Original Report

# Vulval Pain in Pediatric and Adolescent Patients

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A B S T R A C T

*Study Objective:* To describe the experience of a tertiary pediatric and adolescent gynecology service that provides care to children and adolescents who present with vulval pain. Their presentation, associated symptoms, and management is described.
*Design:* A retrospective analysis of all girls younger than 18 years of age who presented to the gynecology clinic of our tertiary referral Children's Hospital between Jan 2010 and July 2016. Electronic medical records were reviewed and parameters recorded using a standardized data sheet.
*Setting:* Gynecology clinic of a tertiary referral children's hospital and private rooms of our director of gynecology.
*Participants:* Young women younger than 18 years who presented with symptoms suggestive of vulvodynia.
*Interventions and Main Outcome Measures:* Presenting symptoms, characteristics of associated features, treatment options, and treatment outcomes.
*Results:* Forty-seven patients with a mean age of 11 years (range, 3-18 years) were identified. At the time of diagnosis 31/47 (65.9%) were premenarchal. Many presented with a symptom other than pain alone. In particular, 35/47 (74.4%) presented with coexisting or previous urinary symptoms. Of patients examined, most had positive cotton tip examination findings (16/17 (94.1%) and 11/13 (84.6%) for pre- and postmenarchal, respectively) with clinical inspection otherwise unremarkable.
*Conclusion:* Children and adolescents with vulval pain have varied presentations. Many of the pre- and postmenarchal patients had coexisting urinary tract symptomatology at the time of diagnosis. This review of patients seen over 5.5 years at a pediatric tertiary referral center provides information on the presenting symptoms, examination features, and response to clinical management.
*Key Words:* Vulvodynia, Adolescent, Vulvar pain, Pediatric

## Introduction

Vulvodynia encompasses varied and heterogeneous presentations in women of all ages. The terminology for vulval pain changed in 2015 during a consensus review by the International Pelvic Pain Society, the International Society for the Study of Vulvovaginal Disease, and International Society for the Study of Women's Sexual Health.[1] Changes were made to represent the evolving evidence since the last update in 2003. According to the new classification, vulval pain is now divided into: (1) vulval pain with an identified cause; and (2) vulvodynia, consisting of vulval pain present for at least 3 months with nil identifiable cause but possible associated features.[1,2]

There has been a presumption that the literature on adult vulval pain, including the associations, pathophysiology, and management can be extrapolated and applied to the pediatric and adolescent population. Nevertheless, the literature on pediatric and adolescent vulval pain has been limited to a small number of case reports and series.

The symptom of distressing vaginal pain in the prepubertal population has been described as a specific entity. This genital pain problem does not occur in adolescent or adult women and differs from vulval pain in its etiology and treatment. Vaginal pain in pediatric patients not related to vulvovaginitis usually presents as nocturnal shooting or stabbing pain lasting for a number of hours at night and often wakes the child from sleep. This pain is often associated with "lost" pinworms crawling on the hymen at night thus causing the pain.[3] This entity does not appear to exist in postmenarchal or adult women. Thus there is a possibility that there might be differences in associations, pathophysiology, and management of vulval pain between pediatric, adolescent, and adult women.

Chronic pain has a large disease burden within our society and there is growing evidence in adult women that chronic vulval pain is linked to additional pain syndromes such as irritable bowel syndrome and fibromyalgia.[4,5] A population-based study in 2013 showed that women with vulvodynia are 2-3 times more likely to have one or more other chronic pain conditions compared with controls without vulvodynia.[5] The 2015 consensus terminology and classification statement noted these comorbid pain syndromes as potential factors associated with vulvodynia.[1,2] There are no studies in premenarchal populations or adolescent populations to clarify if these associations exist in younger girls.

In view of the absence of information about vulvodynia in our younger cohort of women and children, we undertook an audit of our clinical experience over a 6-year time

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interval at a tertiary pediatric hospital. Outcomes of interest were presenting symptoms, characteristics, treatment options, and outcomes.

## Materials and Methods

In this retrospective cohort we identified all patients seen in the pediatric and adolescent gynecology service of a tertiary pediatric hospital or in the private practice of a specialist pediatric and adolescent gynecologist. The Royal Children's Hospital provides tertiary pediatric services for a population of 5 million people in Victoria, Australia. Clinic attendance coding was used to search and identify patients who had attended between January 2010 and July 2016 in both settings under the clinical coding category of a vulval symptom. Patients were included if they met diagnostic criteria for vulvodynia as per the latest International Society for the Study of Vulvovaginal Disease, International Society for the Study of Women's Sexual Health, and International Pelvic Pain Society consensus guidelines at the time they were reviewed.[1,6] Patients were excluded if they were older than 18 years of age at the time of diagnosis or had presented with features of vulvovaginitis that resolved after simple medical management (barrier cream, vulval care advice, vinegar baths, worm treatment). Adolescent girls identified because of an inability to insert tampons were excluded if their problem was resolved after correction of a hymenal anomaly.

Data regarding patient age, presenting symptoms, associated symptoms, relevant medical history, clinical findings, and management were extracted from the medical records. After explanation to the patient and her parent/guardian, clinical examination was undertaken. This routinely consisted of external inspection for structural anomalies of the vulva and hymen, followed by examination of the vulval area and vestibule with a cotton tip for altered sensation or hypersensitivity. This was performed in a clock face pattern with specific areas of tenderness documented. Examination techniques of the 4 consultants were similar with yearly Pediatric and Adolescent Gynecology Fellows being trained in this particular technique by 1 of the 4 consultants. Swabs were taken when clinically indicated.

Information was recorded on standardized data sheets with individual files checked by 2 people to ensure consistent data collection. Descriptive statistics were used to present results with Fisher exact test for group comparison. Institutional research and ethics approval for the audit project was granted.

## Results

A total of 47 girls with a median age of 11 years (range, 3-18 years) were identified. The median age of the premenarchal girls was 9 years. The presenting symptoms were varied and are shown in Table 1. Postmenarchal girls were much more likely to present describing pain alone or pain and difficulty inserting tampons compared with premenarchal girls who reported altered sensations such as bubbling sensation or nocturnal exacerbations. The only presenting symptom to reach a statistically significant difference between the 2 groups was difficulty inserting tampons. A history of atopy or anaphylaxis was reported by 14/47 (29.7%).

**Table 1**
Presenting Symptoms

| Symptom | Premenarchal (n = 31), % | Postmenarchal (n = 16), % | *P* (Fisher Exact Test) |
|---|---|---|---|
| Urinary | 25 (80.6) | 10 (62.5) | .289 |
| Nocturnal pain | 6 (19.3) | 0 | .081 |
| Vulval itch | 6 (19.3) | 0 | .081 |
| Trouble inserting tampon | 0 | 6 (37.5) | .0007 |
| Vulval swelling | 2 (6.4) | 0 | .541 |
| Bubbling sensation | 3 (9.6) | 0 | .541 |
| Burning sensation | 1 (3.2) | 2 (12.5) | .264 |
| Dyspareunia | 0 | 1 (6.25) | .340 |

Menstrual exacerbation of symptoms was not a consistent finding. Overall, 7/23 (30.4%) of postmenarchal patients experienced a menstrual flare of pain. Of the 7 premenarchal patients who reached menarche during their follow-up period, 3 reported a sudden increase in vulval pain during menstruation.

Urinary symptoms were the most commonly reported associated symptoms. They were reviewed for character and temporal relationship to pain. Dysuria, urinary frequency, or incontinence were present in 25/31 (80.6%) of premenarchal and 10/16 (62.5%) postmenarchal girls. For more than half of the premenarchal girls, urinary symptoms occurred before the onset of vulval pain. The postmenarchal cohort was much more likely to have pain symptoms present first before the presence of urinary symptoms.

Diagnostic delay was common in this cohort. The median time from onset of symptoms to diagnosis was 1 year. For the premenarchal girls this delay was longer (2.34 years). Additionally, range was greater (0-11 years) including 4 girls who had experienced more than 6 years of diagnostic uncertainty, compared with the postmenarchal girls whose delay to diagnosis was 1.46 years (range, 0-3 years). The longest delay was greater than 11 years in an adolescent who reported symptoms at age 3 years and had been seen by more than 4 clinicians. The timeframe for follow-up of our cohort ranged from 0 to 6 years (median follow-up, 1 year).

The appearance of the external genitalia was normal in most patients. In premenarchal patients, 8/31 (25.8%) had mild erythema and 5/31 (16.1%) had labial adhesions (Table 2). Examination with cotton tip revealed vestibulodynia in 16/17 (94.1%) and 11/13 (84.6%) of pre- and postmenarchal patients, respectively. Postmenarchal girls were

**Table 2**
Examination Findings

| Finding | Premenarchal, n (%) | Postmenarchal, n (%) | *P* (Fisher Exact Test) |
|---|---|---|---|
| Normal appearance | 17 (54.8) | 14 (87.5) | .049 |
| Pain on cotton tip testing | 16 (94.1) | 11 (84.6) | .354 |
| Mild erythema | 8 (25.8) | 0 | .038 |
| Hymenal anomaly | 1 (3.2) | 4 (25) | .039 |
| Adhesions | 5 (16.1) | 0 | .150 |
| Skin changes | 0 | 0 | 1 |

**Table 3**
Management Strategies

| Management | Premenarchal, n (%) | Postmenarchal, n (%) |
|---|---|---|
| Neuromodulation | 29 (93.5) | 15 (93.7) |
| Regular physiotherapy | 2 (6.4) | 4 (25) |
| Surgery (hymenectomy) | 0 | 4 (25) |
| Oxybutynin | 2 (6.4) | 2 (12.5) |

**Table 4**
Clinical Status at Last Review

| Symptomatology | Premenarchal, n (%) | Postmenarchal, n (%) | *P* (Fisher Exact Test) |
|---|---|---|---|
| Improved | 25 (80.6) | 14 (87.5) | .697 |
| Unchanged | 6 (18.3) | 2 (12.5) | .697 |
| Ceased or weaning medication | 12 (38.7) | 5 (31.2) | .752 |

more likely to have normal appearing external genitalia ($P = .049$) whereas premenarchal patients often had mild erythema ($P = .38$).

Six patients presented with an inability to use tampons secondary to pain or trouble passing the introitus. Four of these patients were found to have structural hymen anomalies on clinical examination (septate hymen, n = 1; annular hymen, n = 3). Each of these young women underwent corrective surgery. Despite surgery, these adolescents had ongoing perihymenal pain or vestibulodynia requiring additional management in the form of physiotherapy and neuromodulation (amitriptyline).

Treatment strategies within our cohort are outlined in Table 3. Neuromodulation was achieved using amitriptyline, gabapentin, pregabalin, or imipramine (occasionally in combination). The most commonly used agent within our service was amitriptyline with doses ranging from 5 to 40 mg every night. Physiotherapy was more commonly used in the postmenarchal group and multiple-agent medical treatment was equal between the 2 groups. Table 4 shows clinical status at the most recent review. Most had improvement or complete resolution of symptoms (25/31 (80.6%) premenarchal, 14/16 (87.5%) postmenarchal). Both groups had equal likelihood of clinical improvement with *P* values failing to reach statistical significance. Neuromodulator treatment had been ceased or weaned at last review in 12/31 (38.7%) and 5/16 (31.2%) of pre- and postmenarchal girls, respectively.

## Discussion

Vulval and vestibular pain have been recognized for many decades, in women of reproductive and postmenopausal ages, yet the etiology and clear and effective management pathways remain poorly defined.[7] This clinical problem does occur in the younger populations but has not been well described, apart from a small series of 6 prepubertal girls[8] and the mention of a single case in an audit of the experiences of a pediatric vulval clinic where more than 100 girls had been seen.[9] In a slightly older population of sexually active 12- to 19-year-old girls, 20% (n = 251) were reported to have severe pain with first tampon use.[10] It is likely that this cohort reflects a population of unrecognized vulvodynia.

Previous studies have identified that there is a considerable delay in diagnosis for many adult patients with vulval pain. A National Institutes of Health-funded study in 2003 to estimate the prevalence of chronic vulval pain showed that 40% of women remained without a diagnosis after 3 consultations and 60% had seen at least 3 clinicians before formal diagnosis.[11] They reported an average delay in diagnosis of more than 2 years with some patients tolerating symptoms for 11 years. The timelines to diagnosis in our younger cohort are not dissimilar, which is somewhat surprising in view of the lack of medical knowledge and literature about this clinical problem in the younger age group.

One clear association shown in this review is urinary symptomatology presenting before or concurrently with vulval pain. This association was described in adult patients in whom 26.6% of adults with vulvodynia report a history of childhood enuresis.[12] The small report of 6 prepubertal girls over a 10-year time span by Reed and Cantor also reported this association in 2 of the 6 girls studied.[8] The coexistence of vulval pain and urinary tract symptomatology has been reported a number of times.[5,13,14] Acknowledgement of this association is made in the consensus terminology and classification statement from 2015, which formally described painful bladder syndrome as a possible factor associated with vulvodynia.[1] It seems this association holds true for younger women and girls.

The apparent association with atopy seen in 14/47 (29.7%) of the cohort might reflect a predisposition or simply the prevalence of these conditions. Previous studies have quoted a prevalence of 30% within the general population.[15] It is an association that might benefit from additional study in larger cohorts.

The limitations of this study are those inherent to the retrospective design including differences in practice between care providers and incomplete data entry. There is potential for examination technique differences among the providers reviewing these patients. The strengths of this cohort come from the large referral base yielding large numbers within the clinic and a predominance of complex cases.

Although vulval pain has a heterogeneous and complex pathophysiology, we have found many common themes in the presentation and natural history of vulvodynia in young women and children. This population is vulnerable to long-term sexual dysfunction and body image issues. We have shown that the symptoms often exist far earlier than had been previously described. If diagnosed and appropriately managed early, this represents a unique opportunity to address the pain before long-term issues arise.

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