameter of 5 cm, erythematous borders, without pearl-shape-edge. A slightly moderate edema was associated. However there was no limitation of mobility of fingers. There were no major inflammatory signs. Faced with this aspect, the diagnosis of squamous cell carcinoma was evoked and a biopsy of the lesion was done. The patient was reviewed two weeks later with a

near total regression of the lesion. Initial histological examination revealed parakeratotic hyperkeratosis with architectural cellular atypia, thickened epidemic crests, cytonuclear atypia of the basal layer, globular cells with abundant cytoplasm difficult to identify by standard staining. A complementary immunohistochemical study found overexpression of CD68 in dermal cells with negativity of other markers (citokeratin and melanic marker HMB45) consistent with the diagnosis of an atypical fibroxanthoma.

The follow-up shows no sign of recurrence at 8 months after the initial biopsy. However we note persistence of a slightly hyperpigmented scar, which did the patient no discomfort.

3. DISCUSSION

Atypical fibroxanthoma (FXA) is a fibrohistiocytic tumor of intermediate malignancy, described for the first time by Helwig in 1961 [2]. Its incidence is low, in the order of 0.24% among all skin tumours [1]. FXA preferentially sits in photoexposed regions such as the head and neck in the elderly population. Location at the limb level is infrequent and accounts for about 20% of cases.



Fig. 1. Initial aspect of the tumor : Ulcerobudding lesion with erythematous borders of the dorsal side of the hand



Fig. 2. Spontaneous regression of the lesion 2 weeks after biopsy, note the presence of scaly skin with little irritation



Fig. 3. Evolution 8 months after biopsy. Note the persistence of hyperpigmented skin with no sign of recurrence

FXA comes across as a single reddish or brownish lump, painless, non-pruritic, classically less than 2 cm, localized at the photoexposed areas.

Histologically, this exophytic tumour is formed by fusiform, epithelial and multinucleated giant cells. It is strictly limited to the dermis and classified among tumours with uncertain differentiation according to the 2013 WHO classification [3]. However, the epidermis may be thin and occasionally ulcerated. Anatomico-pathological aggressiveness criteria include invasion of vessels or subcutaneous fat as well as tumor

necrosis. Diagnosis may be difficult [4] because of the similarity of FXA with other tumours such as squamous cell carcinoma, desmoplastic melanoma and undifferentiated pleomorphic sarcoma. For this reason, an additional immunohistochemical study is needed to eliminate these diagnoses.

The locoregional extension may be significant and thus require wide cross-sections [5]. Recurrences of FXA are infrequent after surgical excision, and are of the order of 8% [6]. Furthermore, the metastatic potential is low.

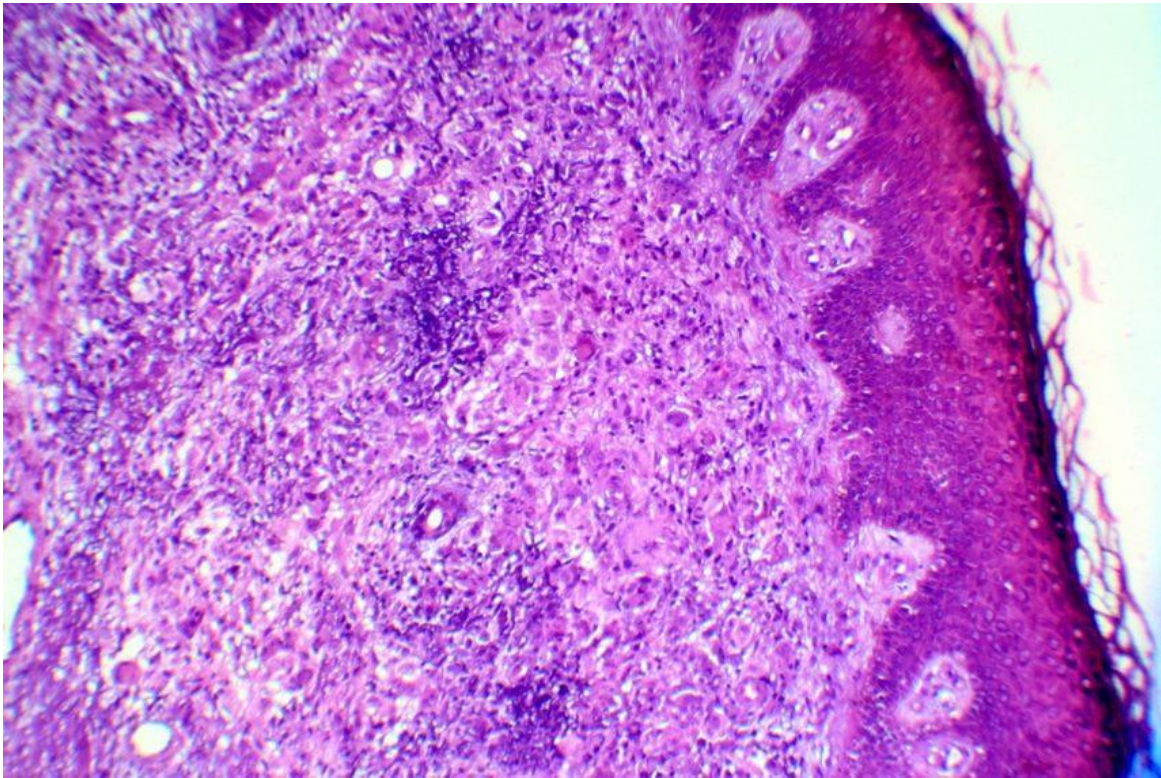


Fig. 4. Microscopic examination of the tumor: dermic proliferation of pleomorphic cells, architectural and cytonuclear atypia (magnification factor 40X)

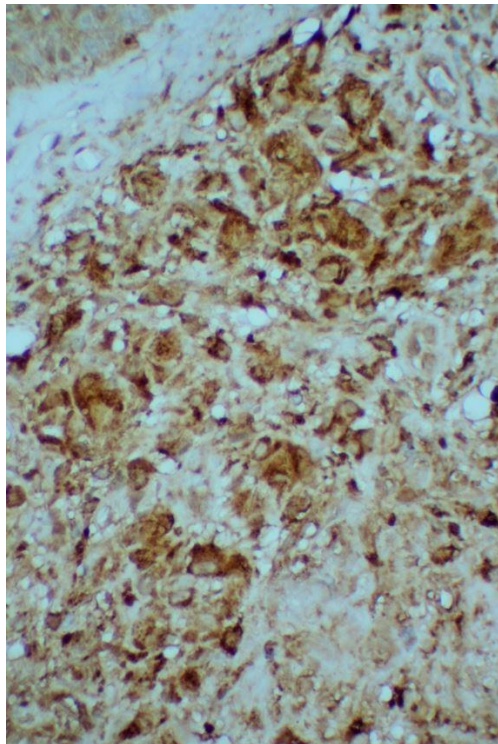


Fig. 5. Immunohistological examination : overexpression of antibodies anti-CD68 (magnification factor 100X)

Spontaneous regression is an exceptional phenomenon and only a few cases have been reported in literature [7,8]: The team at the Boulogne-sur-mer reported the case of an 81-year-old male with retroauricular fibroxanthoma receiving initial surgical removal with early local recurrence at 3 weeks post-operative which spontaneously regressed within 6 weeks [7]. A possible immune response is suspected to be at the origin of this evolution, as is the case with Merkel tumor for which biopsy could be the cause of a systemic leakage of few tumoral cells which activates an immune response leading to tumor apoptosis . Histologically, partial regressions could be noted and are characterized by the appearance of lamellar sclerosis and a predominant cheloidal aspect [9].

Studies of molecular immunohistochemistry and biology are probably necessary to try to shed the light on the mechanisms that allow the spontaneous disappearance of FXA.

4. CONCLUSION

Atypical fibroxanthoma is a tumor with intermediate malignancy and rare metastatic potential. Its spontaneous regression is an

exceptional phenomenon related to a probable immune response. It is recommended to re-examine the patient at regular intervals of 6 months, looking for recurrence, metastasis or development of other skin tumours.

CONSENT

As per international standard or university standard, patients' written consent has been collected and preserved by the author(s).

ETHICAL APPROVAL

As per international standard or university standard written ethical approval has been collected and preserved by the author(s).

COMPETING INTERESTS

Authors have declared that no competing interests exist.

REFERENCES

1. Iorizzo III, LUCIANO J, et Brown, Marc D. Atypical fibroxanthoma: A review of the literature. *Dermatologic surgery*. 2011;37(2):146-157.
2. Helwig EB. Atypical fibroxanthoma. *Tex State J Med*. 1963;59:664-667.
3. Karanian, Marie et COINDRE, Jean-Michel. Quatrième édition de la classification OMS des tumeurs des tissus mous. In : *Annales de Pathologie*. Elsevier Masson. 2015 ;71-85.
4. High AS, Hume WJ, et Dyson D. Atypical fibroxanthoma of oral mucosa: a variant of malignant fibrous histiocytoma. *British Journal of Oral and Maxillofacial Surgery*. 1990 ;28(4):268-271.
5. Andrawes, Peter Hill, David A, Marilyn NG, et al. Malignant Fibrous Histiocytoma of the Scalp with Skull Invasion: A Rare and Aggressive Presentation. *Cureus*. 2020;12(4).
6. Koch, Michael Freundl, Anne J, Agaimy Abbas, et al. Atypical fibroxanthoma-histological diagnosis, immunohistochemical markers and concepts of therapy. *Anticancer Research*. 2015;35(11):5717-5735.
7. Bagazgoitia L, Morais P, Carrillo R, Jaén P. Complete regression of an atypical fibroxanthoma. *European Journal of Dermatology*. 2012;22(1):157-158.
8. Capelle C, Jacobsoone A, Le Moing A, et al. Une surprenante régression de fibroxanthome atypique. In : *Annales de Dermatologie et de Vénérologie*. Elsevier Masson. 2017 ;S162.
9. Stefanato CM, Robson A, Calonje JE. The histopathologic spectrum of regression in atypical fibroxanthoma. *Journal of Cutaneous Pathology*. 2010;37(3):310-315.

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