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ORIGINAL ARTICLE

Factitious dermatitis in childhood: a retrospective study of a case series

Claudia Santos Oliveira[1], Vânia Oliveira de Carvalho[1], Renata Robl Imoto[1]

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Abstract

Objective: To describe a series of cases of patients with factitious dermatitis (FD) evaluated in a Brazilian tertiary medical care service. **Methods:** This is a retrospective and descriptive study with data collection from the medical records of patients with FD, under the age of 16, seen in a Pediatric Dermatology service from 2007 to 2017. We used the T-test for statistical analysis, and the Pearson's chi-square test considered a 5% significance level. **Results:** The sample consisted of 50 patients and 35 (70%) were female. The mean age at onset of symptoms was 9.8 ± 3.0 years and the median time to seek care was 6.5 months (3 days to 8 years). Twenty-one (42%) patients had some type of learning disability. Thirteen (26%) patients had a personal history of psychiatric illnesses. All patients had more than one type of skin lesion and 33 (66%) had an erythematous-scaling plaque. The upper limbs were the most affected region. Thirteen (26%) patients had psychiatric illnesses. Among the patients who showed clinical improvement, 13 (65%) were undergoing psychological follow-up and disease recurrence occurred in 14 (28%) patients. **Conclusions:** FD most frequently affects females and presents with varied lesions and in easily accessible areas. The diagnosis should be suspected in view of a vague evolutionary history associated with polymorphic and symmetrical lesions. Psychological and psychiatric follow-up are essential, in addition to dermatological, as there is a risk of progression with psychiatric comorbidities.

[1] Hospital de Clínicas - Universidade Federal do Paraná, Departamento de Pediatria - Curitiba - Paraná - Brazil

Correspondence to:
Claudia Santos Oliveira
Hospital de Clínicas da Universidade Federal do Paraná. Rua Padre Camargo, nº 285, Alto da Glória. Curitiba - PR. Brazil. CEP: 80.060-240. E-mail: clau.s.oliveira@hotmail.com

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INTRODUCTION

Factitious Dermatitis (FD) is the name given to skin or mucosal lesions caused or aggravated deliberately and secretly by the patient himself with the objective of secondary gain[1]. It is characterized by a clinical history not compatible with the observed aspect, bizarre, geometric, well-demarcated lesions, preferably located in easily accessible areas with the hands[2,3,4].

In general, the patient and guardian denied any causal link to the injuries, which are produced mainly by mechanical and chemical means, such as nails, sharp or blunt objects, burns by cigarettes and caustic chemicals[5].

The diagnosis is difficult and can lead to unnecessary and non-resolving investigations and treatments[6,7]. The diagnosis is of exclusion, because the FD mimics several common skin disorders. There are clues in history that guide clinical reasoning, such as the complaint of injuries that appeared suddenly and the absence of a description of the evolutionary characteristics of the disease.

It is believed to be more frequent than reported, as it is poorly recognized, and thus underdiagnosed[8]. It is estimated that FD represents from one in 200 to one in 2,000 dermatological consultations[5]. Dermatosis is recognized by dermatologists, but little diagnosed by pediatricians, who are more likely to suspect abuse by third parties[1]. In addition, it is this professional, in most cases, responsible for the patient's first care. As lesions can be misdiagnosed and in order to avoid unnecessary treatments and delays in treatment, knowledge about FD must be improved and widely disseminated in the medical literature. FD's diagnosis and management in children should be performed by the pediatrician, both concerning the recognition of the disease and the referral for psychological treatment[9].

There are some case reports in the literature[2,3,7,8,10,11] and few case series[12,13] have been published on the topic. For this reason, this study aims to describe data from a series of FD cases in children and adolescents.

METHODS

This is a retrospective and descriptive study approved by the institution's Research Ethics Committee. We collected our data through the evaluation of the medical records that were located from the spreadsheets of a Pediatric Dermatology service with the terms factitious dermatitis, pathomy or self-induced injury. We included all the children under the age of 16 diagnosed with FD, seen from 2007 to 2017.

The data evaluated were: gender, age at disease onset, school modality, educational level, academic performance, learning disability, age and marital status of the parents, characteristic of family life, past morbid history, morbid family and personal psychiatric history, use of systemic medication, time of disease progression until seeking medical attention, improvement factors, triggering factors, lesion type and topography, associated symptoms, complementary tests and biopsy carried out, consultations with other specialties, number of consultations at the outpatient clinic pediatric dermatology, clinical evolution and recurrence. We also assessed the service's image bank.

The academic performance was classified as regular when there was school failure or school difficulties in the clinical history. Family life was categorized as bad according to situations found in the medical records that are described in Table 1.

The statistical analysis was carried out according to the nature of the variables, number of study groups and types of analysis required with the aid of the JMP® program. Independent t-test and Pearson's chi-square test were used with a significance level of 5%.

RESULTS

General sample characteristics

The sample consisted of 50 children, and 35 (70%) were females. The mean age of symptom onset was 9.8 ± 3.0 years and the mean age of lesion onset for girls and boys was similar (Table 2). The median age at the first consultation was 10 years (2.5 to 14 years) and the median time from the onset of symptoms to seeking medical care was 6.5 months (3 days to 8 years).

Thirty-six (72%) attended regular school, and the academic performance of 18 (36%) was considered good (Table 1). Twenty-one (42%) had some type of learning disability (Graph 1), 16 (32%) had no difficulty, and for 13 (26%) patients there was no information in the medical records. The learning difficulties and the academic performance of boys and girls was similar (Table 2).

Concerning family members' data, the mean age was 35.7 ± 7.7 years for mothers and 39.7 ± 7.0 years for fathers. Fifty-six percent (28) were married, and family life was described as good in 34% (17), the other data is shown in Table 1.

Associated comorbidities

There was some comorbidity in 33 (66%) participants and the same patient had more than one comorbidity (Table 1).

Thirteen (26%) patients had a personal history of psychiatric illness (conduct, mood, anxiety and obsessive compulsive disorders), 5 (10%) had a family history of psychiatric illnesses, and one of the patients developed FD 4 months after his sister's suicide. Eight (16%) patients used systemic medication for neurological or psychiatric problems.

Regarding FD's triggering factors, twenty-nine (58%) had no clear factor, nineteen (38%) reported that it was triggered by: anxiety attacks, family disagreements, sexual abuse, birth of a younger brother, lack of parental attention,

Table 1. Frequency distribution of the participants' demographic data

	Males	Females	*p*
	15 (30%)	35 (70%)	
Age of onset in years (mean ±SD) *	8.7 ±3.21	10.3 ±2.8	0.10
School performance n (%)			
Good	6 (40)	12 (34.28)	0.7
Regular	5 (33.33)	16 (45.71)	
No data	4 (26.67)	7 (20)	
Learning difficulties n (%)			
With learning difficulties	7 (46.66)	14 (40)	0.85
Without learning difficulties	4 (26.67)	12 (34.28)	
No data	4 (26.67)	9 (25.72)	
Clinical progress n (%)			
With clinical improvement	5 (33.33)	15 (42.86)	0.52
Without improvement by the latest assessment	10 (66.67)	20 (57.14)	

*SD=standard deviation

worsening of chronic illness, bullying at school, parents' divorce, instability in the family environment, change of city, family violence and maternal rejection, and 2 (4%) did not have the information in the medical record.

Skin lesions' characteristics

No lesion improvement factor was identified in 46 (92%); two (4%) reported improvement with psychotherapy, one (2%) with antihistamine and one (2%) after moving to another city.

All patients had more than one type of skin lesion and 33 (66%) had an erythematous-scaling plaque (Table 1). Thirty-five (70%) patients had lesions in more than one region of the body, and the upper limb was the most affected region; Figure 1 shows the distribution of the lesions' topography.

Among the associated signs and symptoms there were 16 (32%) itching, 8 (4%) secondary infection, 8 (4%) onychophagia and 8 (4%) trichotillomania.

Evolutionary clinical follow-up

During clinical follow-up, laboratory tests were requested for 4 (8%) patients, 3 (6%) were hospitalized for investigation, and skin biopsy was performed on 8 (16%) patients. Follow-up with other specialties was frequent in the sample, mainly psychology (Table 2).

The participants had a median of 3 (1 to 36) consultations at the Pediatric Dermatology outpatient clinic; 20 (40%) progressed with improvements, 8 (16%) did not show improvement until the last visit and 22 (44%) lost follow-up. Fifteen (42.85%) female patients improved (Table 1). Of the patients who showed clinical improvement during evolution, 13 (65%) were undergoing psychological monitoring and 7 (35%) were not (p = 0.41). Patients who evolved with improvements had a mean age of onset of 10.0 ± 3.05 years and those who did not improve until the last evaluation had 9.7 ± 3.05 years of age (p = 0.72). FD recurrence occurred in 14 (28%) patients.

DISCUSSION

FD is rarely reported in the pediatric population[14]. Considering that there were no FD case series publications in the pediatric range in Brazil, this is the first study that presents the profile of a series of Brazilian children and adolescents.

FD occurs more commonly in adolescents, young people and adult women[15]. In the present study, the girls were more affected (70%), similar to what has been reported by other authors[10,12]. Saez-de-Ocariz et al reported that over a 19-year period in Mexico, females were the most prevalent (6.2: 1)[12]. Comparing average ages, we found that the patients were younger - 9.8 ± 3.0 years, compared to Saez-de-Ocariz et al, whose patients were 11.17 ± 4.12 years of age[12].

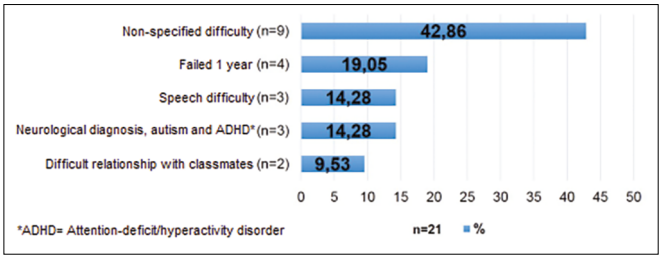
The pathophysiology is still poorly understood, and multifactorial causes include genetic, psychosocial factors and personal or family history of psychiatric illnesses[8]. In the present study, five (10%) patients had a family history of psychiatric illnesses and 13 (26%) had a personal history. There was also a case in which self-mutilation started after the sister's suicide, probably as a request for attention in a situation of disruption of family relationships.

Inquiring about problems in living together at home and at school, and assessing the quality of life and the ability to interact in these environments can help identify problems in specific areas to be addressed in FD[16]. Regular academic performance was present in 21 (42%) patients in the present

Table 2. Characteristics of participants and their families

Characteristic	n (%)	Characteristic	n (%)
School type		**Comorbidities**	33 (66)
Regular School	36 (72)	Obesity	8 (24)
Special School	4 (8)	Epilepsy	7 (21)
No info in chart	10 (20)	Asthma	4 (12)
School performance		Atopic dermatitis	3 (9)
		Heart disease	3 (9)
Good	18 (36)	Psoriasis	2 (6)
Regular	21 (42)	Others#	10 (30)*
No info in chart	11 (22)		
Learning difficulty		**Characteristics of the skin lesions**	
Yes	21 (42)	Erythematous-shedding plaque	33 (66)
No	16 (32)	Bruise	32 (64)
No info in chart	13 (26)	Residual dyschromia	30 (60)
Marital status of parents		Hematic crusts	21 (42)
		Weird aspect	14 (28)
Married	28 (56)	Linear shape	13 (26)
Divorced	12 (24)	Meliceric crusts	10 (20)
Single	5 (10)	Purplish spot	6 (12)
With someone	1 (2)	Lichenification	6 (12)
No info in chart	4 (8)	Bite	1 (2)
Family life		**Follow up with specialties**	
Good	17 (34)	Psychology	28 (56)
Bad**	10 (20)	Neurology	6 (12)
No info in chart	23 (46)	Psychiatry	6 (12)
		Cardiology	2 (4)
		Endocrinology	1 (2)
		Speech therapy	1 (2)
		Not followed up	14 (28)

** Father's alcoholism, sexual abuse, Family instability after parents' divorce, difficulties during the divorce, abandonment.
# Alopecia *universalis*, height and weight deficit, blood dyscrasia, nocturnal enuresis, strabismus, systemic arterial hypertension, congenital hypothyroidism, treated lymphoma, gigantic nevus and down syndrome.
* We followed the same frequency for each one of the items.



Graph 1. Distribution of the types of learning difficulties.

study. It is up to the pediatrician or dermatologist to seek this information in the interview and plan the best form of intervention with the multidisciplinary team and the family.

Family life crises are described in the genesis of FD[3,7]. The families involved in the present study had parents with an average age over 30 years; predominantly married (56%) and approximately a quarter of them were divorced. Family life was categorized as bad in 20% of the cases. In these families, there were situations of an alcoholic father, abandonment by the parents, attempted sexual abuse by the stepfather, separation from the mother and divorce from the parents. This confirms the reports by Moss, that in the cases of children and adolescents, FD is like a manifestation in response to some type of pressure or a "cry for help"[1], as in cases of sexual abuse.

The prevalence of comorbidities was higher (66%) when compared to that found in the literature, in which Saez-

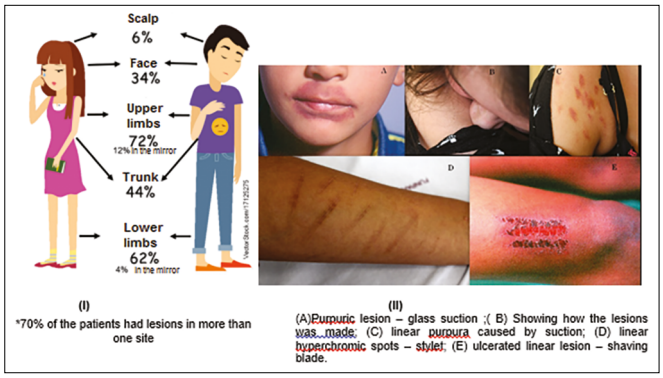


Figure 1. (I) Topographic distribution of the frequency of lesions, considering each region alone *; (II) Clinical aspects of FD lesions.

de-Ocariz et al., had 41% of patients with some associated disease[12]. This difference could be justified by the sample of the present study being composed of patients being monitored at the tertiary level. Obesity was the most frequent comorbidity (24%), and stigmatization of obese people is frequent and causes damage to self-esteem, that culminates in disorders in global mental health[17]. In the context of the present study, it is possible that obesity lead to social isolation and bullying, thus self-mutilation would be the escape valve from the social pressure suffered by these children and adolescents.

The median time spent looking for medical care and diagnosis was 6.5 months, but it took as long as 8 years. For Saez-de-Ocariz et al, patients took an average of 10 months until diagnosis, with a maximum of 4 years[12]. Thus, there were patients who did not seek immediate medical attention for the lesions and there were those who did not receive a correct initial diagnosis, and possibly underwent unnecessary interventions and/or exams until the proper diagnosis was established. FD in children is among the most difficult diagnoses, and should be considered when other skin conditions are excluded[13].

Thirty-eight percent of the patients had triggering factors for FD. Possibly because during the consultation the other patients chose to hide them. According to Soong et al, patients with FD frequently describes unsatisfactory or "hidden" stories and deny self-mutilation[10].

For those patients who noticed a symptom improvement factor (8%), a placebo factor was suggested in one case, when the improvement in FD was related to an antihistamine. The change of city as an improvement factor leaves the hint of a stressing factor being responsible for the injuries. A minority attributed the improvement to psychotherapy (4%); however, those who were being followed up by psychologists showed a higher percentage of improvement, which demonstrated the importance of clinical and multiprofessional monitoring, which is difficult due to low adherence to treatment[18]. In addition, consultations should be frequent, because patients must be followed up carefully, so that new injuries are not caused during follow-up.

Skin lesions are described as polymorphic with superficial abrasions, residual hyperchromia, necrosis, ulceration, and crusts, purple and bullous lesions[19]. Lesions with bizarre and/or geometric morphology are common[16]. In the present study, the most frequent clinical form was the erythematous-scaling plaque, followed by excoriation, residual dyschromia, hematic crusts, bizarre and linear lesions. In agreement with the polymorphic character of the disease, not often found in other dermatoses and with its post-inflammatory character represented by residual dyschromia, result from the sequela of skin diseases. As for the location, the upper limbs were the most frequently affected, with mirror injuries, that is, bilateral and symmetrical injuries. In agreement with what has been published in the literature[6], in which FD lesions are typically bilateral, symmetrical and easy to reach the dominant hands.

According to Yamada et al., children with FD may have systemic complaints, such as unexplained pain, fatigue, nausea, dizziness, verbal aphasia and age regression[11]. The data obtained did not confirm what this author exposed, as the most prevalent symptom was pruritus (32%). Possibly the itching resulted from self-inflicted excoriation, which creates a reaction on the skin that leads to the symptom. A limitation in obtaining these data is the retrospective character and the bias is in performing it in a Pediatric Dermatology service, so the evaluation was restricted to patients whose main symptoms were skin-related.

There were also patients with trichotillomania (4%) and onychophagia (4%), which, according to Moss, are self-inflicted diseases, but which, unlike FD, did not intend to create a skin disease for secondary gains[1].

FD is associated with requesting unnecessary tests that cause diagnostic delays[7]. In the present study, laboratory tests were requested for four (8%) patients, and three (6%) underwent hospitalization. Skin biopsy helps to rule out masked skin diseases, such as basal cell carcinoma, vasculitis or herpetic lesions[10].

Hospitalization is a recurrent situation in pediatric FD cases[3,6,8,11,15]. It is justified as a form of diagnostic clarification, because it is possible to control the evolution of the lesions and to hide from the patient tools that can be used to commit them. In addition, for those families resistant to the diagnosis, hospitalization reinforces the doctor-patient relationship and enables family acceptance that the problem is not organic.

The 44 patients with FD assessed by Luna et al. had an average of four medical consultations, and three patients (6.8%) agreed to undergo psychiatric follow-up[13]. In the present study, the number of consultations at the outpatient clinic had a median of three, and there was greater adherence to psychiatric assessment and monitoring (12%), and 56% were followed up with a psychologist. We still challenge which is the ideal psychotherapeutic approach for FD. Possibly a good doctor-patient relationship is an important point in therapeutic compliance.

FD's prognosis is poor when considered that 14 (28%) had recurrences, a value similar to that mentioned by Ikenaga et al. in a survey of 43 patients, with 30% of recurrences[6]. There was no statistical difference between those who were under psychological counseling or not. At this point, it is possible to expose a difficulty in controlling external stressors that trigger self-mutilation.

In conclusion, FD was characterized by a higher frequency in girls, presented through lesions with a vague evolutionary history, located in easily accessible areas and in a mirror, and there was an association with psychiatric comorbidities. It is expected that with the results obtained, more academics, doctors and pediatricians will include FD among their diagnostic hypotheses, avoiding iatrogenesis and the delay in multidisciplinary therapy.

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