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Review Article

Assessment of Pediatric Hidradenitis Suppurativa at a Regional Medical Center

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ABSTRACT

Objective: Hidradenitis Suppurativa (HS) is a chronic, recurrent inflammatory disorder involving hair follicular occlusion, inflammation, and scarring. Few studies have been designed to characterize HS in pediatric patients. This retrospective study assessed the pediatric HS population at an academic medical center.

Methods: A retrospective chart review was performed of all encounters for HS from April 2018 to July 2022 at Children's Hospital in New Orleans, Louisiana. A total of 202 patients diagnosed with HS (ICD10=L73.2) were reviewed. **Results:** Most patients identified as African American (72.4%). The mean age at HS onset was 12.1 years (SD: 1.9; range: 7-16); mean age of diagnosis was 13.0 years (SD: 1.9; range: 9-17). Family history of HS was not documented in 81.6% of pediatric patients. Pre-teen onset of disease (0-12 y) was recorded in 53.9% of patients. 50% of patients presented with Hurley stage I; 43.4%, stage II; and 6.6%, stage III. No association was found between Hurley stage at diagnosis and gender (p=0.610), race (p=0.603), or pre-teen onset (p=0.716). Eight patients were undergoing biologic therapy (infliximab, n=2; adalimumab, n=6), including 10.9% of females, 8.3% of males, 12.7% of African Americans, and 7.7% of Caucasians.

Conclusions: Family history is an important aspect of clinical history to ascertain in pediatric HS, as it is associated with early onset disease. Pediatric HS disproportionately affects non-Caucasian populations presenting with more severe disease. Clinicians should be aware of this trend in caring for pediatric patients. 50% of patients on biologic therapy reported pre-teen onset of disease. In HS, early onset disease has been correlated with greater overall disease severity. Thus, more patients with early onset HS and likely moderate-to-severe disease will be initiated on biologic therapy in the future. It is crucial to detect pediatric HS in a timely manner, and counsel patients with pediatric HS on the possibility of biologic therapy.

Keywords: Hidradenitis suppurativa; Body mass index

INTRODUCTION

Hidradenitis Suppurativa (HS) is a chronic, recurrent inflammatory disorder involving hair follicular occlusion, inflammation, and scarring [1,2]. The disease is characterized clinically by episodes of painful, inflamed, purulent nodules, abscesses, sinus tracts, and scar formation at intertriginous areas [3,4]. HS is a chronic, debilitating disease that can have significant psychosocial impacts (i.e. depression, anxiety) on the affected individual, as it is often recalcitrant to standard therapeutic options [4]. The average age of onset is in the early 20's while only 2%-3% of patients with HS report disease onset pre-pubertally (before 11 years old) [2,4,5]. HS is more common in females than males by a ratio of 2:1 [6]. One single-center study found that pre-pubescent onset of HS is more likely to occur in males, while post-pubescent onset is more likely in females [7]. Pediatric cases (<18 years old) of HS are rare and can indicate endocrine axis anomaly, such as hormonal imbalance, adrenal hyperplasia, and premature adrenarche [6,8-13]. Pediatric

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onset of disease is associated with a positive family history and more widespread disease, and clinical disease manifestations are also influenced by known risk factors such as smoking and Body Mass Index (BMI) [2,14,15]. Case control studies indicate that pediatric patients with HS have higher rates of depression, anxiety, acne vulgaris, obesity, metabolic syndrome, inflammatory bowel disease, and Down syndrome [16-18].

Standard therapeutic options for the treatment of HS include topical and systemic antibiotics, oral retinoids, hormonal therapy, immunosuppressive drugs, biologic agents, and surgical excision [4,19,20]. Early diagnosis and appropriate treatment of pediatric HS is critical in minimizing the deleterious effects of the disease; however, therapeutic management of HS in pediatric population has been poorly studied. Mild cases are typically treated with topical antibiotics, while both topical and oral agents are used to treat moderate disease. Modalities such as immunosuppressive agents, biologic therapies, laser treatment, and surgery are reserved for severe cases of HS [4,19,20]. Few studies, reviews, and practice guidelines have been designed to characterize the demographics, clinical characteristics, and therapeutic management of HS in pediatric patients. Given that research in pediatric HS is limited; this retrospective study assessed the pediatric HS population at a large regional academic medical center. The objective of this study is to characterize the pediatric HS population and evaluate therapeutic and surgical regimens currently in use.

METHODS

A retrospective chart review was performed of all encounters (i.e., emergency room visits, clinic visits with dermatologists and non-dermatologists) for HS from April 2018 to July 2022 at Children's Hospital in New Orleans, Louisiana (CHNOLA). A total of 202 patients diagnosed with HS (ICD10=L73.2) were reviewed. Patients included in the analysis (n=77) were less than 18 years old at time of HS diagnosis by a dermatologist or in 2 subsequent visits by a non-dermatologist at CHNOLA. Diagnoses were confirmed if physical examination found typical lesions of HS at anatomic sites known to commonly be affected by HS (e.g., axillae, buttocks, groin, and inframammary region). Data extracted from the patients' medical chart included age, gender, race, payor status, age at diagnosis of HS, age at onset of HS, disease severity (defined according to Hurley stages; often documented; if not, based on examination findings), anatomical distribution of disease at presentation, family history, and treatment regimen. Data was analyzed using the SAS/STAT software, Version 9.4 of the SAS System for PC. Copyright 2014 SAS Institute Inc., Cary, NC. The association between Hurley stage at diagnosis and gender, race, and pre-teen onset were assessed using either two-sided χ^2 test or 2-sided Fisher exact test. Significance was declared at P-value < 0.05.

RESULTS

Demographics

Patients' characteristics are reported in **Table 1**. A total of 77 children with HS were included in study (64 female [84.2%], 12 male [15.8%]). One patient was excluded because chart review revealed inadequate documentation of medical history and clinical presentation. Most patients identified as African American (72.4%), and the remaining patients identified as Caucasian (17.1%), other race (6.6%), or declined to report their race (3.9%). Most pediatric pa-

tients with HS utilized Medicaid (84.2%), with the remaining payors being commercial insurance (10.5%), self-pay (2.6%), financial assistance (1.3%), and Medicare (1.3%).

Table 1: Demographic features of pediatric patients with hidradenitis suppurativa (n=76)

ltem	Mean (SD)		
Age at onset, y	12.1 (1.9)		
Age at diagnosis, y	13.0 (1.9)		
Gender	% (n)		
Female	84.2 (64)		
Male	15.8 (12)		
Race	0		
African American	72.4 (55)		
Caucasian	17.1 (13)		
Other	6.6 (5)		
Patient Declined	3.9 (3)		
Payor type	0		
Medicaid	84.2 (64)		
Commercial	10.5 (8)		
Self-Pay	2.6 (2)		
Medicare	1.3 (1)		
Financial Assistance	1.3 (1)		
Pre-teen (age 0 y-12 y) onset	53.9 (41)		
Pre-teen onset in females (n=64)	51.6 (33)		
Pre-teen onset in males (n=12)	66.7 (8)		
Family history of HS	0		
Yes	17.1 (13)		
No	1.3 (1)		
Not recorded	81.6 (62)		
Hurley stage at diagnosis	0		
I	50.0 (38)		
II	43.4 (33)		
III	6.6 (5)		
SD=Standard Deviation.			

The mean age at HS onset was 12.1 years (Standard Deviation [SD]:1.9; range: 7-16) and the mean age of diagnosis was 13.0 years (SD: 1.9; range: 9-17). Only 17.1% of patients (n=13) reported a family history of HS, 1.3% (n=1) of patients reported no family history of HS, and family history was not documented in 81.6% (n=62) of pediatric patients. Pre-teen onset of disease (0 y-12 y) was recorded in 53.9% (n=41) of patients. Across gender, 51.6% of females (n=33) and 66.7% of males (n=8) experienced preteen onset of disease.

Disease Severity and Anatomic Distribution of Sites

Overall, 50.0% of patients presented with Hurley stage I, followed by 43.4% stage II (n=33), and 6.6% stage III (n=5). No significant association was found between Hurley stage at diagnosis and gender (p=0.610), race (p=0.603), nor pre-teen onset (p=0.716) (Table 2).

The anatomic distribution of sites affected in pediatric patients with HS is summarized in **Table 3**. Approximately one-third of patients were affected at two or more sites (38.2%, n=29). Of patients

affected at >2 sites (38.2%), most were affected at 2 sites (58.6%). Many patients, specifically 68.4% (n=52), presented with bilateral involvement. The axilla was the most affected site (79.0%), followed by the inguinal fold(s) (19.7%), inner thigh(s) (14.5%), and mons pubis (13.2%).

Table 2: Univariable association between Hurley stage and patient characteristics (n=76).

Item	l (n=38)	II (n=33)	III (n=5)	p-value
Gender		% (n)		0.61
Female	81.6 (31)	87.9 (29)	80.0 (4)	0
Male	18.4 (7)	12.1 (4)	20.0 (1)	0
Race		0		0.603
African American	68.4 (26)	75.8 (25)	80.0 (4)	0
Caucasian	21.1 (8)	15.2 (5)	0 (0)	0
Other	7.9 (3)	6.0 (2)	0 (0)	0
Patient Declined	2.6 (1)	3.0 (1)	20.0 (1)	0
Pre-teen (age 0 yr-12 yr) onset		0		0.716
Yes	42.1(16)	48.5 (16)	60.0 (3)	0
No	57.9 (22)	51.5 (17)	40.0 (2)	0

Table 3: Anatomic Distri	bution of Sites	in Pediatric Pa	tients with Hidrac	deni-
tis Suppurativa				

ltem	% (n)	
Site location ¹	Total	
Axilla	79.0 (60)	
Inguinal fold (s)	19.7 (15)	
Inner thigh (s)	14.5 (11)	
Mons pubis	13.2 (10)	
Chest	9.2 (7)	
Labia	7.9 (6)	
Buttocks	5.3 (4)	
Inframammary fold (s)	5.3 (4)	
Abdomen/panniculus	4.0 (3)	
Intramammary fold (s)	1.3 (1)	
Scalp	1.3 (1)	
Breast	1.3 (1)	
>2 Sites Involved	38.2 (29)	
Bilateral involvement	68.4 (52)	
¹ Locations are not mutually exclusive		

Treatment Regimen

The topical, oral, and procedural treatment regimens utilized for pediatric patients with hidradenitis suppurativa are summarized in Figure 1 and Table 4. Many patients were treated with a single oral antibiotic (67.1%), with doxycycline being the most frequently utilized oral antibiotic (41.1%). Approximately 61.8% of patients were treated with a topical antibiotic, with clindamycin being the most frequently used (76.6%). A total of 59.2% of patients utilized an antiseptic wash; chlorhexidine was the most common (73.3%). Approximately 26.3% of patients utilized a regimen of combination oral antibiotics, of which clindamycin and rifampin was the most common (65.0%). Patients also utilized other systemic medications (metformin, prednisone, sulfasalazine; 11.8%), biologic therapy (10.5%), and hormonal treatment (spironolactone, oral contraceptives; 9.2%). A total of 24 patients (31.7%) underwent a procedure (intralesional corticosteroid, incision and drainage, and/ or excision) in the treatment of HS (Table 4). Of those, 25.0% were treated with intralesional corticosteroids, 58.3% were treated with

incision and drainage, and 37.5% were treated with excision.

 Table 4: Topical, oral, and procedural treatment regimens for pediatric patients with hidradenitis supparativa (n=76)

Item	% (n)	
Single oral antibiotic (n=51)	0	
Doxycycline	41.1 (21)	
Trimethoprim-Sulfamethoxazole	21.6 (11)	
Clindamycin	13.7 (7)	
Cephalexin	5.9 (3)	
Minocycline	5.9 (3)	
Amoxicillin	3.9 (2)	
Rifampin	3.9 (2)	
Dapsone	2.0 (1)	
Erythromycin	2.0 (1)	
Topical antibiotic (n=47)	0	
Clindamycin	76.6 (36)	
Mupirocin	23.4 (11)	
Antiseptic wash (n=45)	0	
Chlorhexidine	73.3 (33)	
Benzoyl peroxide	20.0 (9)	
Rubbing alcohol	4.4 (2)	
Bleach baths	2.2 (1)	
Combination Oral Antibiotics (n=20)	0	
Clindamycin/Rifampin	65.0 (13)	
Metronidazole/Moxifloxacin/Rifampin	10.0 (2)	
Clindamycin/Doxycycline	5.0 (1)	
Clindamycin/Trimethoprim-Sulfamethox- azole	5.0 (1)	
Ampicillin-Sulbactam/Doxycycline	5.0 (1)	
Other systemic medication (n=9)	0	
Metformin	81.8(9)	
Prednisone	9.1(1)	
Sulfasalazine	9.1 (1)	
Biologic Therapy (n=8)	0	
Adalimumab	80.0 (8)	
Infliximab	20.0 (2)	
Hormonal Treatment (n=7)	0	
Spironolactone	71.4 (5)	
Oral contraceptive	28.6 (2)	
Procedure (n=24)	0	
Incision and drainage	58.3 (14)	

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Excision	37.5 (9)
Intralesional corticosteroids	25.0 (6)

Biologic Therapy

Eight patients were undergoing biologic therapy (infliximab, n=2; adalimumab, n=6), consisting of 10.9% of females (n=7), 8.3% of males (n=1), 12.7% of African Americans (n=7), and 7.7% of Caucasians (n=1). These findings are summarized in **Table 5**. Only one patient on biologic therapy reported a positive family history of HS. Four patients on biologic therapy experienced pre-teen onset of disease, and three were Hurley stage II at presentation, while 5 were Hurley stage III at presentation. Five patients on biologic therapy (n=5) reported involved at >2 sites.

 Table 5: Clinical characteristics of pediatric patients with hidradenitis supparativa utilizing biologic therapy (n=8)

Gender	n
Female	7
Males	1
Race	0
African American	7
Caucasian	1
Pre-teen Onset	4
Hurley Stage I	0
II	3
111	5
>2 involved sites	5



Figure 1: Treatment regimens for pediatric patients with hidradenitis supparativa (n=76)

DISCUSSION

Demographics

In this study, pediatric HS was largely a diagnosis in young, African American women who utilize Medicaid, which is consistent with prevalence estimates and other pediatric HS cohort studies among children and adolescents [5,7,21,22]. Early onset HS has been linked to female sex and this study showed a female preponderance for HS similar to gender ratios reported in the literature (5:1 ratio vs 4:1, 4:1, 3:6:1, and 3:8:1) [5,21,23,24]. A small gap existed between age at onset (12.1 y) and age at diagnosis (13.0 y). This diagnostic gap on the order of months is smaller than other pediatric cohort studies, except for Braunberger et al. who reported a diagnostic gap on the orders of months (11 years vs 11.9 years) as well [7,19,21]. In the general population, 35% of patients report a positive family history of HS [25]. Only 16.9% of patients in our study reported a positive family history of HS; family history was not reported in 81.6% of patients. This is an important aspect of the clinical history to ascertain in pediatric HS, as family history of HS is associated with early onset disease. Over half of patients (53.9%) reported pre-teen onset of disease, consistent with other pediatric cohort studies [19]. This underscores the need for various clinicians, i.e., primary care, emergency, endocrinology, dermatology, etc., to be aware of the prevalence and presentation of HS in pediatric populations.

Disease Severity and Anatomic Distribution of Sites

No univariable association was found between Hurley stage at diagnosis and gender, race, and pre-teen onset. Despite these findings, pediatric HS disproportionately affects non-Caucasian populations presenting with more severe disease, and thus clinicians should be aware of this trend in caring for pediatric patients. Larger studies are needed to further characterize pediatric HS and its variable manifestations across gender and race.

Most patients presented with bilateral disease involvement (68.4%), on par with similar pediatric cohort studies [21]. The axilla is typically the most involved site in pediatric HS across gender, consistent with our study findings [19,21]. Continued awareness of differences in HS presentation according to gender are crucial in early diagnosis and minimizing long-term disease sequelae.

Treatment

Antibiotics, both topically and systemically, were used frequently to treat pediatric patients with HS. While previously thought that HS lesions are aseptic, recent research looking at HS lesions challenges this notion [26-28]. A higher incidence of cutaneous infection has been reported in children and adults with HS, and Liy-Wong et al. suggests that traditional methods of skin swabbing may be less useful in pediatric patients with HS [21,29]. Further high-quality studies are needed to elucidate effective treatment regimens and interventions for patients with pediatric HS. In our study, most patients undergoing procedures for HS had an incision and drainage(s) performed. These procedures are typically performed acutely in the setting of actively inflamed HS lesions. Emergency and operating room visits are common throughout the disease course of HS and can likely be mitigated in our pediatric populations with earlier detection of disease, patient education and appropriate treatment.

Biologic Therapy

While antiseptic washes and topical and oral antibiotics are the mainstays of treatment, biologic therapies are being more frequently used in the treatment of moderate-to-severe pediatric HS. Only adalimumab, a TNF-a inhibitor, has been approved by the Food and Drug Administration (FDA) for the treatment of HS in children 12 years of age and older [30]. Additionally, studies in adults with HS maintain highest treatment efficacy with TNF-a inhibitors such as adalimumab and infliximab [31-33]. Of those patients in our study being treated with biologic therapy, the majority were African American females, consistent with the study population. Family history of HS was nominal. Most patients were Hurley stage III at time of presentation and had >2 involved sites. Interestingly, half of the patients on biologic therapy reported pre-teen onset of disease [34]. In HS, early onset disease has been correlated with greater overall disease severity [3,14,15]. It is thus probable that more patients with early onset HS and thus likely moderate-to-severe disease will be initiated on biologic therapy in the future. It is crucial to detect pediatric HS in a timely manner, and counsel patients with pediatric HS on the possibility of biologic therapy.

Limitations

Limitations of this work include its single-center, retrospective design.

CONCLUSION

The goal of this retrospective analysis was to characterize the population of patients with pediatric-onset HS at a large academic medical center. Characteristics of pediatric HS epidemiology, presentation, and treatment have traditionally been understudied. Our results maintain that pediatric onset HS is a disease largely affecting minority populations across race and gender. Early detection and effective treatment regimens are critical in mitigating long-term sequelae of this debilitating disease, especially in pediatric populations. Varying manifestations of pediatric HS across gender and race deserves further study, as well as the likelihood of initiating biologic therapy in patients with pediatric onset disease.

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CONFLICT OF INTEREST

The author's declared that they have no conflict of interest.

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