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Parental decisional regret and views about optimal timing of female genital restoration surgery in congenital adrenal hyperplasia

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Summary

Purpose
The role of female genital restoration surgery (FGRS) in girls with congenital adrenal hyperplasia (CAH) is controversial, with no long-term parent-reported outcomes available. Decisional regret (DR) affects most parents after their children's treatment of pediatric conditions, including hypospadias. We aimed to assess parental DR after FGRS in infancy or toddlerhood and explore optimal timing for surgery.

Materials and methods
One-hundred and six parents of females with CAH undergoing FGRS before 3 years old and followed at our institution (1999–2017) were invited to enroll online. Higher Decision Regret Scale (DRS) scores indicated greater DR (range 0–100). Participants also reported preferred FGRS timing relative to their surgery (earlier, same, later/delayed). Non-parametric statistical tests were used.

Results
Thirty-nine parents (median 4.4 years after FGRS) participated (36.8% response rate). Median age at FGRS was 9 months. Median DRS score was 0 (mean: 5.0). Overall, 20.5% of parents reported some regret (all mild-moderate) (Figure). Fewer parents reported DR after FGRS compared with published DR after hypospadias repair (50–92%, $p \leq 0.001$) or adenotonsillectomy (41–45%, $p \leq 0.03$). No parent preferred delayed FGRS. Seven parents (18.1%) preferred earlier surgery, especially when performed after birthday (80.0% vs. 8.8%, $p = 0.004$).

Discussion
We present the first report of validated long-term parent-reported outcomes after FGRS in infant and toddler girls with CAH. One limitation is that this is largely a single surgeon series. Reasons for the observed low levels of DR are likely multifactorial. Far from a definitive study, we aimed to provide parents willing to share about their experience an opportunity to do so. For that reason, selection bias may exist in our study. While parents with higher DR were potentially less likely to participate because of mistrust of the medical establishment, those with a negative experience may in fact be more likely to voice their opinions. A low participation rate was likely a result of the sensitive nature of FGRS, a desire for privacy, and inability to locate parents. A larger study will be required to assess how DR is affected by sexual function, genital appearance and complications, and DR among women with CAH.

Conclusions
Parents of females with CAH report low levels of DR after FGRS in infancy and toddlerhood. This appears to be lower than after other genital and non-genital pediatric procedures. When present, parental DR is usually mild. No parents preferred delayed surgery, even among those with DR. Some preferred earlier surgery.

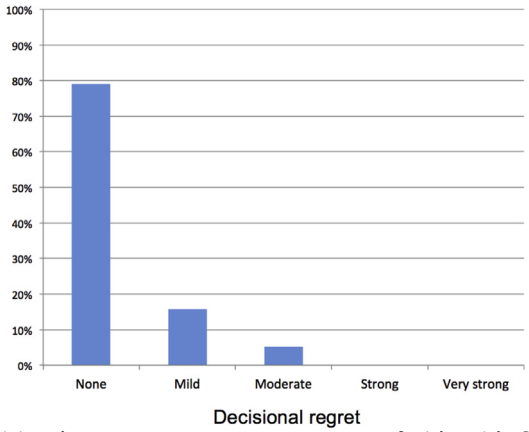

Figure Histogram of decisional regret scores among parents of girls with CAH after female genital restoration surgery.

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Introduction

Congenital adrenal hyperplasia (CAH) is the most common cause of ambiguous genitalia in newborns [1–3]. Historically, female genital restoration surgery (FGRS) was performed in infancy to prevent urinary pooling and to match external genitalia to chromosomal sex. Psychological benefits to the child, family, and caregivers [3–6], often cited as a reason for early surgery, remain to be proven. Recently, some advocacy groups, ethicists, and physicians have challenged early surgery, some calling for moratoria on genital surgery in all disorders of sexual development, until children are old enough to decide for themselves [7]. Adding to this controversy are concerns that clinical outcomes may differ for surgeries performed in infancy versus after puberty [8,9]. Reliable data to support either approach are limited, as the literature consists primarily of surgeon-reported outcomes of historical procedures [10,11].

Long-term patient- or parent-reported outcomes (PROs) are lacking to support any position regarding the role of FGRS in CAH [10]. One of the goals of any CAH management strategy is to minimize patient and parental decisional regret (DR). DR can follow any treatment decision, including a decision to forgo treatment. DR has been reported in parents of children undergoing various treatments, and in fact, DR affects 50–92% of parents after their sons' hypospadias surgery [12,13].

We aimed to assess DR of parents of females with CAH after FGRS performed in infancy and toddlerhood and to explore their opinion of optimal timing for surgery. We hypothesized that prevalence of DR after FGRS is similar to other procedures performed in childhood and few prefer delayed surgery.

Methods

We performed an IRB-approved cross-sectional online study of parents of girls with CAH followed at our institution after FGRS by age 3, mostly performed by a single surgeon (1999–2017). A minimum of 3 months since FGRS was required for inclusion. Of 118 potential parents, 106 with contact information were eligible. Adult women with CAH were also invited to participate. This report focuses on the results form parents, due to a very low response rate from women (women's responses are summarized at the end of the Results section).

Eligible participants were mailed generic letters inviting them to participate without disclosing the diagnosis or treatment history. Interested participants were emailed an individualized link to the online survey, with a reminder emailed a week later. Study data were managed using REDCap, a secure web-based platform [14].

Decisional regret

DR was assessed using the validated Decision Regret Scale (DRS) [15,16]. DRS consists of five items scored on a 5-point Likert scale (strongly disagree, disagree, neither disagree nor agree, agree, strongly agree). Scores range from 0 to 100, with higher scores indicating greater DR. Scores were classified as no DR (0), mild DR (1–25), moderate (26–50), as previously described [17], further dividing higher DR into strong (51–75) and very strong (76–100).

Sensitivity analysis

DRS is a sensitive instrument, classifying the slightest answer indicating potential regret as regretful, with no established clinically meaningful cutoff (0 = no DR vs. 1–100 = some DR). To adjust for this low threshold, sensitivity analyses were performed. First, we used cutoffs which may be more clinically meaningful: >10 [18], >25 [17,19], and >30 [20]. Second, we used a non-neutral regret definition: DR present in those who either disagreed/strongly disagreed with any of items 1, 3, or 5, or agreed/strongly agreed with items 2 or 4 [18].

Risk factors of parental decisional regret (exploratory)

Several potential predictors of higher DR were selected *a priori*: age at FGRS (<1 year old vs. 1–2 vs. >2), preoperative Prader scale (3 vs. 4–5), being a mother (vs. other), undergoing another genital surgery, and time since FGRS (<5 years, 5–10, >10 years).

Parental decisional regret after other procedures

A systematic search was completed in April 2017 to identify relevant articles in Medline (from 1950), PubMed (from 1946), Embase (from 1949), and GoogleScholar (from 1990). Combinations of the terms: decisional regret, parent, child were used. Included publications needed to use the DRS. To determine the average percentage of parents reporting DR for all reported conditions, a weighed mean of DR was calculated. For each study, the percentage of parents reporting DR was multiplied by the number of participants. The sum of these values was divided by the total number of participants.

Preferred timing of surgery

Participants were asked about their preferred timing of surgery relative to when FGRS was actually performed ("Looking back, when would you have done the original surgery for your child?"). Answer options included: earlier in life, same time, later in life.

Risk factors of earlier or later surgery (exploratory)

Similar to the analysis of DR, we screened the following predictors: age at FGRS, preoperative Prader scale, being a mother, additional surgery, time since FGRS and DR.

Power calculation

Although a power calculation was not carried out at study inception, we performed it at study completion. To detect a 50% difference in DR reported by parents of girls with CAH

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compared with other procedures (25% vs. 50% DR), a sample of 39 parents achieved 91% power at a 5% significance level.

Statistics

Categorical variables were analyzed using Fisher's exact test. Continuous variables were analyzed using the Mann–Whitney *U* (2 groups) and Kruskal–Wallis tests (3 groups). A critical $p = 0.05$ was used (Stata v10.1).

Results

Thirty-nine parents of 39 girls participated (36.8% response rate), including 10 of the last 11 (90.9%) eligible consecutive patients (Fig. 1). Twenty-three parents who initially expressed interest did not participate, some citing study fatigue and privacy concerns.

Parents participated at a median of 4.4 years after FGRS (Table 1). The majority of parents were mothers (94.9%). Children were 4.9 years old during the study. Salt-wasting CAH caused by 21-hydroxylase deficiency affected 92.3%. Preoperative Prader scale was documented for 17 girls: Prader 3 in 47.1%, 4 in 41.2%, and 5 in 11.8%. FGRS was performed at a median age of 9 months (range 2–37 months). FGRS was performed by a single surgeon in 37 girls. Overall, 89.7% of girls underwent a partial urogenital sinus mobilization, 87.2% vaginoplasty, and 94.9% clitoroplasty (82.1% had both).

Fifteen (38.5%) underwent a 6-month postoperative cystovaginoscopy to ensure adequate healing and vaginal caliber. Within the first 10 years of follow-up, two (5.1%) underwent another surgical procedure (labioplasty for redundancy, vaginoplasty for stenosis).

Decisional regret

Median DRS score was 0 (mean 5.0). Overall, 20.5% of parents reported some DR. All DR was mild to moderate, with no strong or very strong DR reported (Summary Figure). Parental DR was 0 for the two children who underwent another surgery. Of the last 10 consecutive patients, seven (70.0%) reported no DR and three mild DR (overall median DR 0, mean 5.5) at a median 4 months after FGRS.

On sensitivity analysis, 15.4% of parents reported DR with a 10-point cutoff, 5.1% with higher cutoffs, and 5.1% with non-neutral definition.

Risk factors of parental decisional regret

Parents were just as likely to report DR regardless of the child's age at FGRS, preoperative degree of virilization, being a mother, or undergoing another surgery ($p \geq 0.62$). Parents whose children underwent FGRS more recently appeared to be more likely to report DR (27.3% within 5 years vs. 20.0% at 5–10 years and 0.0% after 10 years), but this did not reach statistical significance ($p = 0.42$).

Comparison with parental decisional regret after other procedures

A systematic search yielded eight published articles [12,13,18,19,21–24] and one abstract [25] (Table 2). Two studies reported parental DR after genital surgery (hypospadias) and seven after non-genital procedures (3 oncology/immunology, 4 otolaryngology). One study reported 0% DR among 17 parents after tracheostomy, while remaining, larger studies reported DR in 40–92% of parents. The weighed mean DR was 60%.

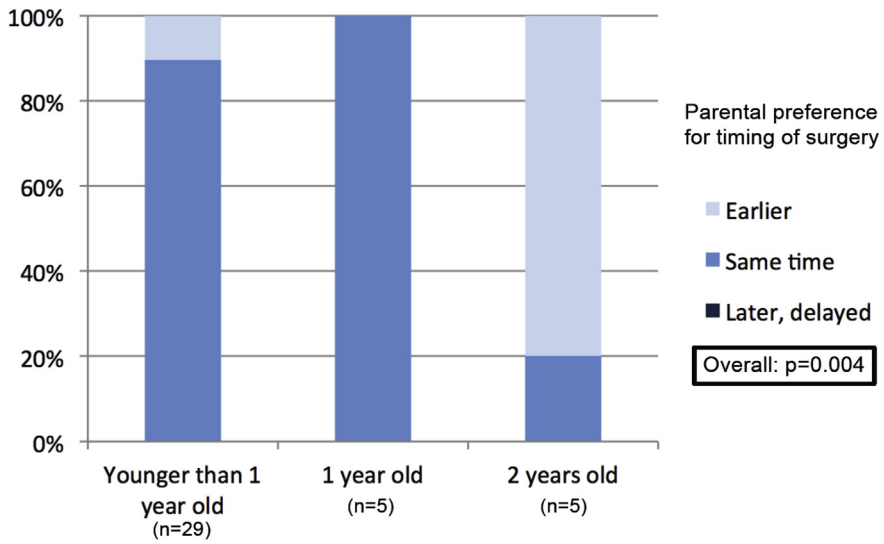

Figure 1 Parental preference for timing of surgery for children who underwent female genital restoration surgery at different ages. (No parents preferred delayed surgery.)

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Table 1 Population characteristics.

Variable	Parents of girls with CAH (n = 39)
CAH subtype	
21-Hydroxylase deficiency (all classic)	
Salt-wasting	35 (89.7%)
Simple virilizing	1 (2.6%)
17-Hydroxylase deficiency	1 (2.6%)
Unknown	2 (5.1%)
Age at FGRS (months; median, range)	9 (2–37)
Type of vaginoplasty	
None	5 (12.8%)
Cutback	1 (2.6%)
Pull-through	3 (7.7%)
Posterior sinus flap	6 (15.4%)
Posterior skin flap	24 (61.5%)
Urogenital mobilization	
None	4 (10.3%)
Partial	35 (89.7%)
Total	0 (0.0%)
Clitoroplasty	
None	2 (5.1%)
Folding only	2 (5.1%)
Erectile tissue excision and folding	34 (87.2%)
Erectile tissue/tunica albuginea excision, neurovascular bundle mobilization and folding	0 (0.0%)
Unknown technique	1 (2.9%)
Labioplasty	
None	0 (0.0%)
Labia minora	37 (94.7%)
Labia majora	37 (94.7%)
Preoperative Prader scale[a]	
3	8 (47.1%)
4	7 (41.2%)
5	2 (11.8%)
Patient age at time of questionnaire (years; median, range)	5.4 (11 months–18.6)
Parental data	
Mother	37 (94.9%)
Father	2 (5.1%)
Age (years; median, range)	38 (22–59)

[a] Preoperative Prader scale was not available for all patients.
(Numbers may not add up to 100% because of rounding.)

DR was reported by 50–92% of parents after hypospadias surgery, higher than after FGRS in our study ($p \leq 0.001$). Parental DR was reported by 41–45% after adenotonsillectomy and 61–72% after pediatric cancer or inflammatory bowel treatments was, higher than after FGRS ($p \leq 0.03$). Differences in parental DR after FGRS and other treatments did not reach statistical significance ($p \geq 0.05$).

Preferred timing of surgery

No parent preferred later or delayed FGRS and 82.1% preferred surgery to be performed at the same time it was done. Seven parents (18.1%) would have preferred FGRS to have been performed even earlier. Among parents of the last 10 consecutive patients, one mother expressed a preference for earlier surgery along with mild DR.

Risk factors of preferring for earlier surgery

Daughters of parents who preferred earlier surgery underwent FGRS at a median 24 months, compared with 8 months for parents who preferred FGRS at the same time it was performed ($p = 0.01$). In other words, parents were more likely to prefer earlier FGRS the later it occurred: 10.5% of parents preferred earlier FGRS when performed in the first year of life (3/29), 0% in the second year (0/5), and 80.0% in the third year (4/5) ($p = 0.004$) (Fig. 1). Preoperative virilization, mother, additional surgery, or having longer follow-up were not associated with preferring earlier surgery ($p \geq 0.63$). There was no association between preferring earlier surgery and DR ($p = 0.99$).

Responses from adult women

Four women participated (response rate 10.8%) at a median 29.0 years after FGRS, being 30.2 years old at the time of the study. All had salt-wasting CAH caused by 21-hydroxylase deficiency and were not initially managed by our team. FGRS occurred at a median age of 11 months (vaginoplasty in all, clitoroplasty in 3). All women had multiple prior procedures, typically using older techniques and approaches, before being referred to our center. They underwent a median of 2.5 general anesthetics in the first decade of follow-up, and 2 others in the second decade. Median DRS score was 0 (mean 3.8) and one woman reported mild DR (score of 15). No woman preferred earlier or later/delayed FGRS.

Discussion

We present the first report of validated long-term PROs after FGRS in infant and toddler girls with CAH. Most parents reported no DR. When present, it was typically mild. No one reported strong or very strong DR. Contrary to our hypotheses, parental DR may be lower after FGRS than after all reported genital and most non-genital pediatric procedures. Parents preferring even earlier surgery did not have higher levels of DR. Rather, their daughters tended to undergo FGRS after their second birthday.

The birth of a child with ambiguous genitalia is an extremely difficult time for parents. Generating accurate, honest, and meaningful data should be a priority for all CAH advocates, patients, and their families. Current arguments on both sides of the FGRS debate are based on anecdotal data and expert/advocate opinions. An analysis of DR, and other meaningful PROs, has never been reported for delayed surgery to allow for a comparison with our study. We strongly urge future researchers to not report physician interpretations of patient/parental views [26], as these are prone to bias, under-reporting, and underestimating patient concerns [27,28]. Patient and parental experiential expertise should be given a voice and be strongly considered in guiding CAH management, especially the value and timing of FGRS.

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Table 2 Studies on decisional regret reported by parents of children undergoing surgery or medical treatments.

Paper	Clinical scenario	*n*	No regret (0)	Mild regret (1—25)	Moderate regret (26—50)	Strong regret (51—75)	Very strong (76—100)	Median regret (IQR, range)	Mean regret
Current study	Female genital restoration surgery	39	79%	15%	5%	0%	0%	0 (0-0, 0—50)	5.0
Hypospadias									
Lorenzo et al. [12] (2014)	Distal hypospadias repair	116	50%	41%	9%			n/a	9[a]
Ghidini et al. [13] (2016)	Distal hypospadias repair	172	8%	52%	40%			n/a	n/a
Otolaryngology									
Hebert et al. [25] (2015)	Tracheostomy	17	100%	0%	0%	0%	0%	0	0
Hong et al. [24] (2016)	Otoplasty	62	60%	36%	3%	0%	0%	0 (0—5, n/a)	n/a
Hong et al. [21] (2016)	Adenotonsillectomy or tympanostomy tube insertion	64	55%	44%	2%	0%	0%	0 (0—15, 0—35)	7[a]
Carr et al. [19] (2016)	Adenotonsillectomy	94	59%	33%	10%	0%	0%	0 (n/a, 0—45)	9
Oncology and immunology									
Mack et al. [18] (2016)	Pediatric cancer treatment	346	39%	45%	15%	1%	0%	10 (0—20, n/a)	12
Lipstein et al. [22] (2016)	Biologics in inflammatory bowel disease or juvenile idiopathic arthritis	201	28%	32%	40%			n/a	18
Pentz et al. [23] (2016)	Pediatric stem cell transplant donation	30	n/a	n/a	n/a	n/a	n/a	0 (0—5, n/a)	n/a

[a] When not directly reported, values were calculated from published subgroups. Numbers may not add up to 100% because of rounding.

Reasons for the observed low levels of DR are multifactorial. Participants may have been satisfied with the original decision, experiencing low decisional conflict and good functional outcomes, factors linked to lower DR [12,20]. Unfortunately, these variables were not captured in this study. The decision regarding FGRS may not have been questioned because of other, constant and more imminent health concerns, such as death caused by an adrenal crisis. Although DR may increase over time, starting even 6 months after a decision [29], we did not detect this in our study.

Although it is a validated PRO, DRS is extremely sensitive and has no established cutoff to make it more meaningful to patients, families, and clinicians. It would be a misinterpretation to suggest that and one in five parents regret FGRS. Rather, DR after FGRS in infancy/toddlerhood is rare and may be lower than after most reported pediatric procedures. We did not detect an association between DR and wishes regarding the timing of surgery, potentially because no participants reported high regret and no participants wished for delayed surgery. Importantly, DR is conceptually distinct from anatomical and functional results, areas that were not addressed in this study.

Our study has several limitations. Study participants went through FGRS mostly by a single surgeon by age 3. Their views and opinions may not reflect those who underwent surgery elsewhere, in adolescence or adulthood, or not at all. Second, most participants were mothers of girls with Prader 3—4 virilization. Although neither DR nor preferred timing of surgery was associated with these variables, our results may be of limited generalizability to fathers or children with more significant virilization.

We received few responses from women with CAH and no responses from teenage girls after FGRS, although parents received a link to an anonymous survey for girls 14—17 years old. Based on the feedback received from parents, most did not want their daughter to fill out the survey. Low participation rates were likely a result of the sensitive nature of FGRS, a desire for privacy, and inability to locate patients. Participation has improved since hiring a nurse clinic coordinator (HF) and establishing an IRB-approved assessment of outcomes among recently treated patients and families.

Far from a definitive study, we aimed to provide individuals willing to share about their experience an opportunity to do so. For that reason, selection bias may exist in our study. Unlike many of the studies of parental DR after treatments performed on their children, not all participants were consecutive patients. While it is possible that those with higher DR were less likely to participate because of mistrust of the medical establishment, patients and families with a negative experience may in fact be more likely to voice their opinions. Indeed, most consecutive parents reported no DR and none preferred delayed

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surgery. No eligible participants who did not enroll responded with negative feedback. Future studies will focus on assessing reasons for when FGRS is preferred among patients and parents followed at multiple centers. A larger study will be required to assess how DR is affected by sexual function, menstruation, genital appearance, and complications.

Conclusions

Parents of girls with CAH report low levels of DR after FGRS performed in infancy and toddlerhood. This may be lower than after other reported genital and non-genital pediatric procedures. When present, DR is usually mild. Parents did not prefer delayed surgery, even among those with DR. Some preferred even earlier surgery.

Conflicts of interest

None.

Funding

None.

Acknowledgments

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Appendix A. Supplementary data

Supplementary data related to this article can be found at https://doi.org/10.1016/j.jpurol.2017.11.012.

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