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## BMJ Paediatrics Open

### Maternal attitude towards delaying normal/precocious puberty in girls with/without disability

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Complete List of Authors:	Deeb, Asma; Mafraq Hospital, Paediatric Endocrinology Akle, Mariette; Paediatric Endocrinology Al Zaabi, Abrar; Mafraq Hospital, Paediatric Department Siwji, Zohra; Mafraq Hospital, Paediatric Department Attia, Salima; Mafraq Hospital, Paediatric Endocrinology Al Suwaidi, Hana; Mafraq Hospital, Paediatric Endocrinology Al Qahtani, Nabras; Mafraq Hospital, Paediatric Endocrinology Ehtisham, Sarah; Mediclinic City Hospital, Paediatric Endocrinology
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Manuscripts

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3 **Maternal attitude towards delaying puberty in girls with and without disability presenting**  
4 **with normal or precocious puberty**  
5

6 *Asma Deeb<sup>1</sup>, Mariette Akle<sup>1</sup>, Abrar Al Zaabi<sup>1</sup>, Zohra Siwji<sup>1</sup>, Salima Attia<sup>1</sup>, Hana Al Suwaidi<sup>1</sup>,*  
7 *Nabras Al Qahtani<sup>1</sup> & Sarah Ehtisham<sup>2</sup>*  
8  
9

10  
11 1) Paediatric Endocrinology Department  
12 Mafrag Hospital  
13 PO Box 2951  
14 T + 971-50-8350568  
15 F +971-2-  
16 Abu Dhabi, United Arab Emirates  
17  
18

19 Asma Deeb (corresponding author)

20 [adeeb@seha.ae](mailto:adeeb@seha.ae)  
21

22 Mariette Akle  
23

24 [makle@seha.ae](mailto:makle@seha.ae)  
25

26 Abrar Al Zaabi  
27

28 [abzaabi@seha.ae](mailto:abzaabi@seha.ae)  
29

30 Zohra Siwji  
31

32 [zoali@seha.ae](mailto:zoali@seha.ae)  
33

34 Salima Attia  
35

36 [sabood@seha.ae](mailto:sabood@seha.ae)  
37

38 Hanah Al Suwaidi  
39

40 [hsuwaidi@seha.ae](mailto:hsuwaidi@seha.ae)  
41

42 Nabras Al Qahtani  
43

44 [nmqahtani@seha.ae](mailto:nmqahtani@seha.ae)  
45

46 2) Mediclinic City Hospital  
47 Dubai Healthcare City  
48 PO Box 505004  
49 T +97144355644  
50 F +97144359900  
51 Dubai, United Arab Emirates  
52  
53

54 - Sarah Ehtisham  
55  
56  
57  
58  
59  
60

1  
2  
3 [sarah.ehtisham@mediclinic.ae](mailto:sarah.ehtisham@mediclinic.ae)  
4

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**What is already known about the subject**

- Puberty suppression may be sought for girls with disability for improving quality of life.
- Gonadotropin analogs are an option for puberty suppression in normal and girls with disability.
- Surgical intervention for permanent puberty suppression is occasionally considered by families of girls with disability.

**What this study adds about the subject**

- Mothers of normal and girls with disability have different reasons for seeking advice on puberty suppression.
- Cultural and religious issues might affect attitude of mothers on puberty suppression.
- Difficulty of hygiene management during menstruation and fear of short adult height are the commonest reasons for seeking puberty suppression in normal and girls with disability respectively.

## Abstract

**Background:** Children with neurologic impairment are at a risk of precocious puberty (PP). There is a parental anxiety regarding the impact of menses, particularly in girls with severe disability. Therapeutic puberty suppression is sought to reduce morbidity and improve quality of life for patients and caregivers. We aim to explore maternal attitude and reasons for seeking puberty suppression.

**Methods:** Mothers of girls on gonadotropin analogs were approached to enroll in the study. Participants were interviewed and asked to fill in a questionnaire detailing possible reasons for delaying puberty. Medical information pertaining to the underlying etiology for disability, details of puberty presentation and investigations were collected. The study group was divided into girls with disability presenting with PP or normal puberty and girls with no disability presenting with PP. The study was approved by Mafraq Research and Ethics committee.

**Results:** 42 mothers were enrolled. 15 had normal girls with central PP. 27 girls had a degree of disability. 10 presented with PP. Most parents aimed to delay the puberty, commonly sought by mothers with normal girls, while 56% of mothers desired to completely halt puberty in girls with disability. 7% wanted surgical intervention. Fear of short stature (100%), inability to psychologically cope with puberty (67%) and fear of peer rejection (60%) were the main concerns for the mothers with normal girls. Mothers with girls with disability were mainly concerned about hygiene management during menstruation (92.5%), fear of child abuse or unwanted pregnancy (55%) and fear of inability of expressing pain/discomfort associated with menstruation (30%).

**Conclusion:** PP and pubertal changes in girls with disability is associated with major concerns. Mothers of girls with disability commonly seek medical help to delay/halt puberty due to hygiene management during menstruation, while short final height is the main concern for mothers of normal girls with PP. Culture & religion play an important role in puberty management in girls with disability.

## Introduction

Disability has a major impact on the affected child and families. A Census report from the USA showed that there are over 5 million children under 18 years have disabilities (1). The endocrine system is highly affected by disability with impacts on the hypothalamic pituitary function, bone health, pubertal timing, sexual function and fertility. Accordingly, assessment of pubertal changes should be an essential component of the multidisciplinary care for children with neuro disability (2). In this group of children, puberty poses various challenges particularly in relation to management of menstruation. It is reported that families of 50% of girls with disabilities sought medical advice in relation to menstruation management (3).

People with neurologic impairment have a higher risk of going into precocious puberty (PP). Girls with cerebral palsy undergo puberty earlier, but they reach menarche slightly later than the general population (4). Presentation for counselling by families of adolescents with disabilities is often sought before or after menarche (5).

Mothers' attitude towards their daughters' early puberty is influenced by the maturity and the cognitive ability of the girl. While issues of physical care related to puberty changes might be prominent in girls with disability, other factors might be more of a concern in girls with normal mental development and ability. There is a high level of parental anxiety regarding the impact of menses, particularly on girls with severe disability (3). Increase in linear growth is a major component of puberty and it is well known that untreated children with PP end up with short adult height. The height concern might be different when compared in normal children and others with disability. Parents and care-givers occasionally approach health care professionals for permanent surgical measures to abolish puberty, which raises ethical dilemmas if the patient is not able to participate fully in consent (6).

Treating girls with central PP can alleviate psychological distress and delay menstruation (7). Use of gonadotropin analogs to treat Central PP is proven to be safe and effective (8). Advancing puberty and onset of menstruation can raise clinical dilemmas associated with hormonal intervention either to delay onset of menstruation or to reduce the amount of bleeding. Although various hormonal modalities are available for such a purpose, each of these modalities have

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3 advantages and disadvantages. Individualized choice decided with care for particular patient is of  
4 paramount importance (4, 9). It is recommended that menstrual suppression therapy should not  
5 commence until menarche is achieved (10, 11). Awaiting menarche confirms normal hormonal  
6 function and rule out any obstructive anomaly. In good medical practice, the ultimate goal of  
7 menstrual suppression is to reduce morbidity and improve quality of life both for the girls and for  
8 their caregivers (12). In our region where mothers of girls with disability are helped by male  
9 family members, there are major issues of difficulties in exposing the girl to her helpers. This  
10 stems from both cultural and religious reasons and might be a reason for seeking help to delay or  
11 abolish puberty (personal observation).  
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### 20 **Aim of the study**

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22 We aim to study maternal attitude towards daughters' puberty. We also plan to explore the  
23 reasons for seeking medical help to delay puberty in normal and girls with disability who  
24 presented with normal or precocious puberty.  
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## Patients & Method

The study was undertaken at the Paediatric Endocrinology department of Mafraq Hospital. A list of girls receiving gonadotropin analogs therapy between 2013-2016 for delaying puberty was obtained from the clinic database. The list obtained from the clinic database was cross-checked with the hospital pharmacy electronic records for gonadotropin analogs dispensing.

Mothers of girls on gonadotropin analogs therapy were approached to participate in the study during one of their daughters scheduled visits for gonadotropin analogs injection. The study was explained to the mothers and information sheets were handed to them. Mothers who agreed to participate in the study had their daughters enrolled. The study was approved by Mafraq hospital Research and Ethics committee.

Mothers were interviewed by a clinical psychologist (MA) in one to one setting. A questionnaire detailing various possible reasons for delaying puberty was filled during the interview (Appendix). Mothers were asked to suggest other reasons for seeking puberty suppression if their reasoning was not included in the list.

Medical information were collected by the study team including: detailed underlying medical condition for girls with disability, cause of PP in normal girls (if established), age at puberty presentation, signs of first puberty signs, initial puberty-related biochemistry and radiology tests.

### Inclusion Criteria:

**The following group of girls on gonadotropin analogs were eligible for enrollment:**

- Normal girls with central PP
- Girls with disability presenting with central PP
- Girls with disability presenting with normal puberty

## Results

42 mothers with daughters receiving puberty suppression participated in the study. 27 of the girls had a degree of physical/mental disability and 15 were normal. The underlying causes of disability in the 27 girls were cerebral palsy, spina bifida, Arnold Chiari malformation, hydrocephalus with shunt, microcephaly, hemimegalencephaly, Biotin deficiency, Rett's syndrome, cerebral dysgenesis, ataxia, intractable seizure and idiopathic developmental delay with seizures. All normal girls presented with PP and were diagnosed with idiopathic central PP after performing full investigations including brain MRI scans.

Mean (range) age of the normal girls at enrolment was 9.6 years (7-13). All of them had PP with a mean (range) of presentation of 6.3 years (5.5-7.8) and were on gonadotropin analogs for a period of 2.2 years (0.2- 5.4). Of the 27 girls with disability, 10 presented with PP. Mean (range) of start of puberty was 8.1 years (3-13) and have been on gonadotropin analogs for a mean of 1.8 years (0.3-5).

On the interview, all mothers of the normal girls intended to delay their puberty while 15 (56%) of mothers of girls with disability expressed the wish to stop the puberty. While none of the normal girls' mother considered surgical intervention for stopping puberty, 7 mothers (26%) considered surgical intervention (hysterectomy) to prevent puberty permanently (Table 1).

**Table 1**

<b>Variable</b>	<b>Normal Girls</b>	<b>Girls with disability</b>
<b>Number (total number)</b>	15	27
<b>Number with PP</b>	15	10
<b>Mean (range) of age at enrollment (yrs)</b>	9.6 (7-13)	9.5 (5-14)
<b>Mean (range) of age at onset of puberty (yrs)</b>	6.3 (5.5-7.8)	8.1 (3-13)
<b>Mean (range) duration of treatment (yrs)</b>	2.2 (0.2-5.4)	1.8 (0.3-5)
<b>Number of mothers intending to delay puberty</b>	15 (100%)	12 (44%)
<b>Number of mothers intending to stop puberty</b>	0	15 (56%)

<b>Number of mothers considering surgical intervention to stop puberty</b>	0	7 (26%)
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Regarding the reasons mother gave for puberty delaying, fear of short adult height was the main one given by mothers of normal girl while none of the mothers of girls with disability had height as a concern. Inability to cope with puberty psychologically was the reason in 67% of the normal girl while it was only a cause for concerns in 18.5% in mothers of girls with disability. 60% of mothers of normal girls feared peer rejection for their daughters which was not the case in girls with disability. Concern about hygiene management during menstruation was the main concern which was shared by 92.5% of mothers of girl with disability while it was raised by only a quarter of mothers with normal girls. Fear of abnormal sexual and emotional behavior was raised by both categories of mothers. However, it was highly rated by mothers of girls with disability as a reason to delay puberty compared to the other group. Over half of mothers with girls with disability feared sexual abuse or unwanted pregnancy in their daughters while it was not a major concern in mothers of normal girls.

Inability to express menstrual pain, possible aggravation of convulsions or chest pain due to wheel chair strapping on the developing breast were all expressed by mothers of girls with disability while none was applicable to the normal girls. A quarter of mothers of girls with disability expressed concern about the difficulty of seeking male family member help if the girl shows progressive signs of puberty due to cultural and religious reasons (Table 2).

**Table 2**

<b>Reason for delaying/stopping puberty</b>	<b>Number</b>	
	<b>Girls with no disability</b>	<b>Girls with disability</b>
Fear of short adult height	<b>15 (100%)</b> <i>1</i>	<b>0</b>
Inability to cope with puberty changes psychologically	<b>10 (67%)</b> <i>2</i>	<b>5 (18.5%)</b> <i>7</i>
Fear of peer rejection	<b>9 (60%)</b> <i>3</i>	<b>0</b>

Hygiene management during menstruation	<b>4 (27%)</b> 4	<b>25 (92.5)</b> 1
Fear of abnormal sexual or emotional behavior	<b>3 (20%)</b> 5	<b>11 (41%)</b> 4
Fear of child abuse or unwanted pregnancy	<b>2 (13%)</b> 6	<b>15 (55.5)</b> 3
Difficulties in maintaining hygiene in dependent girls with disability	N/A	<b>23 (85%)</b> 2
Menstrual pain that cannot be properly expressed	N/A	<b>8 (30%)</b> 5
Difficulty of seeking male family member help	N/A	<b>7 (26%)</b> 6
Effect on convulsion frequency	N/A	<b>5 (18.5%)</b> 7
Chest pain due to wheelchair harness on chest	N/A	<b>3 (11%)</b> 8

## Discussion

Parents of girls with early puberty are reported to be worried, embarrassed and concerned about their daughters' inability to cope with early sexual development and possible peer rejection (7). It is reported that girls who mature earlier than their peers find pubertal adjustment more challenging. Those girls appear to go through various psychological difficulties and experience detrimental sequelae (13). Stice et al reported that girls who had PP were at 1.9 times risk for depression (14). In our cohort, two thirds of the mothers interviewed expressed concern that their daughters might not be able to cope with puberty changes psychologically and might face peer rejection. This concern, however, was not major in mothers of girls with disability.

A newly-observed irritability or discomfort might be noted by parents/carers of girls with disability who wear harness while in wheel chairs. This could be caused by chest tenderness with the harness impinging on the developing breast tissue (15). With the girl being unable to express the pain properly, this can be rarely recognized by families. We had 3 mothers who had this issue as a major concern when their daughters started early signs of breast development and 8 mothers expressed concerns that daughters might not be able to express pain associated with menstruation.

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3 There is a high level of parental anxiety regarding the impact of menses, particularly when  
4 disability is severe (3). The feared impact can be an indirect effect on the child's medical  
5 condition like frequency of convulsions for example or a direct effect of the puberty  
6 consequences. The latter mainly includes hygiene issues for girls with disability, menstrual  
7 irregularities, premenstrual symptoms, cyclical pain, which might not be properly expressed,  
8 unwanted sexual contact or pregnancy (16. 17). These issues collectively were more pronounced  
9 in girls with disability in our cohort. Around 85% of mothers feared difficulty in maintaining  
10 hygiene in daughters with disability during menstruation. Fear of inability of expressing  
11 pre/menstrual pain was expressed by about a third of mothers and 18% of them were concerned  
12 about increased convulsion frequency with progression of puberty.  
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21 Less severely disabled children may have deteriorated behavior and demonstrate socially-  
22 inappropriate sexual behavior like masturbation at an early age raising family concern about risk  
23 of sexual contact (15). Abnormal sexual behavior was an issue of concern in 20% of mothers  
24 with normal girl but it was higher at 40% of mothers with girls with disability in our study group.  
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29 Increase in linear growth is a major component of puberty. It is well known that untreated  
30 children with PP end up with short adult height. While, final adult height might not be a concern  
31 for families with disabled children, rapid growth associated with puberty might have an impact  
32 on the family and the child with disability. The impact arises from a possible need of changing  
33 medical equipment, such as provision of an adequately sized wheelchair, progression of scoliosis  
34 bracing or altering the size for chest braces or ankle-foot orthoses (15). All mothers of normal  
35 girls in our study group put the growth issue as a priority when medical advice for puberty delay  
36 was sought. Interestingly, none of the mothers with girls with disability felt that the rapid growth  
37 was an issue.  
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46 Parent and care-givers occasionally approach health care professionals for permanent surgical  
47 measures to abolish puberty, which raises ethical dilemmas if the patient is not able to participate  
48 fully in consent (6). Surgical intervention (hysterectomy) was a measure that was considered by  
49 a quarter of mothers of girls with disability in our group.  
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3 In our region where mothers of girls with disability are helped by male family members, there  
4 are significant issues of difficulties in exposing the girl with disability to her male helpers. This  
5 stems from both cultural and religious reasons and might be a reason for seeking help to delay or  
6 abolish puberty (personal observation). With the girl growing, physical help is required for  
7 various daily routines. Involving a male family member to participate in helping on those daily  
8 routine results in exposure of the growing girl which might not be acceptable by the mother nor  
9 the male helper. 26% of mothers interviewed expressed this issue as a major concern.

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11 Use of gonadotropin analogs to treat central PP is proven to be safe and effective (8). However,  
12 they are relatively expensive when compared with other treatments used for the similar  
13 indication of delaying or abolishing menstruation and mostly not covered by insurance. In  
14 addition, it is an injectable form of medication which is often a traumatic experience and can  
15 cause sterile abscesses. In our cohort, one girl developed an abscess at the site injection which  
16 subsided and did not recur on further injections. Treatment with gonadotropin analogs require  
17 attention to dosage and period of injection to avoid intermittent bleeding. 2 girls in our group had  
18 vaginal spotting at the start of treatment. Another girl had intermittent bleeding during the first 9  
19 months of treatment that had subsided. Overall, gonadotropin analogs are found to be an  
20 acceptable form of treatment in our group particularly with the low frequency of injection by  
21 using the depot form. However, it is a challenge to persuade the mothers to stop the medicine  
22 after the treatment period is completed to avoid long term side effects. Of those side effects, the  
23 concern about the effect of gonadotropin analogs on bone mineral density is often raised.  
24 Although data suggests that reduction in bone mineral density is reversed and its peak at final  
25 height is unaffected (18), it is recommended to avoid the long-term use of puberty antagonists  
26 (11).

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28 Our study shows that early pubertal changes are associated with major concerns. Mothers of girls  
29 with disability seek medical help to delay puberty due to various reasons, particularly, fear of the  
30 difficulty in keeping hygiene during menstruation. Short adult height is the main concern  
31 expressed by mothers of normal girls going into PP. Culture and religion have an impact on  
32 decision making of delaying puberty and some families do consider surgical measures to stop  
33 puberty in girls with disability.

### Authors' contribution

Asma Deeb designed the study, applied for ethical approval and wrote the manuscript. Mariette Akle designed the questionnaire and interviewed the study participants. Abrar Al Zaabi and Zohra Siwji collected patients' data and collated the results. Salima Attia, Hana Al Suwaidi and Nabras Al Qahtani are clinicians who recruited the study subjects. Sarah Ehtisham revised the study proposal, advised on the questionnaire design and revised the manuscript.

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5 **Table legends**  
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7 **Table 1:** Characteristics of study population

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9 **Table 2:** Reasons for seeking puberty suppression by mothers in girls with disability. Numbers  
10 in italic under percentage indicates the order of frequency of the choice  
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## References

- 1) Brault Matthew. Americans With Disabilities: Household Economic Studies. Current Population Reports 2010;P70-131.
- 2) Mithyantha R, Bassi Z. Early Pubertal Changes in Neurodisabled Children. An under-diagnosed issue? Arch dis child 2013;98:A53.
- 3) Zacharin M. The Impact of Menstruation in Adolescents with Disabilities Related to Cerebral Palsy. Arch Dis Child 2010;95(7):526-530.
- 4) Albanese A, Hopper NW. Suppression of menstruation in adolescents with severe learning disabilities. Arch Dis Child 2007;92:629–632.
- 5) Zacharin MR: Puberty, contraception, and hormonal management for young people with disabilities. Clin Pediatr 2009;48:149–155.
- 6) Isaacs D, Tobin B, Hamblin J, Slaytor E, Donaghue KC, Munns C, Kilham HA: Managing ethically questionable parental requests: growth suppression and manipulation of puberty. J Paediatr Child Health 2011;47:581–584.
- 7) MazurT, Clopper RR. Pubertal disorders: Psychology and clinical management. Endocrinology and Metabolism Clinics of North America 1991;20(1):211-230.
- 8) Carel JC, Eugster EA, Rogol A. Consensus statement on the use of gonadotropin-releasing hormone analogs in children. Pediatrics 2009;123(4):e752-62.
- 9) Van Schroyen Lantman-de Valk HM, Rook F, Maaskant MA: The use of contraception by women with intellectual disabilities. J Intellect Disabil Res 2011;55(4):434-440.
- 10) Dizon C, Allen L, Ornstein M. Menstrual and contraceptive issues among young women with developmental delay: a retrospective review of cases at the Hospital for Sick Children, Toronto. J Pediatr Adolesc Gynecol 2005;18:157–162.
- 11) Kirkham Yolanda, Ornstein M, Aggarwal Anjali, McQuillan S et al. Menstrual suppression in special circumstances: SOGC clinical practice guidelines 2014;36(10):915–924.
- 12) Guilbert, E., Boroditsky, R., Black, A., Kives, S., Leboeuf, M., Mirosh, M. et al. Society of Obstetricians and Gynaecologists of Canada. Canadian consensus guideline on continuous and extended hormonal contraception. SOGC Clinical Practice Guidelines. J Obstet Gynaecol Can 2007;29: S1–S32.

- 13) Ge X, Conger RD, Elder GH. Coming of age too early: pubertal influences on girls' vulnerability to psychological distress. *Child Development* 1996;67:3386–3400.
- 14) Stice E, Presnell K, Bearman SK. Relation of early menarche to depression, eating disorders, substance abuse, and comorbid psychopathology among adolescent girls. *Developmental Psychology* 2001;37:608–619.
- 15) Zacharin M. Endocrine Problems in Children and Adolescents Who Have Disabilities. *Horm Res Paediatr* 2013;80:221–228.
- 16) Quint EH: Menstrual issues in adolescents with physical and developmental disabilities. *Ann NY Acad Sci* 2008;1135:230-236.
- 17) Grover SR: Gynaecological issues in adolescents with disability. *J Paediatr Child Health* 2011; 47:610-613.
- 18) van der Sluis IM, Boot AM, Krenning EP, et al. Longitudinal follow-up of bone density and body composition in children with precocious or early puberty before, during and after cessation of GnRH agonist therapy. *J Clin Endocrinol Metab* 2002;87(2):506–512.

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## Appendix: Questionnaire filled on interviewing mothers by clinical psychologist

### Subjects categories

- Normal girl with precocious puberty
- Girl with disability presenting with precocious puberty
- Girl with disability presenting with normal puberty

#### 1) What was the main reason/reasons you wanted to delay or stop puberty

- a) Fear of short adult height
- b) Fear of peer rejection
- c) Fear of abnormal sexual or emotional behavior
- d) Inability to cope with puberty changes psychologically
- e) Difficulties in maintaining hygiene for dependent girls with disability
- f) Fear of a vulnerable child abuse or unwanted pregnancy
- g) Menstrual pain that cannot be properly expressed
- h) Effect on convulsion frequency
- i) Chest pain due to wheelchair harness on chest
- j) Difficulty of seeking male family member help
- k) Hygiene management during menstruation
- l) Other reasons. Please list

#### 2) Was your intention of treatment to delay or stop puberty permanently

- Delay
- Stop

#### 3) If to delay, for how long you wanted to delay the puberty?

- Short period (few months)
- Long period (years)
- According to doctors' advice

#### 4) Would you consider permanent surgical measures like hysterectomy to stop puberty?

- Yes
- No

## BMJ Paediatrics Open

### Maternal attitude towards delaying puberty in girls with and without disability; a questionnaire-based study

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Complete List of Authors:	Deeb, Asma; Mafraq Hospital, Paediatric Endocrinology Akle, Mariette; Mafraq Hospital, Paediatric Endocrinology Al Zaabi, Abrar; Mafraq Hospital, Paediatric Department Siwji, Zohra; Mafraq Hospital, Paediatric Department Attia, Salima; Mafraq Hospital, Paediatric Endocrinology Al Suwaidi, Hana; Mafraq Hospital, Paediatric Endocrinology Al Qahtani, Nabras; Mafraq Hospital, Paediatric Endocrinology Ehtisham, Sarah; Mediclinic City Hospital, Paediatric Endocrinology
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3 **Maternal attitude towards delaying puberty in girls with and without disability; a**  
4 **questionnaire-based study**  
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7 *Asma Deeb<sup>1</sup>, Mariette Akle<sup>1</sup>, Abrar Al Zaabi<sup>1</sup>, Zohra Siwji<sup>1</sup>, Salima Attia<sup>1</sup>, Hana Al Suwaidi<sup>1</sup>,*  
8 *Nabras Al Qahtani<sup>1</sup> & Sarah Ehtisham<sup>2</sup>*  
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11  
12 1) Paediatric Endocrinology Department  
13 Mafraq Hospital  
14 PO Box 2951  
15 T + 971-50-8350568  
16 F +971-2-  
17 Abu Dhabi, United Arab Emirates  
18  
19

20 Asma Deeb (corresponding author)

21 [adeeb@seha.ae](mailto:adeeb@seha.ae)

22 Mariette Akle

23 [makle@seha.ae](mailto:makle@seha.ae)

24 Abrar Al Zaabi

25 [abzaabi@seha.ae](mailto:abzaabi@seha.ae)

26 Zohra Siwji

27 [zoali@seha.ae](mailto:zoali@seha.ae)

28 Salima Attia

29 [sabood@seha.ae](mailto:sabood@seha.ae)

30 Hanah Al Suwaidi

31 [hsuwaidi@seha.ae](mailto:hsuwaidi@seha.ae)

32 Nabras Al Qahtani

33 [nmqahtani@seha.ae](mailto:nmqahtani@seha.ae)

34  
35 2) Mediclinic City Hospital  
36 Dubai Healthcare City  
37 PO Box 505004  
38 T +97144355644  
39 F +97144359900  
40 Dubai, United Arab Emirates  
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43 - Sarah Ehtisham  
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2  
3 [sarah.ehtisham@mediclinic.ae](mailto:sarah.ehtisham@mediclinic.ae)  
4

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### What is already known about the subject

- Puberty suppression may be sought for girls with disability for improving quality of life.
- Gonadotropin releasing hormone analogue (GnRHa) therapy is an option for pubertal suppression in girls with and without disability.
- Surgical intervention for permanent suppression of menstruation is occasionally considered by families of girls with disability.

### What this study adds about the subject

- Mothers of girls with and without disability have different reasons for seeking advice on puberty suppression.
- Cultural and religious issues may influence the attitude of mothers on puberty suppression.
- Concerns about menstrual hygiene and fear of short adult height are the commonest reasons for seeking puberty suppression in girls and without with disability respectively.

## Abstract

**Background:** Parental anxiety about the impact of puberty/menses, particularly in girls with severe disability leads to seeking therapeutic pubertal suppression. We aim to explore maternal attitudes and reasons for seeking pubertal suppression.

**Methods:** Mothers of girls receiving GnRHa therapy were enrolled in the study. A semi-structured interview was conducted to ascertain possible reasons for delaying puberty. The study group was divided into girls with disability with central precocious puberty (CPP) or normal puberty and girls without disability presenting with CPP.

**Results:** 42 mother-daughter pairs were enrolled and divided into 2 groups; Group A: 15 girls with CPP with no disability. Group B: 27 girls with of disability (10 had CPP (Group B1) and 17 had normal pubertal timing (Group B2). Mothers in Group A aimed to delay puberty, whilst in Group B, 13 (48%) of mothers desired to halt puberty and 7 (26%) requested permanent surgical intervention. Fear of short stature (15, 100%), inability to cope psychologically (10, 67%) and fear of peer rejection (9, 60%) were the main concerns in Group A. In Group B, mothers were concerned about menstrual hygiene management (25, 92.5%), fear of child abuse or unwanted pregnancy (15, 55%) and fear of inability to express pain/discomfort with menstruation (8, 30%).

**Conclusion:** Mothers of girls with disability commonly seek medical help to delay/halt puberty due to concerns about menstrual hygiene. Short final height is the main concern for girls without disability. Culture & religion play an important role in puberty management in girls with disability.

## Introduction

Childhood disability has a major impact on the affected child and families. A Census report from the USA showed that there are over 5 million children under 18 years have disabilities (1). The endocrine system is highly affected by disability with effects on the hypothalamic pituitary function, bone health, pubertal timing, sexual function and fertility. Accordingly, assessment of pubertal changes should be an essential component of the multidisciplinary care for children with disability (2). In this group of children, puberty poses various challenges particularly in relation to management of menstruation. It is reported that 50% of families of girls with disabilities seek medical advice in relation to menstruation management (3).

Children with neurologic impairment have a higher risk of central precocious puberty (CPP). Girls with cerebral palsy undergo puberty earlier but reach menarche slightly later than the general population (4). Families of adolescents with disabilities can present for counselling before or after menarche (5).

Maternal attitudes towards their daughters' early puberty is influenced by the maturity and the cognitive ability of the girl. While physical care issues related to puberty can be prominent in girls with disability, other factors might be of more concerns in girls with normal mental development. There is a high level of parental anxiety regarding the impact of menses, particularly in girls with severe disability (3). Increase in linear growth is a major component of puberty and it is well known that untreated CPP is detrimental to adult height. Parental concerns about growth may be different in children with and without disability. Parents and care-givers occasionally approach health care professionals requesting permanent surgical measures to abolish puberty, which raises ethical dilemmas if the patient is not able to participate fully in consent (6).

Treating girls with CPP can alleviate psychological distress and delay menstruation (7). Use of gonadotropin releasing hormone analogue (GnRHa) therapy to treat CPP is proven to be safe and effective (8). Advancing puberty and onset of menstruation can raise clinical dilemmas associated with hormonal intervention either to delay onset of menstruation or to reduce the menstrual flow. Although various non-hormonal and hormonal interventions are available for

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3 such a purpose, each modality of treatment has advantages and disadvantages. It is of paramount  
4 importance to individualise the treatment decisions considering each child's circumstances (4, 9).  
5 It is recommended that menstrual suppression therapy should not commence until menarche is  
6 achieved (10, 11). However, in certain circumstances, delaying puberty and menstrual  
7 suppression can reduce morbidity and improve quality of life both for the girls and for their  
8 caregivers (12).  
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15 In the United Arab Emirate and some other Arab and Asian countries, mothers are the main  
16 carers of girls with disability and are more commonly involved in hospital visits and treatment  
17 decisions than fathers (personal observation). In our region, pubertal changes in a girl with  
18 disabilities can pose significant issues when male family members and helpers are involved in  
19 personal care. This stems from both cultural and religious reasons and may contribute to reasons  
20 for seeking help to delay or suppress puberty (personal observation).  
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26 Understanding the maternal attitude and concerns towards puberty enables health care  
27 professionals to provide appropriate and culture-sensitive solutions. Highlighting specific issues  
28 in a population helps to improve quality of life and maternal satisfaction through provision of  
29 appropriate measures.  
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### 34 **Aims of the study**

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37 We aimed to explore maternal attitudes towards their daughters' puberty. We also planned to  
38 explore the reasons for seeking medical help to delay puberty in girls with and without disability  
39 who presented with normal or precocious puberty.  
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## Patients & Methods

The study was undertaken at the Paediatric Endocrinology department of Mafraq Hospital. A list of girls receiving gonadotropin releasing hormone analogue (GnRHa) therapy for delaying puberty between 2013-2016 was compiled from the clinic database. This list was cross-checked with the hospital pharmacy electronic records for dispensing of GnRHa. Mothers of girls on GnRHa therapy were approached to participate in the study during one of their daughters' scheduled visits for GnRHa injection. The study was explained to the mothers and information sheets were given. Mothers who agreed to participate in the study gave a verbal consent and had their daughters enrolled. The study was approved by Mafraq hospital Research and Ethics committee.

The mothers had a semi-structured interview by a clinical psychologist (MA) in a one to one setting. A questionnaire was designed to include possible reasons for intervening in puberty. The multiple choice of reasons was derived from a literature search on the subject and reflected study team own observation and cultural background of the cohort. The questionnaire was completed during the interview (Appendix). The mothers were asked to suggest other reasons for seeking pubertal suppression if their reason was not included in the list.

Medical information was collected from the electronic medical record by the study team including: detailed underlying medical condition for girls with disability, cause of CPP (if established), age at puberty presentation, the initial signs of puberty, and the results of the initial biochemistry and radiology tests.

No formal power calculation was performed. The intention was to approach all eligible patients and enrol all who consent. All girls receiving GnRHa therapy for puberty were eligible for enrolment and they were subdivided into the following groups for analysis:

- Group A: Girls with CPP without disability
- Group B: Girls with disability – subdivided into Group B1 with CPP and Group B2 with normal puberty

## Results

46 girls receiving GnRHa therapy were identified in the medical and hospital pharmacy records. 42 mothers consented to the study and their daughters were enrolled. All the families were Muslim, 19 were Emirati, 18 from other Arab countries and 5 were Indian or Pakistani. 15 of the girls had no disability (Group A). All presented with CPP and were diagnosed with idiopathic CPP after performing full investigations including brain MRI scans. 27 of the girls had a degree of physical/intellectual disability (Group B) with the following underlying causes: cerebral palsy, spina bifida, Arnold-Chiari malformation, hydrocephalus with shunt, microcephaly, hemimegalencephaly, Biotin deficiency, Rett's syndrome, cerebral dysgenesis, ataxia, intractable seizure and idiopathic developmental delay with seizures. Of these 27 girls, 10 had CPP (Group B1) and 17 had normal puberty (Group B2).

Mean (range) age of Group A at enrolment was 9.6 years (7-13). All had CPP with a mean (range) age of presentation of 6.3 years (5.5-7.8) and were on GnRHa for a period of 2.2 years (0.2- 5.4). Of the 27 girls with disability, those with CPP (Group B1) had a mean (range) age at presentation of 6.5 years (3.0-7.6) with GnRHa treatment for 2.6 years (1.9-5.0). For Group B2 the mean (range) age at start of puberty was 11 years (9.5-13.0) with GnRHa treatment for a mean of 0.9 years (0.3-3.1).

From the interview, all mothers of the Group A girls intended to delay their puberty, while 13 (48%) of the Group B mothers expressed the wish to stop the puberty. While none of the Group A mothers considered surgical intervention for stopping puberty, 7 of the mothers in Group B (26%) would consider surgical intervention (hysterectomy) to prevent puberty permanently (Table 1).

Fear of short adult height was the main reason identified by mothers in Group A for delaying puberty, while none of the mothers in Group B had height as a concern. Inability to cope with puberty psychologically was given as a reason by 10 (67%) of the mothers in Group A, but cited less frequently in mothers in Group B. Nine (60%) of Group A mothers feared peer rejection for their daughters which was not the case in Group B. Concern about hygiene management during menstruation was the main concern which was shared by 25 (92.5%) of mothers in Group B while it was raised by only 4 (27%) of Group A mothers. Fear of abnormal sexual and emotional behaviour was raised in both groups. However, it was more highly rated by mothers in Group B

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3 as a reason to delay puberty compared to Group A. Over half of mothers in Group B feared  
4 sexual abuse or unwanted pregnancy in their daughters while it was not a major concern in  
5 mothers in Group A. Inability to express menstrual pain and possible aggravation of convulsions  
6 or chest pain due to wheel chair strapping on the developing breast were all expressed as  
7 pertinent concerns by mothers in Group B while none were applicable to Group A. Around 23  
8 (85%) of mothers in Group B feared difficulty in maintaining hygiene during menstruation. Fear  
9 of their daughter's inability to express pre/menstrual pain was expressed by 8 (30%) of mothers  
10 and 5 (18.5%) of them were concerned about increased convulsion frequency with progression of  
11 puberty. Seven (26%) mothers in Group B expressed concern about the difficulty of seeking  
12 male family member help if their daughter had progressive signs of puberty, due to cultural and  
13 religious reasons (Table 2).  
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## Discussion

Parents of girls with early puberty are reported to be worried, embarrassed and concerned about their daughters' inability to cope with early sexual development and possible peer rejection (7). It is reported that girls who mature earlier than their peers find pubertal adjustment more challenging. Those girls appear to go through various psychological difficulties and can experience detrimental sequelae (13). Stice et al reported that girls who had CPP were at 1.9 times greater risk for depression (14). In our cohort, two thirds of the mothers of girls without disability expressed concern that their daughters might not be able to cope with puberty changes psychologically and might face peer rejection. This concern, however, was infrequent in mothers of girls with disability. Concerns about how their daughters would cope with the physical changes of puberty were more prevalent in girls with disability. These concerns included painful breast development and menstrual pain which they might not be able to fully express. In girls who wear a harness in a wheel chair, breast development in puberty can cause discomfort due to chest tenderness with the harness impinging on the developing breast tissue (15). If the girl is unable to express the pain, this might not be recognised by families and may just manifest as irritability.

There is a high level of parental anxiety regarding the impact of menses, particularly when disability is severe (3). The anxiety may relate to the impact on the child's medical condition such as on the frequency of convulsions, or it may relate to the consequences of puberty. The latter mainly includes hygiene issues for girls with disability, menstrual irregularities, premenstrual symptoms, cyclical pain (which might not be properly expressed), unwanted sexual contact or pregnancy (16, 17). These issues collectively were more often raised by mothers of girls with disability in our cohort.

Less severely disabled children may experience a deterioration in their behaviour and demonstrate socially-inappropriate sexual behaviour such as public masturbation at an early age, raising family concern about risk of sexual contact (15). Abnormal sexual behaviour was an issue of concern in a far greater proportion of mothers of girls with disability in our study.

Increase in linear growth is a major component of puberty. It is well known that untreated children with CPP have an initial growth spurt then shorter adult height. All mothers of girls in

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3 Group A raised growth as a main issue when medical advice for puberty delay was sought.  
4 Interestingly, none of the mothers with girls with disability felt that short adult height was an  
5 issue. While final adult height might not be a concern for families with disabled children, rapid  
6 growth associated with puberty does have an impact on the family and the child with disability.  
7 It raises issues of needing to change medical equipment, such as provision of an adequately sized  
8 wheelchair, progression of scoliosis bracing or altering the size for chest braces or ankle-foot  
9 orthoses (15).

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16 Parent and care-givers may occasionally approach health care professionals requesting  
17 permanent surgical measures to abolish puberty, which raises ethical dilemmas if the patient is  
18 not able to participate fully in consent (6). All mothers in Group A wanted to delay rather than  
19 stop puberty. However, 13 (48%) of Group B mothers chose stopping of puberty. Surgical  
20 intervention (hysterectomy) was a measure that was considered by 7 mothers of those 13. The  
21 choice of delaying or stopping puberty did not seem to depend on the daughters' degree of  
22 disability. However further sub analysis was not feasible due to the relatively small number of  
23 participants. Concerns about potential treatment side effects, inconvenience or expense may  
24 contribute to why a family may consider more permanent surgical methods to avoid  
25 menstruation. 2 mothers in Group B chose surgical intervention with hysterectomy fearing long  
26 term side effects of medical treatment. Use of GnRHa to treat central PP is proven to be safe and  
27 effective (8). However, these medications are relatively expensive when compared with other  
28 treatments that can be used to delay or abolish menstruation, and GnRHa are very rarely covered  
29 by medical insurance policies. In addition, it is an injectable form of medication which is often a  
30 traumatic experience and can cause sterile abscesses. In our cohort, one girl developed an  
31 abscess at the site injection which subsided and did not recur on further injections. Treatment  
32 with GnRHa require attention to dosage and interval of injection to avoid intermittent bleeding.  
33 The initial injection had a stimulatory effect and 2 girls had vaginal bleeding after commencing  
34 treatment. Another girl had intermittent bleeding during the first 9 months of treatment that  
35 subsequently subsided. Overall, GnRHa were found to be an acceptable form of treatment in our  
36 group particularly with the low frequency of injection by using the depot form. However, it can  
37 be a challenge to persuade the mothers to stop the medicine after the treatment period is  
38 completed to avoid long term side effects, especially the effect of GnRHa on bone mineral  
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3 density. Although final height data suggests that the reduction in bone mineral density on  
4 treatment is reversed by the time final height is attained and peak bone density is unaffected (18),  
5 it is recommended to avoid the long-term use of puberty antagonists (11).  
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10 In our region where mothers may be assisted by male family members and male helpers when  
11 undertaking care such as bathing, transferring and lifting their daughter with disability, there are  
12 significant issues and difficulties in utilising male help as their daughter matures. Involving a  
13 male helper in these daily routines involves physical exposure of the growing girl which might  
14 not be acceptable to the mother or the male helper. This stems from both cultural and religious  
15 reasons and might be a reason for seeking help to delay or abolish puberty (personal  
16 observation). In our cohort, over a quarter of mothers interviewed expressed this issue as a major  
17 concern.  
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24 Our study shows that early pubertal changes in girls with disability are associated with major  
25 concerns. Mothers of girls with disability seek medical help to delay puberty due to various  
26 reasons, particularly, fear of the difficulty in maintaining hygiene during menstruation. In  
27 comparison, short adult height is the main concern expressed by mothers of girls with CPP  
28 without disability. Culture and religion have an impact on the decision making of delaying  
29 puberty and some families would consider surgical measures to stop puberty in girls with  
30 disability.  
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37 Limitations of our study include the use of a quantitative questionnaire methodology in a  
38 relatively small sample size, and it would be valuable to further explore the differences we  
39 observed through a qualitative study in a larger cohort. Another limitation is that we explored  
40 maternal attitudes through a retrospective study of girls already on treatment and the answers  
41 given may be subject to recall bias. It may be more pertinent to explore these questions  
42 prospectively. The maternal attitudes highlighted in this study reflect the social, religious and  
43 cultural norms of this geographical region and further studies in other populations would be  
44 needed to be able to generalise the results more widely.  
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### Authors' contribution

Asma Deeb designed the study, applied for ethical approval and wrote the manuscript. Mariette Akle designed the questionnaire and interviewed the study participants. Abrar Al Zaabi and Zohra Siwji collected patients' data and collated the results. Salima Attia, Hana Al Suwaidi and Nabras Al Qahtani are clinicians who recruited the study subjects. Sarah Ehtisham revised the study proposal, advised on the questionnaire design and revised the manuscript.

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3 **Table legends**  
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5 **Table 1:** Characteristics of study population  
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7 **Table 2:** Reasons for seeking puberty suppression by mothers. Numbers in italic under  
8 percentage indicates the order of frequency of the choice.  
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Confidential: For Review Only

## References

- 1) Brault Matthew. Americans with Disabilities: Household Economic Studies. Current Population Reports 2010;P70-131.
- 2) Mithyantha R, Bassi Z. Early Pubertal Changes in Neurodisabled Children. An under-diagnosed issue? Arch dis child 2013;98:A53.
- 3) Zacharin M. The Impact of Menstruation in Adolescents with Disabilities Related to Cerebral Palsy. Arch Dis Child 2010;95(7):526-530.
- 4) Albanese A, Hopper NW. Suppression of menstruation in adolescents with severe learning disabilities. Arch Dis Child 2007;92:629–632.
- 5) Zacharin MR: Puberty, contraception, and hormonal management for young people with disabilities. Clin Pediatr 2009;48:149–155.
- 6) Isaacs D, Tobin B, Hamblin J, Slaytor E, Donaghue KC, Munns C, Kilham HA: Managing ethically questionable parental requests: growth suppression and manipulation of puberty. J Paediatr Child Health 2011;47:581–584.
- 7) Mazur T, Clopper RR. Pubertal disorders: Psychology and clinical management. Endocrinology and Metabolism Clinics of North America 1991;20(1):211-230.
- 8) Carel JC, Eugster EA, Rogol A. Consensus statement on the use of gonadotropin-releasing hormone analogs in children. Pediatrics 2009;123(4):e752-62.
- 9) Van Schroyen Lantman-de Valk HM, Rook F, Maaskant MA: The use of contraception by women with intellectual disabilities. J Intellect Disabil Res 2011;55(4):434-440.
- 10) Dizon C, Allen L, Ornstein M. Menstrual and contraceptive issues among young women with developmental delay: a retrospective review of cases at the Hospital for Sick Children, Toronto. J Pediatr Adolesc Gynecol 2005;18:157–162.
- 11) Kirkham Yolanda, Ornstein M, Aggarwal Anjali, McQuillan S et al. Menstrual suppression in special circumstances: SOGC clinical practice guidelines 2014;36(10):915–924.
- 12) Guilbert, E., Boroditsky, R., Black, A., Kives, S., Leboeuf, M., Mirosh, M. et al. Society of Obstetricians and Gynaecologists of Canada. Canadian consensus guideline on continuous and extended hormonal contraception. SOGC Clinical Practice Guidelines. J Obstet Gynaecol Can 2007;29: S1–S32.

- 1  
2  
3 13) Ge X, Conger RD, Elder GH. Coming of age too early: pubertal influences on girls'  
4 vulnerability to psychological distress. *Child Development* 1996;67:3386–3400.  
5  
6  
7 14) Stice E, Presnell K, Bearman SK. Relation of early menarche to depression, eating  
8 disorders, substance abuse, and comorbid psychopathology among adolescent girls.  
9 *Developmental Psychology* 2001;37:608–619.  
10  
11 15) Zacharin M. Endocrine Problems in Children and Adolescents Who Have  
12 Disabilities. *Horm Res Paediatr* 2013;80:221–228.  
13  
14 16) Quint EH: Menstrual issues in adolescents with physical and developmental  
15 disabilities. *Ann NY Acad Sci* 2008;1135:230-236.  
16  
17 17) Grover SR: Gynaecological issues in adolescents with disability. *J Paediatr Child*  
18 *Health* 2011; 47:610-613.  
19  
20 18) van der Sluis IM, Boot AM, Krenning EP, et al. Longitudinal follow-up of bone  
21 density and body composition in children with precocious or early puberty before,  
22 during and after cessation of GnRH agonist therapy. *J Clin Endocrinol Metab*  
23 2002;87(2):506–512.  
24  
25  
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Table 1

Variable	Girls with CPP and no disability Group A	Girls with disability Group B	
		CPP Group B1	Normal puberty Group B2
Number	15	10	17
Mean (range) of age at enrolment (years)	9.6 (7.0-13.0)	9.2 (5.0 -11.1)	10.1 (10.3 -14)
Mean (range) of age at onset of puberty (years)	6.3 (5.5-7.8)	6.5 (3.0 - 7.6)	11 (9.5 -13.0)
Mean (range) duration of treatment (years)	2.2 (0.2-5.4)	2.6 (1.9 – 5)	0.9 (0.3 -3.1)
Number of mothers intending to delay puberty	15 (100%)	8 (80%)	6 (35%)
Number of mothers intending to stop puberty	0	2 (20%)	11 (55%)
Number of mothers who would consider surgical intervention to stop puberty	0	2 (20%)	5 (29%)

**Table 2**

Reason for delaying/stopping puberty	Number	
	Girls without disability Group A N=15	Girls with disability Group B N=27
Fear of short adult height	15 (100%) 1	0
Inability to cope with puberty changes psychologically	10 (67%) 2	5 (18.5%) 7
Fear of peer rejection	9 (60%) 3	0
Difficulty in maintaining hygiene during menstruation	4 (27%) 4	25 (92.5%) 1
Fear of abnormal sexual or emotional behaviour	3 (20%) 5	11 (41%) 4
Fear of child abuse or unwanted pregnancy	2 (13%) 6	15 (55.5%) 3
Menstrual pain that cannot be properly expressed	N/A	8 (30%) 5
Difficulty of seeking male family member help	N/A	7 (26%) 6
Effect on convulsion frequency	N/A	5 (18.5%) 7
Chest pain from pressure of wheelchair harness on breasts	N/A	3 (11%) 8

## Appendix: Questionnaire completed by clinical psychologist during the interviews

### Patient categories

- Group A: Girl without disability with precocious puberty
- Group B1: Girl with disability presenting with precocious puberty
- Group B2: Girl with disability presenting with normal puberty

### What was the main reason/reasons you wanted to delay or stop puberty

- a) Fear of short adult height
- b) Inability to cope with puberty changes psychologically
- c) Fear of peer rejection
- d) Difficulties in maintaining hygiene during menstruation
- e) Fear of abnormal sexual or emotional behaviour
- f) Fear of child abuse or unwanted pregnancy
- g) Menstrual pain that cannot be properly expressed
- h) Difficulty of seeking male family member help
- i) Effect on convulsion frequency
- j) Chest pain due to wheelchair harness on chest
- k) Other reasons. Please list

### 1) Was your intention of treatment to delay or stop puberty permanently

- Delay
- Stop

### 2) If to delay, for how long did you want to delay the puberty?

- Short period (few months)
- Long period (years)
- According to doctors' advice

### 3) Would you consider permanent surgical measures like hysterectomy to stop puberty?

- Yes
- No

## BMJ Paediatrics Open

### Maternal attitude towards delaying puberty in girls with and without disability; a questionnaire-based study from the United Arab Emirates

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3 **Maternal attitude towards delaying puberty in girls with and without disability; a**  
4 **questionnaire-based study from the United Arab Emirates**  
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7 *Asma Deeb<sup>1</sup>, Mariette Akle<sup>1</sup>, Abrar Al Zaabi<sup>1</sup>, Zohra Siwji<sup>1</sup>, Salima Attia<sup>1</sup>, Hana Al Suwaidi<sup>1</sup>,*  
8 *Nabras Al Qahtani<sup>1</sup> & Sarah Ehtisham<sup>2</sup>*  
9

10  
11  
12 1) Paediatric Endocrinology Department  
13 Mafraq Hospital  
14 PO Box 2951  
15 T + 971-50-8350568  
16 F +971-2-  
17 Abu Dhabi, United Arab Emirates  
18  
19

20 Asma Deeb (corresponding author)

21 [adeeb@seha.ae](mailto:adeeb@seha.ae)

22 Mariette Akle

23 [makle@seha.ae](mailto:makle@seha.ae)

24 Abrar Al Zaabi

25 [abzaabi@seha.ae](mailto:abzaabi@seha.ae)

26 Zohra Siwji

27 [zoali@seha.ae](mailto:zoali@seha.ae)

28 Salima Attia

29 [sabood@seha.ae](mailto:sabood@seha.ae)

30 Hanah Al Suwaidi

31 [hsuwaidi@seha.ae](mailto:hsuwaidi@seha.ae)

32 Nabras Al Qahtani

33 [nmqahtani@seha.ae](mailto:nmqahtani@seha.ae)

34  
35  
36 2) Mediclinic City Hospital  
37 Dubai Healthcare City  
38 PO Box 505004  
39 T +97144355644  
40 F +97144359900  
41 Dubai, United Arab Emirates  
42  
43

44 - Sarah Ehtisham  
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1  
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3 [sarah.ehtisham@mediclinic.ae](mailto:sarah.ehtisham@mediclinic.ae)  
4

5 **Short running title**

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7 Puberty suppression in girls  
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### What is already known about the subject

- Puberty suppression may be sought for girls with disability for improving quality of life.
- Gonadotropin releasing hormone analogue (GnRHa) therapy is an option for pubertal suppression in girls with and without disability.
- Surgical intervention for permanent suppression of menstruation is occasionally considered by families of girls with disability.

### What this study adds about the subject

- Mothers of girls with and without disability have different reasons for seeking advice on puberty suppression.
- Cultural and religious issues may influence the attitude of mothers on puberty suppression.
- Concerns about menstrual hygiene and fear of short adult height were the commonest reasons for seeking puberty suppression in girls in the United Arab Emirates with and without with disability respectively.

## Abstract

**Background:** Parental anxiety about the impact of puberty/menses, particularly in girls with severe disability leads to seeking therapeutic pubertal suppression. We aim to explore maternal attitudes and reasons for seeking pubertal suppression.

**Methods:** Mothers of girls receiving GnRHa therapy in Mafraq hospital, Abu Dhabi were enrolled in the study. A semi-structured interview was conducted to ascertain possible reasons for delaying puberty. The study group was divided into girls with disability with central precocious puberty (CPP) or normal puberty and girls without disability presenting with CPP.

**Results:** 42 mother-daughter pairs were enrolled and divided into 2 groups; Group A: 15 girls with CPP with no disability. Group B: 27 girls with of disability (10 had CPP (Group B1) and 17 had normal pubertal timing (Group B2). Mothers in Group A aimed to delay puberty, whilst in Group B, 13 (48%) of mothers desired to halt puberty and 7 (26%) requested permanent surgical intervention. Fear of short stature (15, 100%), inability to cope psychologically (10, 67%) and fear of peer rejection (9, 60%) were the main concerns in Group A. In Group B, mothers were concerned about menstrual hygiene management (25, 92.5%), fear of child abuse or unwanted pregnancy (15, 55%) and fear of inability to express pain/discomfort with menstruation (8, 30%).

**Conclusion:** Mothers of girls with disability commonly seek medical help to delay/halt puberty due to concerns about menstrual hygiene. Short final height was the main concern for girls without disability. Culture & religion play an important role in puberty management in girls with disability.

## Introduction

Childhood disability has a major impact on the affected child and families. A Census report from the USA showed that there are over 5 million children under 18 years have disabilities (1). The endocrine system is highly affected by disability with effects on the hypothalamic pituitary function, bone health, pubertal timing, sexual function and fertility. Accordingly, assessment of pubertal changes should be an essential component of the multidisciplinary care for children with disability (2). In this group of children, puberty poses various challenges particularly in relation to management of menstruation. It is reported that 50% of families of girls with disabilities seek medical advice in relation to menstruation management (3).

Children with neurologic impairment have a higher risk of central precocious puberty (CPP). Girls with cerebral palsy undergo puberty earlier but reach menarche slightly later than the general population (4). Families of adolescents with disabilities can present for counselling before or after menarche (5).

Maternal attitudes towards their daughters' early puberty is influenced by the maturity and the cognitive ability of the girl. While physical care issues related to puberty can be prominent in girls with disability, other factors might be of more concerns in girls with normal mental development. There is a high level of parental anxiety regarding the impact of menses, particularly in girls with severe disability (3). Increase in linear growth is a major component of puberty and it is well known that untreated CPP is detrimental to adult height. Parental concerns about growth may be different in children with and without disability. Parents and care-givers occasionally approach health care professionals requesting permanent surgical measures to abolish puberty, which raises ethical dilemmas if the patient is not able to participate fully in consent (6).

Treating girls with CPP can alleviate psychological distress and delay menstruation (7). Use of gonadotropin releasing hormone analogue (GnRHa) therapy to treat CPP is proven to be safe and effective (8). Advancing puberty and onset of menstruation can raise clinical dilemmas associated with hormonal intervention either to delay onset of menstruation or to reduce the menstrual flow. Although various non-hormonal and hormonal interventions are available for

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3 such a purpose, each modality of treatment has advantages and disadvantages. It is of paramount  
4 importance to individualise the treatment decisions considering each child's circumstances (4, 9).  
5 It is recommended that menstrual suppression therapy should not commence until menarche is  
6 achieved (10, 11). However, in certain circumstances, delaying puberty and menstrual  
7 suppression can reduce morbidity and improve quality of life both for the girls and for their  
8 caregivers (12).  
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15 In the United Arab Emirate and some other Arab and Asian countries, mothers are the main  
16 carers of girls with disability and are more commonly involved in hospital visits and treatment  
17 decisions than fathers (personal observation). In our region, pubertal changes in a girl with  
18 disabilities can pose significant issues when male family members and helpers are involved in  
19 personal care. This stems from both cultural and religious reasons and may contribute to reasons  
20 for seeking help to delay or suppress puberty (personal observation).  
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26 Understanding the maternal attitude and concerns towards puberty enables health care  
27 professionals to provide appropriate and culture-sensitive solutions. Highlighting specific issues  
28 in a population helps to improve quality of life and maternal satisfaction through provision of  
29 appropriate measures.  
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### 34 **Aims of the study**

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37 We aimed to explore maternal attitudes towards their daughters' puberty. We also planned to  
38 explore the reasons for seeking medical help to delay puberty in girls with and without disability  
39 who presented with normal or precocious puberty.  
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## Patients & Methods

The study was undertaken at the Paediatric Endocrinology department of Mafraq Hospital. A list of girls receiving gonadotropin releasing hormone analogue (GnRHa) therapy for delaying puberty between 2013-2016 was compiled from the clinic database. This list was cross-checked with the hospital pharmacy electronic records for dispensing of GnRHa. All girls in the database were approached to enrol in the study. Mothers of girls on GnRHa therapy were approached to participate in the study during one of their daughters' scheduled visits for GnRHa injection. The study was explained to the mothers and information sheets were given. Mothers who agreed to participate in the study gave a verbal consent and had their daughters enrolled. The study was approved by Mafraq hospital Research and Ethics committee.

The mothers had a semi-structured interview by a clinical psychologist (MA) in a one to one setting. A questionnaire was designed to include possible reasons for intervening in puberty. The multiple choice of reasons was derived from a literature search on the subject and reflected study team own observation and cultural background of the cohort. The questionnaire was completed during the interview (Appendix). The mothers were asked to suggest other reasons for seeking pubertal suppression if their reason was not included in the list.

Medical information was collected from the electronic medical record by the study team including: detailed underlying medical condition for girls with disability, cause of CPP (if established), age at puberty presentation, the initial signs of puberty, and the results of the initial biochemistry and radiology tests.

No formal power calculation was performed. The intention was to approach all eligible patients and enrol all who consent. All girls receiving GnRHa therapy for puberty were eligible for enrolment and they were subdivided into the following groups for analysis:

- Group A: Girls with CPP without disability
- Group B: Girls with disability – subdivided into Group B1 with CPP and Group B2 with normal puberty

## Results

46 girls receiving GnRHa therapy were identified in the medical and hospital pharmacy records. 42 mothers consented to the study and their daughters were enrolled. All the families were Muslim, 19 were Emirati, 18 from other Arab countries and 5 were Indian or Pakistani. 15 of the girls had no disability (Group A). All presented with CPP and were diagnosed with idiopathic CPP after performing full investigations including brain MRI scans. 27 of the girls had a degree of physical/intellectual disability (Group B) with the following underlying causes: cerebral palsy, spina bifida, Arnold-Chiari malformation, hydrocephalus with shunt, microcephaly, hemimegalencephaly, Biotin deficiency, Rett's syndrome, cerebral dysgenesis, ataxia, intractable seizure and idiopathic developmental delay with seizures. Of these 27 girls, 10 had CPP (Group B1) and 17 had normal puberty (Group B2).

Mean (range) age of Group A at enrolment was 9.6 years (7-13). All had CPP with a mean (range) age of presentation of 6.3 years (5.5-7.8) and were on GnRHa for a period of 2.2 years (0.2- 5.4). Of the 27 girls with disability, those with CPP (Group B1) had a mean (range) age at presentation of 6.5 years (3.0-7.6) with GnRHa treatment for 2.6 years (1.9-5.0). For Group B2 the mean (range) age at start of puberty was 11 years (9.5-13.0) with GnRHa treatment for a mean of 0.9 years (0.3-3.1).

From the interview, all mothers of the Group A girls intended to delay their puberty, while 13 (48%) of the Group B mothers expressed the wish to stop the puberty. While none of the Group A mothers considered surgical intervention for stopping puberty, 7 of the mothers in Group B (26%) would consider surgical intervention (hysterectomy) to prevent puberty permanently (Table 1).

Fear of short adult height was the main reason identified by mothers in Group A for delaying puberty, while none of the mothers in Group B had height as a concern. Inability to cope with puberty psychologically was given as a reason by 10 (67%) of the mothers in Group A, but cited less frequently in mothers in Group B. Nine (60%) of Group A mothers feared peer rejection for their daughters which was not the case in Group B. Concern about hygiene management during menstruation was the main concern which was shared by 25 (92.5%) of mothers in Group B while it was raised by only 4 (27%) of Group A mothers. Fear of abnormal sexual and emotional behaviour was raised in both groups. However, it was more highly rated by mothers in Group B

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3 as a reason to delay puberty compared to Group A. Over half of mothers in Group B feared  
4 sexual abuse or unwanted pregnancy in their daughters while it was not a major concern in  
5 mothers in Group A. Inability to express menstrual pain and possible aggravation of convulsions  
6 or chest pain due to wheel chair strapping on the developing breast were all expressed as  
7 pertinent concerns by mothers in Group B while none were applicable to Group A. Around 25  
8 (95%) of mothers in Group B feared difficulty in maintaining hygiene during menstruation. Fear  
9 of their daughter's inability to express pre/menstrual pain was expressed by 8 (30%) of mothers  
10 and 5 (18.5%) of them were concerned about increased convulsion frequency with progression of  
11 puberty. Seven (26%) mothers in Group B expressed concern about the difficulty of seeking  
12 male family member help if their daughter had progressive signs of puberty, due to cultural and  
13 religious reasons (Table 2).  
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## Discussion

Parents of girls with early puberty are reported to be worried, embarrassed and concerned about their daughters' inability to cope with early sexual development and possible peer rejection (7). It is reported that girls who mature earlier than their peers find pubertal adjustment more challenging. Those girls appear to go through various psychological difficulties and can experience detrimental sequelae (13). Stice et al reported that girls who had CPP were at 1.9 times greater risk for depression (14). In our cohort, two thirds of the mothers of girls without disability expressed concern that their daughters might not be able to cope with puberty changes psychologically and might face peer rejection. This concern, however, was infrequent in mothers of girls with disability. Concerns about how their daughters would cope with the physical changes of puberty were more prevalent in girls with disability. These concerns included painful breast development and menstrual pain which they might not be able to fully express. In girls who wear a harness in a wheel chair, breast development in puberty can cause discomfort due to chest tenderness with the harness impinging on the developing breast tissue (15). If the girl is unable to express the pain, this might not be recognised by families and may just manifest as irritability.

There is a high level of parental anxiety regarding the impact of menses, particularly when disability is severe (3). The anxiety may relate to the impact on the child's medical condition such as on the frequency of convulsions, or it may relate to the consequences of puberty. The latter mainly includes hygiene issues for girls with disability, menstrual irregularities, premenstrual symptoms, cyclical pain (which might not be properly expressed), unwanted sexual contact or pregnancy (16, 17). These issues collectively were more often raised by mothers of girls with disability in our cohort.

Less severely disabled children may experience a deterioration in their behaviour and demonstrate socially-inappropriate sexual behaviour such as public masturbation at an early age, raising family concern about risk of sexual contact (15). Abnormal sexual behaviour was an issue of concern in a far greater proportion of mothers of girls with disability in our study.

Increase in linear growth is a major component of puberty. It is well known that untreated children with CPP have an initial growth spurt then shorter adult height. All mothers of girls in

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3 Group A raised growth as a main issue when medical advice for puberty delay was sought.  
4 Interestingly, none of the mothers with girls with disability felt that short adult height was an  
5 issue. While final adult height might not be a concern for families with disabled children, rapid  
6 growth associated with puberty does have an impact on the family and the child with disability.  
7 It raises issues of needing to change medical equipment, such as provision of an adequately sized  
8 wheelchair, progression of scoliosis bracing or altering the size for chest braces or ankle-foot  
9 orthoses (15).

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16 Parent and care-givers may occasionally approach health care professionals requesting  
17 permanent surgical measures to abolish puberty, which raises ethical dilemmas if the patient is  
18 not able to participate fully in consent (6). All mothers in Group A wanted to delay rather than  
19 stop puberty. However, 13 (48%) of Group B mothers chose stopping of puberty. Surgical  
20 intervention (hysterectomy) was a measure that was considered by 7 mothers of those 13. The  
21 choice of delaying or stopping puberty did not seem to depend on the daughters' degree of  
22 disability. However further sub analysis was not feasible due to the relatively small number of  
23 participants. Concerns about potential treatment side effects, inconvenience or expense may  
24 contribute to why a family may consider more permanent surgical methods to avoid  
25 menstruation. 2 mothers in Group B chose surgical intervention with hysterectomy fearing long  
26 term side effects of medical treatment. Use of GnRHa to treat central PP is proven to be safe and  
27 effective (8). However, these medications are relatively expensive when compared with other  
28 treatments that can be used to delay or abolish menstruation, and GnRHa are very rarely covered  
29 by medical insurance policies. In addition, it is an injectable form of medication which is often a  
30 traumatic experience and can cause sterile abscesses. In our cohort, one girl developed an  
31 abscess at the site injection which subsided and did not recur on further injections. Treatment  
32 with GnRHa require attention to dosage and interval of injection to avoid intermittent bleeding.  
33 The initial injection had a stimulatory effect and 2 girls had vaginal bleeding after commencing  
34 treatment. Another girl had intermittent bleeding during the first 9 months of treatment that  
35 subsequently subsided. Overall, GnRHa were found to be an acceptable form of treatment in our  
36 group particularly with the low frequency of injection by using the depot form. However, it can  
37 be a challenge to persuade the mothers to stop the medicine after the treatment period is  
38 completed to avoid long term side effects, especially the effect of GnRHa on bone mineral  
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3 density. Although final height data suggests that the reduction in bone mineral density on  
4 treatment is reversed by the time final height is attained and peak bone density is unaffected (18),  
5 it is recommended to avoid the long-term use of puberty antagonists (11).  
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10 In our region where mothers may be assisted by male family members and male helpers when  
11 undertaking care such as bathing, transferring and lifting their daughter with disability, there are  
12 significant issues and difficulties in utilising male help as their daughter matures. Involving a  
13 male helper in these daily routines involves physical exposure of the growing girl which might  
14 not be acceptable to the mother or the male helper. This stems from both cultural and religious  
15 reasons and might be a reason for seeking help to delay or abolish puberty (personal  
16 observation). In our cohort, over a quarter of mothers interviewed expressed this issue as a major  
17 concern.  
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24 Our study shows that early pubertal changes in girls with disability are associated with major  
25 concerns. Mothers of girls with disability seek medical help to delay puberty due to various  
26 reasons, particularly, fear of the difficulty in maintaining hygiene during menstruation. In  
27 comparison, short adult height is the main concern expressed by mothers of girls with CPP  
28 without disability. Culture and religion have an impact on the decision making of delaying  
29 puberty and some families would consider surgical measures to stop puberty in girls with  
30 disability.  
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37 Limitations of our study include the use of a quantitative questionnaire methodology in a  
38 relatively small sample size, and it would be valuable to further explore the differences we in a  
39 larger cohort. Another limitation is that we explored maternal attitudes through a retrospective  
40 study of girls already on treatment and the answers given may be subject to recall bias. It may be  
41 more pertinent to explore these questions prospectively. The maternal attitudes highlighted in  
42 this study reflect the social, religious and cultural norms of this geographical region and further  
43 studies in other populations would be needed to be able to generalise the results more widely.  
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### Authors' contribution

Asma Deeb designed the study, applied for ethical approval and wrote the manuscript. Mariette Akle designed the questionnaire and interviewed the study participants. Abrar Al Zaabi and Zohra Siwji collected patients' data and collated the results. Salima Attia, Hana Al Suwaidi and Nabras Al Qahtani are clinicians who recruited the study subjects. Sarah Ehtisham revised the study proposal, advised on the questionnaire design and revised the manuscript.

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3 **Table legends**  
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5 **Table 1:** Characteristics of study population  
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7 **Table 2:** Reasons for seeking puberty suppression by mothers. Numbers in italic under  
8 percentage indicates the order of frequency of the choice.  
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## References

- 1) Brault Matthew. Americans with Disabilities: Household Economic Studies. Current Population Reports 2010;P70-131.
- 2) Mithyantha R, Bassi Z. Early Pubertal Changes in Neurodisabled Children. An under-diagnosed issue? Arch dis child 2013;98:A53.
- 3) Zacharin M. The Impact of Menstruation in Adolescents with Disabilities Related to Cerebral Palsy. Arch Dis Child 2010;95(7):526-530.
- 4) Albanese A, Hopper NW. Suppression of menstruation in adolescents with severe learning disabilities. Arch Dis Child 2007;92:629–632.
- 5) Zacharin MR: Puberty, contraception, and hormonal management for young people with disabilities. Clin Pediatr 2009;48:149–155.
- 6) Isaacs D, Tobin B, Hamblin J, Slaytor E, Donaghue KC, Munns C, Kilham HA: Managing ethically questionable parental requests: growth suppression and manipulation of puberty. J Paediatr Child Health 2011;47:581–584.
- 7) Mazur T, Clopper RR. Pubertal disorders: Psychology and clinical management. Endocrinology and Metabolism Clinics of North America 1991;20(1):211-230.
- 8) Carel JC, Eugster EA, Rogol A. Consensus statement on the use of gonadotropin-releasing hormone analogs in children. Pediatrics 2009;123(4):e752-62.
- 9) Van Schroyen Lantman-de Valk HM, Rook F, Maaskant MA: The use of contraception by women with intellectual disabilities. J Intellect Disabil Res 2011;55(4):434-440.
- 10) Dizon C, Allen L, Ornstein M. Menstrual and contraceptive issues among young women with developmental delay: a retrospective review of cases at the Hospital for Sick Children, Toronto. J Pediatr Adolesc Gynecol 2005;18:157–162.
- 11) Kirkham Yolanda, Ornstein M, Aggarwal Anjali, McQuillan S et al. Menstrual suppression in special circumstances: SOGC clinical practice guidelines 2014;36(10):915–924.
- 12) Guilbert, E., Boroditsky, R., Black, A., Kives, S., Leboeuf, M., Mirosh, M. et al. Society of Obstetricians and Gynaecologists of Canada. Canadian consensus guideline on continuous and extended hormonal contraception. SOGC Clinical Practice Guidelines. J Obstet Gynaecol Can 2007;29: S1–S32.

- 1  
2  
3 13) Ge X, Conger RD, Elder GH. Coming of age too early: pubertal influences on girls'  
4 vulnerability to psychological distress. *Child Development* 1996;67:3386–3400.  
5  
6  
7 14) Stice E, Presnell K, Bearman SK. Relation of early menarche to depression, eating  
8 disorders, substance abuse, and comorbid psychopathology among adolescent girls.  
9 *Developmental Psychology* 2001;37:608–619.  
10  
11 15) Zacharin M. Endocrine Problems in Children and Adolescents Who Have  
12 Disabilities. *Horm Res Paediatr* 2013;80:221–228.  
13  
14 16) Quint EH: Menstrual issues in adolescents with physical and developmental  
15 disabilities. *Ann NY Acad Sci* 2008;1135:230-236.  
16  
17 17) Grover SR: Gynaecological issues in adolescents with disability. *J Paediatr Child*  
18 *Health* 2011; 47:610-613.  
19  
20 18) van der Sluis IM, Boot AM, Krenning EP, et al. Longitudinal follow-up of bone  
21 density and body composition in children with precocious or early puberty before,  
22 during and after cessation of GnRH agonist therapy. *J Clin Endocrinol Metab*  
23 2002;87(2):506–512.  
24  
25  
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Table 1

Variable	Girls with CPP and no disability Group A	Girls with disability Group B	
		CPP Group B1	Normal puberty Group B2
Number	15	10	17
Mean (range) of age at enrolment (years)	9.6 (7.0-13.0)	9.2 (5.0 -11.1)	10.1 (10.3 -14)
Mean (range) of age at onset of puberty (years)	6.3 (5.5-7.8)	6.5 (3.0 - 7.6)	11 (9.5 -13.0)
Mean (range) duration of treatment (years)	2.2 (0.2-5.4)	2.6 (1.9 – 5)	0.9 (0.3 -3.1)
Number of mothers intending to delay puberty	15 (100%)	8 (80%)	6 (35%)
Number of mothers intending to stop puberty	0	2 (20%)	11 (55%)
Number of mothers who would consider surgical intervention to stop puberty	0	2 (20%)	5 (29%)

**Table 2**

Reason for delaying/stopping puberty	Number	
	Girls without disability Group A N=15	Girls with disability Group B N=27
Fear of short adult height	15 (100%) 1	0
Inability to cope with puberty changes psychologically	10 (67%) 2	5 (18.5%) 6
Fear of peer rejection	9 (60%) 3	0
Difficulty in maintaining hygiene during menstruation	4 (27%) 4	25 (92.5%) 1
Fear of abnormal sexual or emotional behaviour	3 (20%) 5	11 (41%) 3
Fear of child abuse or unwanted pregnancy	2 (13%) 6	15 (55.5%) 2
Menstrual pain that cannot be properly expressed	N/A	8 (30%) 4
Difficulty of seeking male family member help	N/A	7 (26%) 5
Effect on convulsion frequency	N/A	5 (18.5%) 6
Chest pain from pressure of wheelchair harness on breasts	N/A	3 (11%) 7

## Appendix: Questionnaire completed by clinical psychologist during the interviews

### Patient categories

- Group A: Girl without disability with precocious puberty
- Group B1: Girl with disability presenting with precocious puberty
- Group B2: Girl with disability presenting with normal puberty

### What was the main reason/reasons you wanted to delay or stop puberty

- a) Fear of short adult height
- b) Inability to cope with puberty changes psychologically
- c) Fear of peer rejection
- d) Difficulties in maintaining hygiene during menstruation
- e) Fear of abnormal sexual or emotional behaviour
- f) Fear of child abuse or unwanted pregnancy
- g) Menstrual pain that cannot be properly expressed
- h) Difficulty of seeking male family member help
- i) Effect on convulsion frequency
- j) Chest pain due to wheelchair harness on chest
- k) Other reasons. Please list

### 1) Was your intention of treatment to delay or stop puberty permanently

- Delay
- Stop

### 2) If to delay, for how long did you want to delay the puberty?

- Short period (few months)
- Long period (years)
- According to doctors' advice

### 3) Would you consider permanent surgical measures like hysterectomy to stop puberty?

- Yes
- No