Received: 29 April 2020 | Revised: 25 June 2020 | Accepted: 10 July 2020
DOI: 10.1111/dth.14012

DERMATOLOGIC THERAPY WILEY

LETTER

Infantile digital fibromatosis successfully treated with topical imiquimod 5% combined with methylprednisolone aceponate 0.1%

Dear Editor,

Infantile digital fibromatosis (IDF) is a rare, benign fibromatous tumor of childhood.[1,2] While mostly asymptomatic and spontaneous resolution is a rule, treatment may be demanded in IDF cases due to complications.[1] Treatment is usually challenging due to possible complications and relapsing nature of tumor after surgery.[1] Reports about other options are few such as intralesional steroid injections (ILS), cryotherapy, intralesional 5-fluorouracil injections.[3-6] In this paper we report a case with IDF treated with topical imiquimod and methylprednisolone aceponate 0.1%.

One-year-old boy was referred to us with three pink, firm nodules on dorsal, lateral, and medial aspects of right fifth finger with sizes of 1.5 cm × 1.5 cm, 0.5 × 0.5 cm, 1.5 × 0.8 cm, respectively (Figure 1A). His parents told that nodules began from dorsal aspect when he was 1 month old and spread lateral and medial aspects of his finger in 2 months. There was no trauma history and his past medical and family history were totally normal. Histopathological findings consisted with dermal hyalinized tumor and whorl-like arranged fibroblast like spindle cells with eosinophilic inclusion bodies which were suggestive for IDF (Figures 2 and 3). Due to high concerns of parents and erosion in distal phalanx, we applied ILS treatment with 20 mg/mL triamcinolone acetonide, but lesions did not respond, and new lesion appeared on right fourth finger. Therapy was switched to imiquimod 5% cream 3 days per week with occlusion and methylprednisolone aceponate 0.1% ointment on other days. At week of eight lesions started to resolve and at week of 22, lesions were markedly faded and flattened with complete healing of lesion on the fourth digit. We ceased treatment at that examination. After 6 weeks from treatment cessation other lesions were almost totally healed (Figure 1B). No side effect was observed during treatment.

IDF is a fibromatous tumor characterized by growing, erythematous, or skin colored nodules primarily affecting one or more digits, hands or foots.[1] Tumor spontaneously regresses in 6 months to 5 years.[4] Wait and watch approach is suitable in many cases but, tumor may leads to complications such as joint movement limitation, deformities, cosmetical, and functional disturbances.[3] Excision of lesions may be preferred but conservative surgeries have high recurrence rates, on the other hand wide excisions may lead to deformities.[1] ILS was reported as beneficial and well-tolerated, but still it can be traumatic and also effectiveness is questionable.[3] In our case we did not achieve good response with ILS furthermore new lesions appeared on adjacent finger. Other reported effective therapies are intralesional 5-fluorouracil and cryotherapy which are also traumatic and only one case report for each exist.[5,6] After failure of ILS treatment we tried imiquimod concomitant with topical corticosteroid due to their wide range of use in dermatology and successful results exist in Keloid, another fibroblastic tumor.[7,8] Imiquimod was also tried previously in IDF by Failla et al. but they did not observe successful result.[9] We combined topical methylprednisolone aceponate with imiquimod and achieved almost complete healing that occurred more rapidly than expected in watchful approach.[4] Imiquimod induces interferon-α which can downregulate keloidal collagen synthesis.[10] Topical corticosteroids also have

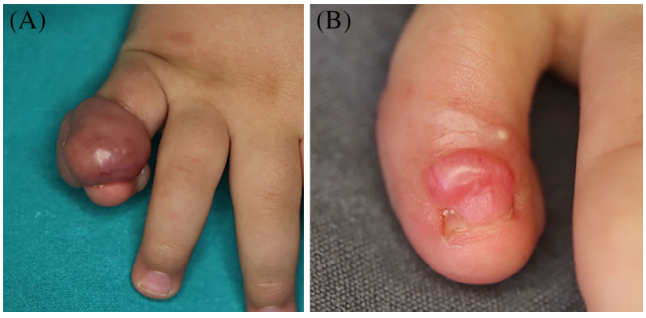

FIGURE 1 A, IDF lesions on fifth digit of right hand. IDF, infantile digital fibromatosis. B, Almost totally flattened and faded lesions at week of 28

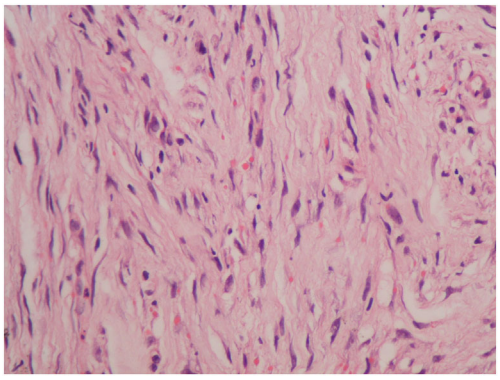

FIGURE 2 Hyalinized tumor with whorl-like arranged fibroblast like spindle cells with Hematoxylin and Eosin stain

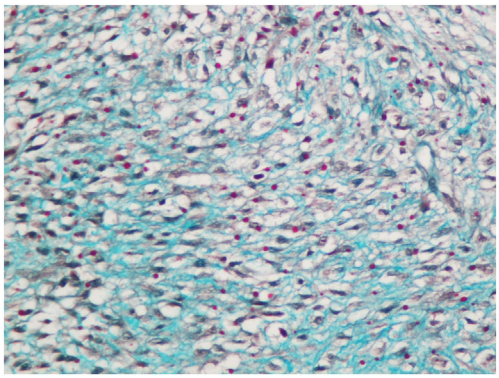

FIGURE 3 Eosinophilic intracytoplasmic inclusion bodies in spindle cells with Masson Trichrome stain

inhibitory effects on fibroblasts.[11] Therefore, these may improve IDF by altering myofibroblast biology similarly. Usually watchful approach is the best option for these cases but, if intervention is demanded, it seems that combined therapy of imiquimod and topical corticosteroids are relatively safe and worth to try prior to ILS and surgery in IDF.

CONFLICT OF INTEREST

Authors have no conflict of interest to declare.

AUTHOR CONTRIBUTIONS

Ayşe Serap Karadağ: Author is primary physician of concerning patient, supervisor of process. Author created the idea, designed the process, analyzed outcome, reviewed the article. Mahmut Can Koska: Author took part in patient care, deciding treatment, follow-up, photography, data collection, literature review, writing and preparation of manuscript. Filiz Cebeci: Author took part in diagnosis, deciding treatment, creation of idea and reviewed the article. Bilge Bilgiç: Author took part in diagnosis and investigation of pathologic materials, reviewed the article. Ali Burak Bostan: Author took part in diagnosis, deciding treatment, reviewed the article. Korhan Özkan: Author took part in diagnosis, deciding treatment, reviewed the article.

INFORMED CONSENT FORM

Consent form was signed by patient's parents.

Ayşe Serap Karadağ[1]
Mahmut Can Koska[1]
Filiz Cebeci Kahraman[1]
Bilge Bilgiç[2]
Ali Burak Bostan[3]
Korhan Özkan[3]

[1]*Department of Dermatology and Venereology, Istanbul Medeniyet University, School of Medicine, Göztepe Education and Research Hospital, Istanbul, Turkey*
[2]*Department of Pathology, Istanbul University, Istanbul Medical Faculty, Istanbul, Turkey*
[3]*Department of Orthopedics and Traumatology, Istanbul Medeniyet University, School of Medicine, Göztepe Education and Research Hospital, Istanbul, Turkey*

Correspondence
Mahmut Can Koska, İstanbul Medeniyet Üniversitesi Göztepe Eğitim ve Araştırma Hastanesi, Dermatoloji Anabilim Dalı, Eğitim Mah., Dr. Erkin Cad. 34722 Kadıkoy/Istanbul, Turkey.
Email: cankoska90@gmail.com

ORCID

Ayşe Serap Karadağ https://orcid.org/0000-0003-4333-8274
Mahmut Can Koska https://orcid.org/0000-0002-4564-0322
Filiz Cebeci Kahraman https://orcid.org/0000-0002-9109-3892

REFERENCES

1. Laskin WB, Miettinen M, Fetsch JF. Infantile digital fibroma/fibromatosis: a clinicopathologic and immunohistochemical study of 69 tumors from 57 patients with long-term follow-up. *Am J Surg Pathol*. 2009;33(1):1-13.
2. Coffin CM, Dehner LP. Fibroblastic-myofibroblastic tumors in children and adolescents: a clinicopathologic study of 108 examples in 103 patients. *Pediatr Pathol*. 1991;11(4):569-588.
3. Holmes WJ, Mishra A, McArthur P. Intra-lesional steroid for the management of symptomatic infantile digital fibromatosis. *J Plast Reconstr Aesthet Surg*. 2011;64(5):632-637.
4. Eypper EH, Lee JC, Tarasen AJ, Weinberg MH, Adetayo OA. An algorithmic approach to the Management of Infantile Digital Fibromatosis: review of literature and a case report. *Eplasty*. 2018;18:e19.
5. Oh CK, Son HS, Kwon YW, Jang HS, Kwon KS. Intralesional fluorouracil injection in infantile digital fibromatosis. *Arch Dermatol*. 2005;141(5):549-550.
6. Kramer A, Har-Shai Y, Metanes I, Harel H, Wollstein R. The use of cryotherapy to treat infantile digital fibromatosis with a functional deficit: a case report. *J Hand Surg Asian Pac Vol*. 2018;23(2):278-281.
7. Hanna E, Abadi R, Abbas O. Imiquimod in dermatology: an overview. *Int J Dermatol*. 2016;55(8):831-844.
8. Giannotti B, Pimpinelli N. Topical corticosteroids. Which drug and when? *Drugs*. 1992;44(1):65-71.
9. Failla V, Wauters O, Nikkels-Tassoudji N, Carlier A, Andre J, Nikkels AF. Congenital infantile digital fibromatosis: a case report and review of the literature. *Rare Tumors*. 2009;1(2):e47.
10. Berman B, Kaufman J. Pilot study of the effect of postoperative imiquimod 5% cream on the recurrence rate of excised keloids. *J Am Acad Dermatol*. 2002;47(4 Suppl):S209-S211.
11. Priestley GC, Brown JC. Effects of corticosteroids on the proliferation of normal and abnormal human connective tissue cells. *Br J Dermatol*. 1980;102(1):35-41.