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# Retrospective Analysis of the Cases With Terra Firma-Forme Dermatosis: An Entity Related With Allergic Diseases?

Terra Firma Forme Dermatozlu Olguların Retrospektif Olarak Degerlendirilmesi Allerjik Hastalıklarla İlgili Yeni Bir Antite mi?

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#### **ABSTRACT**

**Introduction:** Terra Firma-Forme Dermatosis (TFFD) is a little-known dermatosis. The etiology is not fully known. We aimed to investigate the clinical characteristics of TFFD, accompanying diseases, and a possible connection to allergic diseases.

Material and Method: A total of 869 pediatric patients were examined within the study period. Forty-one patients (2.19%) diagnosed with TFFD were included in the study. Age, gender, medical history, family history of the patients, anatomic localizations of the lesions, time that the complaints were noticed, complaints on admission, accompanying diagnoses, laboratory tests, and total IgE levels were evaluated.

**Results:** Truncal involvement was the most observed area in patients. At least one allergic disease was detected in 29 (70.7%) patients. The difference between the allergy ratio in the patients who were admitted to the outpatient clinic (0.107) and the patients with TFFD (0.707) was found statistically significant (t-value=8,345, P = 0.000). Total IgE levels were found to be high in 15 (55.5%) of 27 (65.85%) patients in whom the total IgE level was examined. In March-August, when allergic patients applied more frequently to the clinic, also cases with TFFD were seen more frequently.

**Conclusion:** Our study suggests that TFFD may be a finding associated with allergic diseases, considering the seasonal distribution of the disease, concurrence with allergic diseases, and high IgE levels.

**Key Words:** terra firma-forme Dermatosis, allergy, pediatric, IgE

#### Introduction

Terra firma-forme dermatosis (TFFD) was first defined by Duncan in 1987 (1). It is characterized

#### ÖZET

Amaç: Terra Firma-Forme Dermatozu (TFFD) az bilinen bir dermatozdur. Etiyolojisi tam olarak bilinmemektedir. TFFD'nun klinik özelliklerini, eşlik eden hastalıkları ve alerjik hastalıklarla olası bir bağlantıyı araştırmayı amaçladık.

Gereç ve Yöntem: Çalışma sırasında toplam 869 pediatrik hasta muayene edildi. TFFD tanısı konmuş 41 hasta (% 2.19) çalışmaya dahil edildi. Hastaların yaşı, cinsiyeti, tibbi öyküsü, aile öyküsü, lezyonların anatomik lokalizasyonları, fark edilme zamanı, başvuru şikayetleri, eşlik eden tanılar, laboratuar testleri ve total IgE düzeyleri değerlendirildi.

**Bulgular:** Trunkal tutulum hastalarda en çok gözlenen alandı. 29 hastada (% 70.7) en az bir allerjik hastalık tespit edildi. Polikliniğe başvuran hastalar (0.107) ile TFFD'lu (0.707) hastalar arasındaki allerji oranı farkı istatistiksel olarak anlamlı bulundu (t-değeri = 8,345, P = 0.000). Total IgE düzeyleri incelenen 27 (% 65.85) hastanın 15'inde (% 55.5) yüksek bulundu. Alerjik hastaların kliniğe daha sık başvurdukları Mart-Ağustos aylarında TFFD'lu vakalar da daha sık görüldü.

Sonuç: Çalışmamız, TFFD'nun mevsimsel dağılımı, alerjik hastalıklarla eşzamanlılığı ve yüksek IgE düzeylerini dikkate alarak alerjik hastalıklarla ilişkili bir bulgu olabileceğini düşündürmektedir.

Anahtar Kelimeler: Terra Firma Forme Dermatozu, alerji, pediatric, IgE

by dirty brownish asymptomatic plaques caused by idiopathic hyperkeratosis. The patient's hygiene is normal, unlike in dermatitis neglecta. Patients state that they repeatedly washed the lesions with

Table 1. Lesion localizations

Localization	Number of cases	Percent
Trunk	21	51.22%
Upper extremity	15	36.59%
Lower extremity	1	2.44%
Neck	1	2.44%
Neck + lower extremity	1	2.44%
Neck + trunk	1	2.44%
Neck + upper extremity	1	2.44%
Total	41	

Table 2. Accompanying diseases

ICD-10	Diagnosis	Number	Percent
J01.9	Acute sinusitis, undefined	9	11.54%
L20.9	Atopic dermatitis, undefined *	8	10.26%
J30.2	Seasonal allergic rhinitis, other*	7	8.97%
J45.9	Asthma, undefined*	4	5.13%
B82.9	Intestinal peristaltism, undefined	4	5.13%
K52.9	Gastroenteritis and colitis, non-infectious, undefined	4	5.13%
H65.0	Acute serous otitis media	4	5.13%
E66.8	Obesity, other	4	5.13%
R11	Nausea and vomiting	3	3.85%
K52.8	Gastroenteritis and colitis, other, defined, non-infectious	3	3.85%
J35.0	Chronic tonsillitis	3	3.85%
E61.1	Iron deficiency	2	2.56%
R10.4	Abdominal pain, other and undefined	2	2.56%
F98.0	Non-organic enuresis	2	2.56%
N39.0	Urinary tract infection, not localized	2	2.56%
J35.2	Adenoid hypertrophy	1	1.28%
L04.9	Acute lymphadenitis, undefined	1	1.28%
J01.0	Acute maxillary sinusitis	1	1.28%
J30.4	Allergic rhinitis, undefined*	1	1.28%
K60.0	Anal fissure, acute	1	1.28%
J45.0	Asthma, allergic*	1	1.28%
Q21.1	Atrial septal defect	1	1.28%
R51	Headache	1	1.28%
R42	Dizziness (Vertigo)	1	1.28%
R01.0	Benign and innocent heart murmur	1	1.28%
Z00.2	Examination in rapidly growing period in childhood	1	1.28%
K21.9	Gastro-esophageal reflux disease, without esophagitis	1	1.28%
R05	Cough	1	1.28%
L08.0	Pyoderma	1	1.28%
Z00.1	Routine child examination	1	1.28%
L50.9	Urticaria, undefined	1	1.28%
W57	Bite by non-toxic insect and arthropod	1	1.28%

Note: \* Allergic diseases



Fig. 1. Diagnosis was made with the disappearance of the lesions when cleaned with 70% ethyl alcohol

soap, but they were not cleaned away (2). Diagnosis and treatment involve cleaning with 70% alcohol which causes the lesions to disappear (1).

Case reports and a few clinical studies are available in the literature about this disease. Research studies are available about its incidence, gender distribution, involved areas, comorbidities, and laboratory tests. In our study, we aimed to investigate the clinical characteristics of terra firma-forme retrospectively, accompanying diseases, and any connection to allergic diseases.

# Material and Method

Patients under 18 years who were admitted to the Pediatrics Outpatient Clinic for one year from September 2014 suspected to have TFFD during physical examination and who consulted with the dermatology clinic were included in the study. Data were retrospectively obtained from the automation system of the hospital. Diagnosis was made upon the disappearance of the lesions when cleaned with 70% ethyl alcohol (Figure 1). Patients for whom hygiene could not be exactly decided were not included in the study. Lesions lost by washing with water and soap were accepted as normal skin pollution and were left out of work. Biopsy was obtained in no patients. Information on age, gender, medical history, and family history of the patients was obtained; and the anatomic localizations of the lesions, the time it took to notice the lesions, the patient's complaints on diagnoses, admission, the accompanying laboratory tests, and total IgE levels were

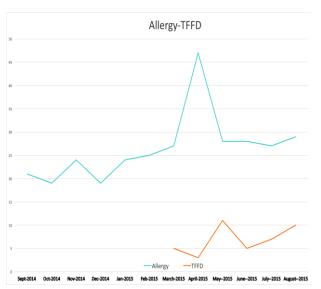


Fig. 2. A peak was observed in TFFD cases following the increase in allergic diseases

evaluated. Ethics committee approval was obtained from the local committee prior to the study (project number: 17-9-2015/135).

**Statistical Analysis:** Results were analyzed in an IBM SPSS statistics subscription (Ver:05-2017) program. One sample t-test was done to determine the difference in allergy ratio between the patients who were admitted to the outpatient clinic and the patients with TFFD.

### Results

A total of 869 patients were examined in the pediatric outpatient clinic within the study period. Forty-one patients (2.19%) diagnosed with TFFD were included in the study.

Of the patients, 27 (65.90%) were girls and 14 (34.10%) were boys. The mean age of the study group was  $7.3 \pm 2.90$  years (range 22 months-13.10 years).

There was the history of allergy in the mother or the father in 8 (19.50%) patients.

Two patients were sisters aged 6 and 13 years.

There was the history of smoking in 19 families (46%) but not in 19 (46%). Data were not available for three patients.

When patients were evaluated with regard to the complaint for admission to pediatric clinic, it was learned that 12 admitted with a complaint of coughing, and coughing was observed to be the most common cause of admission. The other causes of admission were fever (n=7), headache (n=4), vomiting (n=4), nasal discharge (n=3), rash (n=3), request for an approval report for swimming in the pool (n=2), wheezing (n=2),

**Table 3.** Comparison of the patients with TFFD to all patients

Total patients	TFFD patients	Total patients Allergy ratio	TFFD patients Allergy ratio	t value
869	41	0.107	0.707	8.345 (P=0.000)

constipation (n=2), abdominal pain (n=2), dyspnea (n=2), growth assessment, growth retardation, skin spots, swelling in the lower lip, dizziness, blood in the diaper, sore throat, throat irritability, sniffing, imbalance, enuresis, genital discharge, fatigue, sneezing, snoring, urinary incontinence, diarrhea, loss of appetite, failure to thrive, stomachache, malar rash, and dysphagia.

None of the patients reported skin lesions which are the symptoms of TFFD as their complaint, So there was no statement about the time of onset. Lesions disappeared one to two months after the examination, so we could not precisely evaluate the duration of the lesions. Three mothers stated that they had seen and cared for these lesions before. Mothers stated that they defined these lesions as "Yerli Kara" (in English, "indigenous black"); they were not cleaned away by washing, and they gave up with time. The mothers' statement in Turkish is almost same as the Latin name of the disease.

When the lesions were evaluated with regard to localizations, truncal (chest + abdomen) involvement was observed in 21 (51.22%) patients. Involvement areas were described as trunk, flexor side of the elbow, abdomen, flexor side of the arm, back, neck, elbow, chest, extensor side of the elbow, leg, medial side of the knee, upper quadrants of the abdomen, abdomen, right upper quadrant of the abdomen, extensor side of the knee, medial side of the hand, inguinal region, around breast, anterior side of the right chest, left chest, anterior side of the chest, medial side of the leg, and forearm. Lesions were symmetrical in 10 (24%) patients and asymmetrical in 31 (76%) patients. Lesion localizations are classified and given in (Table 1).

When patients were evaluated regarding accompanying diseases, it was observed that a total of 78 diagnoses were made for 41 patients. The most common diagnosis was acute sinusitis (11.50%, n=9). The other accompanying diseases are shown in (Table 2).

When atopic dermatitis, allergic rhinitis, and asthma were accepted as allergic diseases, at least one of these diseases was detected in a total of 29 (70.7%) patients (during examination in 21 patients and in medical history of eight patients).

There was an allergic disease in 200 out of 1869 patients (10.70%) who were admitted to the outpatient clinic.

One-sample t-test was done to determine whether the difference between the allergy ratio in the patients who were admitted to the outpatient clinic (0.107) and the patients with TFFD (0.707) where t was found to be 8,345 (P = 0.000); thus, the difference was significant (Table 3).

A statistical analysis was done by date of admission in order to evaluate the relationship between TFFD and season. The results are summarized in (Table 4). Distribution according to months is indicated on the graph along with allergic diseases in the patients who were admitted to the outpatient clinic. A peak was observed in the TFFD cases following an increase in allergic diseases (Figure 2).

The total IgE levels were found to be high in 15 (55.5%) of 27 (65.85%) patients in whom the total IgE level was examined. The total IgE levels varied between 1.31 and 3845 ng/ml, the mean total IgE level was found to be 493.5 ng/ml, and the standard deviation to be 799 ng/ml.

The total IgE levels was found to be high in 5 (62.5%) of 8 patients with atopic dermatitis.

When patients were evaluated with regard to thyroid functions, TSH (thyroid stimulating hormone) was found to be mildly elevated in only one patient out of 26 (63%) patients in whom TSH was examined. The TSH levels varied between 0.56 and 4.76  $\mu\text{IU/ml}$ , the mean TSH level was found to be 2.56  $\mu\text{IU/ml}$ , the mean free T4 value was found to be 18.5 pmol/L, and the free T4 level was found to be elevated in one patient.

Hemoglobin values were measured in 25 (61%) patients and were found to be within normal ranges in all age groups.

Plasma eosinophil values were high in seven out of 25 (28%) patients. Eosinophilia is present in 3 (37.5%) of 8 patients with atopic dermatitis. However, Eosinophilia is only present in 4 (12%) of 33 non-atopic cases (eosinophil ratio > 10% or > 250 count/mm³). We detected that TFFD was more frequent in March-August when allergic patients are admitted to the clinic more frequently (Figure 2).

**Table 4.** Distribution of the time of admission according to months in TFFD patients

March	5	12.20%
April	3	7.32%
May	11	26.83%
June	5	12.20%
July	7	17.07%
August	10	24.39%

## Discussion

Although terra firma-forme dermatosis was defined in the 20th century, its etiopathogenesis has not yet been discovered. It is thought to develop due to delay in keratinocyte maturation, melatonin retention and accumulation of sebum, sweat, corneocyte, and microorganisms in the regions like neck, trunk, and umbilicus where hygiene care is less scrupulous (3).

Keratinocyte retention rather than keratinocyte proliferation is considered to play a role in the etiopathogenesis of the disease although it develops from adhered dirty crust formation resulting from insufficient exfoliation of keratinocytes (3).

In a series of four cases published by Laith Akkash et al., lamellar hyperkeratosis, papillomatosis, curly orthokeratosis, and mild acanthosis were detected in the biopsy of three cases (4).

Siblings with TFFD were reported (5). In our study, two sisters were diagnosed with TFFD concurrently, and intermarriage was detected in two patients. This condition suggests the possibility of familial and genetic susceptibility. Sun exposure was proposed as a triggering factor in some studies (6),(7),(2). We could not associate the lesions with sun exposure as they were mostly located in clothed areas.

Whether there is a relationship between allergic diseases and TFFD has not been reported before. However, allergic rhinitis was reported in four cases, and atopic dermatitis was reported in six cases (8). Atopic dermatitis was detected in 12 patients in the study by Berk et al. (2). TFFD was associated with emollients and urea-containing preparations in another study, and one patient was reported to have used them due to xerosis, and one patient was reported to have used them due to atopic dermatitis (1). In our study, when atopic dermatitis, allergic rhinitis, and asthma were accepted as allergic diseases, at least one these diseases was detected in a total of 29 (70.7%)

patients with TFFD. The difference between the allergy ratio in the patients who were admitted to the outpatient clinic (0.107) and the patients with TFFD (0.707) was found significant statistically (t-value=8,345, P=0.000). These data support the theory that there is a significant relationship between TFFD and allergic diseases.

We detected that TFFD was more frequent in March-August when allergic patients are admitted to the clinic more frequently (Figure 2). These data also support the fact that TFFD may be associated with allergic diseases like atopic dermatitis. More study is required on this issue since these diseases could help to explain the etiology of TFFD.

Elevated serum IgE levels are associated with allergic diseases because IgE is the primary antibody of immediate hypersensitivity reactions. Virenda et al. found IgE levels to be elevated in 92% of the children with atopic dermatitis in different age groups (9). To the best of our knowledge, our study is the first to examine the total IgE levels in TFFD cases. The total IgE levels were found to be elevated in half (n=15) of the patients in whom total IgE levels were examined (n=27). This condition also indicates that TFFD could be associated with allergic diseases.

In our study, lesions were mostly detected on the trunk, followed by abdomen and extremities in our study. Lesions were mostly detected on the neck in the study by Berk et al. and in the trunk in the study by Aslan et al., similar to our study (8). Lesions were usually reported to be symmetrical, and localized or generalized less frequently (1),(2),(10). In our study, 24% (n=10) of the lesions were observed to be symmetrical. A whole body examination is important in order to detect TFFD in children. All of the patients were referred from the general pediatrics clinic where a whole-body examination was performed.

In previous studies, TFFD was not associated with thyroid diseases. TSH was found to be elevated in only one out of 26 patients (61%), the accompanying disease was obesity in this patient, and the fT4 level was normal. Also, in our study, a relationship was not found between thyroid dysfunctions and TFFD.

In conclusion, our study suggests that TFFD may be a finding associated with allergic diseases considering the seasonal distribution of the disease, and its concurrence with allergic diseases and IgE levels. It is beneficial to question and examine the children with TFFD for allergic diseases. More comprehensive and multi-center studies are required on this issue.

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