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Exploring Factors Associated with Decisions about Feminizing Genitoplasty in Differences of Sex Development

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Abstract

Study Objective: Infants with genital development considered atypical for assigned female sex may undergo feminizing genitoplasty (clitoroplasty and/or vaginoplasty) in early life. We sought to identify factors associated with parent/caregiver decisions regarding genitoplasty for their children with genital virilization.

Design: Longitudinal, observational study.

Setting: 12 pediatric centers in the United States with multi-disciplinary DSD clinics, 2015–2020

Participants: Children <2 years with genital appearance atypical for female sex of rearing and their parents/caregivers.

Interventions/Outcome Measures: Data were extracted from the medical record on the child's diagnosis and anatomic characteristics pre-surgery. Parents/caregivers completed questionnaires on psychosocial distress, experience of uncertainty, cosmetic appearance of their child's genitalia, and demographics. Urologists rated cosmetic appearance. For 58 patients from the study cohort with genital virilization being raised as girls or gender-neutral, we compared these data across three groups based on the child's subsequent surgical intervention: (i) no surgery (n=5), (ii) vaginoplasty *without* clitoroplasty (V-only) (n=15), and (iii) vaginoplasty *and* clitoroplasty (V+C) (n=38).

Results: Fathers' and urologists' ratings of genital appearance were more favorable in the no-surgery group than in the V-only and V+C groups. Clitorophallic length was greater in the V+C group compared to the V-only group, with substantial overlap between groups. Mothers' depressive and anxious symptoms were lower in the no-surgery group compared to the V-only and V+C groups.

Conclusions: Surgical decisions were associated with fathers' and urologists' ratings of genital appearance, the child's anatomic characteristics, and mothers' depressive and anxious symptoms. Further research on surgical decision-making is needed to inform counseling practices.

Keywords

Intersex persons; Disorders of Sex Development; congenital adrenal hyperplasia

Introduction

Differences/disorders of sex development (DSDs) are a group of conditions characterized by genital and/or gonadal development that is atypical.[1] Since the 1950's, surgical intervention to alter the appearance of genital structures has been a common practice for individuals with DSDs.[2] However, in recent years, the decision-making process around genital and gonadal surgical procedures for individuals with DSDs has come under scrutiny, in the context of broader conversations about performing surgeries in patients too young to participate in the process. This has drawn attention to the need to determine factors associated with decision-making about early genital surgery in children with DSDs.[3] Vaginoplasty and clitoroplasty before age 2 years are considered by many pediatric surgeons and pediatric urologists to

be preferable options for the management of genital virilization in 46,XX children with congenital adrenal hyperplasia.[4] Perspectives on the optimal age for feminizing surgery vary considerably among adults with DSD.[5]

In this study, we sought to identify objective and subjective factors associated with parents'/caregivers' decisions to pursue feminizing genital procedures, namely clitoroplasty (reduction of the size of the clitorophallus) and vaginoplasty (vaginal reconstruction), typically performed in conjunction with mobilization/separation of a common urogenital sinus.[6] We conducted a secondary analysis of data from a study on outcomes for individuals with moderate to severe genital atypia. We hypothesized that decisions about surgery would be most closely associated with physical features such as the length of the clitorophallus and the degree of virilization of the external genitalia. We also explored whether decisions about surgery were associated with parents'/caregivers' assessment of cosmetic appearance of their child's genitalia and parents'/caregivers' psychosocial wellbeing.

Materials and Methods

We conducted a secondary analysis of baseline data collected by a longitudinal, observational, multicenter study of children with DSDs in the U.S.[7] The purpose of the broader study, which started in 2015, was to examine outcomes of early genital surgery for patients with moderate to severe genital atypia. Assessing surgical decision-making was not a specified goal of this study at its outset; rather, we re-analyzed data gathered by the study to identify associations with later decisions around surgery. We focused on individuals undergoing feminizing procedures, as these surgeries represent an area of controversy, due to the irreversibility of clitoroplasty in particular. Surgical outcomes in the study subcohort undergoing feminizing procedures have previously been described in detail.[6]

The study was approved by the IRB at each institution.[7] Written informed consent was obtained from at least one parent/caregiver of each participant. Inclusion criteria for the study were age <2 years at enrollment, Prader score 3–5, and lack of prior genitoplasty. Exclusion criteria were significant medical concerns involving non-urogenital organs and inability of parents/caregivers to complete questionnaires in English or Spanish. For this secondary analysis on decision-making around feminizing procedures, an additional criterion for inclusion in the subcohort was being raised female or nonbinary, as individuals raised male would generally not be considered for such procedures.

Surgical decision-making was conducted as part of usual clinical care conducted at each site; as this was an observational study, this study did not attempt to standardize the clinical team's counseling about surgery across sites. The study collected information regarding pediatric urologists' and/or endocrinologists' assessment of genital structures, including clitorophallic length.

As part of this study, parents/caregivers completed a demographic questionnaire (including self-report of relationship to the participant), the Beck Depression and Anxiety Inventories (BDI-II and BAI, respectively), the Parent Perception of Uncertainty Scale (PPUS), and a 4-point Likert scale rating of cosmetic appearance (1 = "good," 2 = "satisfied," 3 =

"dissatisfied," 4 = "very dissatisfied").[8] Urologists also rated cosmetic appearance. All measures were completed before the child underwent surgery, though it was not documented whether families had already made the decision to pursue surgery at the time when measures were completed.

Results are reported as mean ± standard deviation for continuous measures with symmetric sampling distributons (e.g., clitorophallic length, PPUS scores, parental age), as median [interquartile range (IQR)] for continuous measures with non-symmetric sampling distributons (e.g., Prader score, cosmetic score, BAI and BDI-II scores), and as count (percentage) for categorical measures. Cosmetic rating was treated as an approximately continuous variable. Statistical comparisons for these measures between groups used Student's t-tests (ST), Wilcox rank tests (WR), or Pearson chi-squared tests (PC), respectively. To identify factors associated with the decision to undergo clitoroplasty, the vaginoplasty *without* clitoroplasty (V-only) group was compared to the vaginoplasty *and* clitoroplasty (V+C) group. To identify factors associated with the decision to undergo any surgery, the no-surgery group was compared to both surgery groups combined. Additional comparisons examined factors associated with cosmetic rating scores. Given prior work showing differences in depressive and anxious scores between mothers and fathers,[8] we analyzed mothers and fathers separately. R (version 3.6.1) was used to perform calculations and to create figures. Results corresponding to p-values < 0.05 are described as significant. Cohen's effect size (D), the degree of difference between two variables, is also reported; a difference was considered large when $D > 0.7$.

Results

Fifty-eight patients were included in this secondary analysis and were categorized into three groups based on surgical procedures performed: no surgery (n=5), vaginoplasty *without* clitoroplasty (V-only, n=15), and vaginoplasty *and*clitoroplasty (V+C, n=38). Our analyses included 38 pairs of mothers and fathers and an additional 13 mothers (51 mothers total; all parents/caregivers self-identified as mothers or fathers). Patient diagnoses included congenital adrenal hyperplasia (n=50), ovotesticular DSD (n=2) partial androgen insensitivity syndrome (n=1), and sex chromosome mosaic DSD (n=3), trisomy 21 with virilization (n=1), and unknown (n=1) [6,7] (Supplemental Table 1).

Physical Features

We first tested whether decisions about surgery were associated with differences in the children's physical features. Prader score[9] (a scale describing the degree of genital virilization) did not differ significantly between the three groups (Figure 1). Clitorophallic length was significantly lower in the V-only group compared to the V+C group (1.6±0.9 vs. 2.4±0.8 cm, ST:p=0.009, D=0.9), but despite these overall group differences, there was substantial overlap in clitorophallic length between these two surgical groups (Figure 2). Clitorophallic length did not differ significantly between the no-surgery group and the two surgery groups combined (Figure 2).

We next examined whether parent or urologist ratings of cosmetic appearance of the external genitalia prior to surgery differed between groups and compared cosmetic ratings from

mothers, fathers and urologists of children enrolled in the study (Figure 3). On a 4-point Likert scale (with higher scores indiating greater dissatisfaction), mothers' pre-surgical cosmetic scores did not differ between the surgery groups (Figure 3A) and were not associated with clitorophallic length or Prader score (Figure 4A). Fathers in the V-only group reported greater satisfaction with cosmetic appearance prior to surgery than fathers in the V+C group (2.0 [1.0–3.0] vs. 3.0 [2.0–3.0], WR:p=0.03, D=0.8). There was no significant difference in fathers' cosmetic ratings between the no-surgery group and the two surgery groups combined (2.0 [1.8–2.3] vs. 3.0 [2.0–3.0], WR:p=0.29, D=0.56) (Figure 3B). Stretched clitorophallic length did not differ significantly between children of fathers who provided less vs. more favorable cosmetic ratings (1.8±1.0 vs. 2.3±0.7 cm, ST:p=0.07, D=0.6). Prader score was not associated with fathers' cosmetic ratings (Figure 4B).

Urologists reported less satisfaction with cosmetic appearance in the two surgery groups compared to the no-surgery group (3.0 [3.0–4.0] vs. 2.0 [1.0–3.0], WR:p=0.003, D=1.67) (Figure 3C). Urologists' cosmetic ratings of "dissatisfied" or "very dissatisfied" were associated with higher Prader scores than ratings of "good" or "satisfied" (3.0 [3.0–4.0] vs. 3.0 [3.0–3.0], WR:p=0.04, D=0.87; Figure 4C). The difference in clitorophallic length between children whose urologists provided less vs. more favorable cosmetic ratings was not statistically significant (2.3±0.9 vs. 1.4±0.8 cm, ST:p=0.051, D=1.0).

Parents' Psychological Measures

In addition to investigating the association between surgical decisions and a child's physical features, we compared parents' level of depressive and anxious symptoms between groups. Psychosocial measures were completed prior to surgery but not at a specific point relative to the decision-making process; measures may have been completed before, during, or after the decision-making process.

Mothers in the no-surgery group had lower anxious symptom scores than mothers in the two surgery groups (0.5 [0.0–1.3] vs. 11.0 [3.0–16.0], WR:p=0.006, D=1.23; Figure 5A). Mothers of children who did not undergo surgery also had lower depressive symptom scores (which were highly correlated with anxious symptom scores; Figure 6) than mothers of children who underwent surgery (6.0 [4.5–6.0] vs. 10 [5.0–19.5], WR:p=0.05, D=0.79) (Figure 5B). While scores for both anxious and depressive symptoms in the V-only and V+C groups ranged from the normal to severely elevated range, no elevated scores were observed in the no-surgery group (Figure 5).

Fathers' scores for anxious and depressive symptoms did not differ significantly between the three groups (Figures 5C & 5D). Mothers' and fathers' scores on the PPUS, which measures parental uncertainty associated with the child's diagnosis, also did not differ between the groups (Supplemental Figure 1). Of note, no correlation was noted between parent satisfaction with cosmetic appearance of child's genital structures and anxious/depressive symptom scores for mothers or fathers.

Other Factors

In addition to physical features and psychological adjustment inventories, we also examined demographic factors. We hypothesized that families may adjust to a diagnosis over time, and

thus parents of older children may tend not to pursue surgery. However, we did not find an association between patient age at enrollment and surgical group. We also examined parental age and found no association between maternal or paternal age and surgical decision. Finally, we evaluated whether parental education was associated with surgical decision but did not find a correlation

Discussion

We examined factors associated with decisions around genital surgery among families of children with DSDs being raised as girls or gender-neutral across 12 centers with specialized clinical programs for the care of individuals with DSDs. We initially hypothesized that the greatest differences between surgical groups would lie in objective features such as clitorophallic length and Prader score. Indeed, we found differences in clitorophallic length between children who underwent vaginoplasty only (V-only group) and those who underwent both vaginoplasty and clitoroplasty (V+C group), However, we found no differences in objective features between those who underwent no surgery and those who underwent vaginoplasty with or without clitoroplasty. Urologist ratings of genital appearance differed between the no-surgery group and the two surgery groups in our analysis, a finding that may point to the important role of counseling by urologists, and potentially by other clinical providers, in the decision-making process.

Not surprisingly, we observed a significantly greater average clitorophallic length among children who underwent both vaginoplasty and clitoroplasty compared to those who underwent vaginoplasty alone. However, there was substantial overlap between groups, and there was no threshold for clitorophallic length below which parents/caregivers decided to forgo clitoroplasty for their children. Thus, while clitorophallic length may influence the decision about clitoroplasty, it is not the sole factor contributing to this decision. Fathers' ratings of cosmetic appearance differed between the V+C and V-only groups, and understanding the factors influencing fathers' cosmetic ratings may improve understanding about surgical decision-making. Furthermore, occasional differences between maternal, paternal, and urologist cosmetic ratings demonstrate a potential need for improved communication between medical providers and parents around indications for clitoroplasty and vaginoplasty and expected outcomes for each procedure.

We next examined whether there were associations between psychosocial characteristics and surgical group. We found associations with maternal symptom scores for anxiety and depression, with lower average scores in the no surgery group compared to the V-only V+C groups. There are several non-mutually exclusive possibilities for these differences in depressive/anxious symptom scores between mothers of children in the no-surgery group compared to those in the two surgery groups. First, because parents may have completed questionnaires before, during, or after the decision-making process, the decision to have surgery could itself have heightened distress for some mothers, though this does not explain why similar differences were not seen for fathers. This could be mediated by conflicting feelings about surgery, challenges of the decision-making process (which could include potential partner and/or extended-family pressure and concerns about conforming to advice

from clinical providers), and/or uncertainty about surgical outcomes (though we did not observe significant differences between groups on a measure of disease-related uncertainty).

A second possibility is that increased maternal distress and the decision to pursue surgery could be caused by one or more common antecedents. Factors we examined that might influence both included child's Prader score, clitorophallic length, ratings of cosmetic appearance, and perceived uncertainty about the child's condition, but these were not significantly correlated with either maternal distress or the decision to pursue surgery in our analysis. Other factors, unexamined in our study, could include concern about consequences of the DSD condition, including psychosocial consequences such as stigma (experienced by the child, the family, or both), gender dysphoria, and physical consequences on urinary and reproductive function. Distress could also be caused by disagreement between caregivers or caregivers and members of the medical team regarding goals for the child's care.

These potential explanations for maternal distress are consistent with previous work indicating that a third of parents/caregivers of children with DSD experienced marked distress in association with recalling their child's initial diagnosis, as well as subsequent medical and surgical interventions.[10] Parents identified particular distress related to receiving conflicting medical diagnostic information, as well as uncertainty about their child's future gender identity, and the fear of the child experiencing gender dysphoria.[10,11] Additionally, disagreement between parent/caregivers and members of the medical team has been identified as a challenge in medical decision-making for the care of children with DSDs.[11]

The third possibility is that maternal psychosocial distress could influence decision-making around surgical intervention. For instance, mothers who are highly distressed may, whether consciously or subconsciously, view surgery as a way to relieve anxiety and depression related to their child's physical differences. Indeed, in our cohort, distress did decrease over time after surgery for most parents, though not all, as previously reported.[8] Qualitative work has indicated that surgical reconstruction of atypical genital structures provided relief from distress for some parents of children with DSD, but for other parents, surgery reduced but did not eliminate anxiety about the impact of DSDs in later childhood and adolescence due to perceived differences in genital appearance.[12] It may also be that others involved in the decision-making process (e.g., fathers, clinicians) respond to maternal distress by favoring surgery, though once again, it is difficult to explain why anxiety and depression scores would have been different in mothers and fathers. It is worth noting that in our study, as in much pediatric psychology research,[13] more mothers participated than fathers, which could have led to capture of greater variability in depressive/anxiety scores in mothers than fathers.

Notably, maternal distress was not uniformly high in the surgical groups. While mothers in the no-surgery group all had low symptom scores for anxiety and depression, scores for mothers in the V-only and V+C groups ranged widely, from low (35.7% of V-only and 44.4% of V+C had low anxiety symptom scores) to clinically significant (28.6% of V-only and 22.2% of V+C had clinically significant levels of anxiety symptoms). Thus, the decision to undergo surgery is not solely explained by maternal distress.

One limitation of this study is that while questionnaires were completed prior to the actual surgical procedure, they may have been completed before, after, or during the period during which decision-making around surgery occurred (if such a period can even be discretely defined); this limits our ability to understand the potential causal pathways underlying the associations observed in this study. Similarly, physical exams performed to assess child's Prader score and clitorophallic length could have been completed anytime in the child's life prior to genitoplasty. It is possible that the size and appearance of genital structures may have varied over time for some individuals as infants grew and/or were treated with medications that suppress androgen production. [14,15] Also, the measurement of clitorophallic length and use of the Prader scales may have varied between providers. The study did not collect information regarding surgeons' reasons for performing clitoroplasty in patients whose clitorophallic length fell within the range considered typical for a clitoris in infants and young children (3–10mm.)[16]

Other factors contributing to urologists' counseling were not uniformly documented. Specifically, existence of pre-operative medical concerns related to genitourinary anatomy, such as history of urinary tract infection or obstruction were not collected. Additionally, at all of the sites participating in this study, pediatric urologists were the subspecialists providing surgical care; other surgical subspecialists such as pediatric gynecologists and general pediatric surgeons may approach counseling and clinical care differently. The study did not collect data on details of psychosocial counseling and support for the decision-making process. There was likely variation across sites, given the limited guidance on the optimal approach to psychosocial support of children with DSD and their families, including discussions about gender identity.[3,17]

Another important limitation of this study was the small size of the no-surgery group, though the differences in urologist cosmetic ratings and maternal psychosocial scores were sufficiently pronounced to be statistically significant even with our limited sample size. Nevertheless, the small sample size may limit the generalizability of our findings. Furthermore, our study had statistical power to detect only large differences between groups. Also, because most patients in our analysis had CAH, we were unable to determine the role of the underlying diagnosis in surgical decision-making. Additionally our data do not capture parents' anxious and depressed symptoms prior to having a child with a DSD, which makes the underlying cause of increased distress in the surgical groups difficult to ascertain. Parents of children enrolled in the study were treated as independent actors for our analyses. This may have neglected the interrelatedness of some of our outcomes, e.g., that paternal dissatisfaction with cosmesis may have impacted maternal anxious/depressive symptom scores. Our findings suggest many areas for future research. Understanding any potential influence of parental anxiety and depression on decision-making is of paramount importance and could lead to interventions to reduce parental stress during a trying time. Similarly, identifying additional factors that may influence decision-making around surgery for children with DSDs – such as caregiver and provider values, family dynamics, cultural or religious influences, and goals for the urogenital function for their child – would highlight key topics for education, discussion, and counseling during the decision-making process. Counseling practices, provider biases, cultural factors, and family dynamics – and variation in these factors across sites – are additional areas for subsequent investigation. Most

importantly, data continue to be needed on long-term outcomes for individuals who undergo or do not undergo feminizing procedures in infancy and early childhood so decisions can be informed by empirical evidence.

Conclusions

Our findings have several implications for current clinical practice. First, clinicians must assess whether their own goals for outcomes of genital surgery might differ from those of parents/caregivers, or if parents/caregivers may not be in agreement about surgical decisions. Identifying potential differences in perspective between caregivers and/or between caregivers and the clinical team represents an important step in collaborative decision-making for clitoroplasty and vaginoplasty in patients with virilizing DSDs. Second, explicitly assessing and addressing the distress caregivers may be experiencing can help to ensure that decision-making incorporates their concerns while preserving and prioritizing the interests of the child in both the short and long term. Clinicians should make certain, to the extent possible, that parental distress does not impair caregivers' ability to receive, comprehend, and process all of the information relevant to surgical decisions for their children. Finally, our findings suggest that ongoing psychosocial support for caregivers may be paramount in ensuring optimal outcomes for both patients and caregivers affected by DSDs.

Supplementary Material

Refer to Web version on PubMed Central for supplementary material.

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Abbreviations:

DSDs	Differences/disorders of sex development
V+C	vaginoplasty and clitoroplasty
V-only	vaginoplasty only

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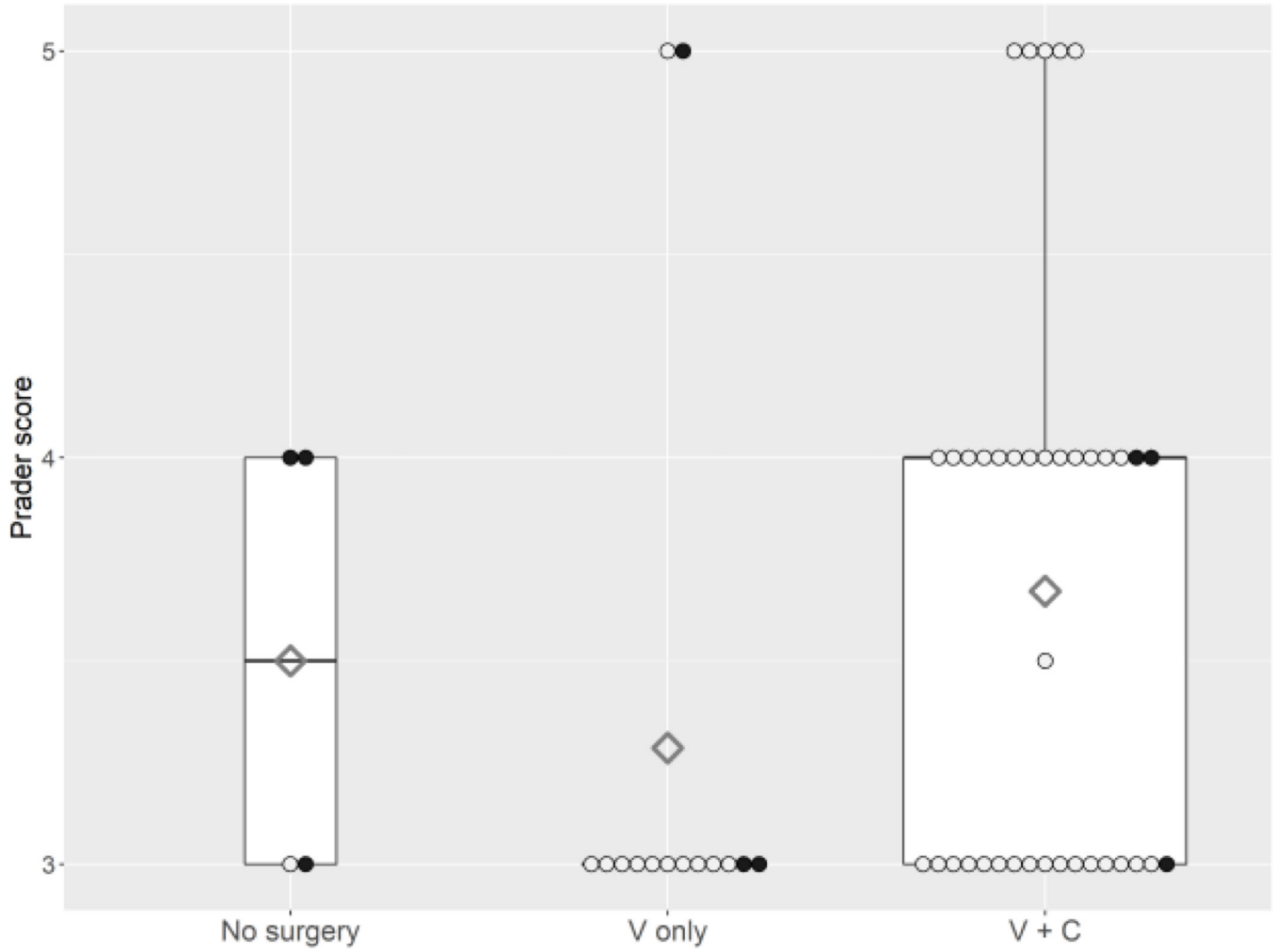

Figure 1.

Prader scores in each surgical group. Higher scores indicate greater virilization of the external genitalia. Darker horizontal lines are medians; diamonds are means; circles are individual data points (open circles denote patients with CAH and closed circles denote patients with a diagnosis other than CAH). Boxes represent the inter-quartile range; whiskers (vertical lines) indicate the 95th percentile of the distribution. V only, vaginoplasty only; V + C, vaginoplasty and clitoroplasty.

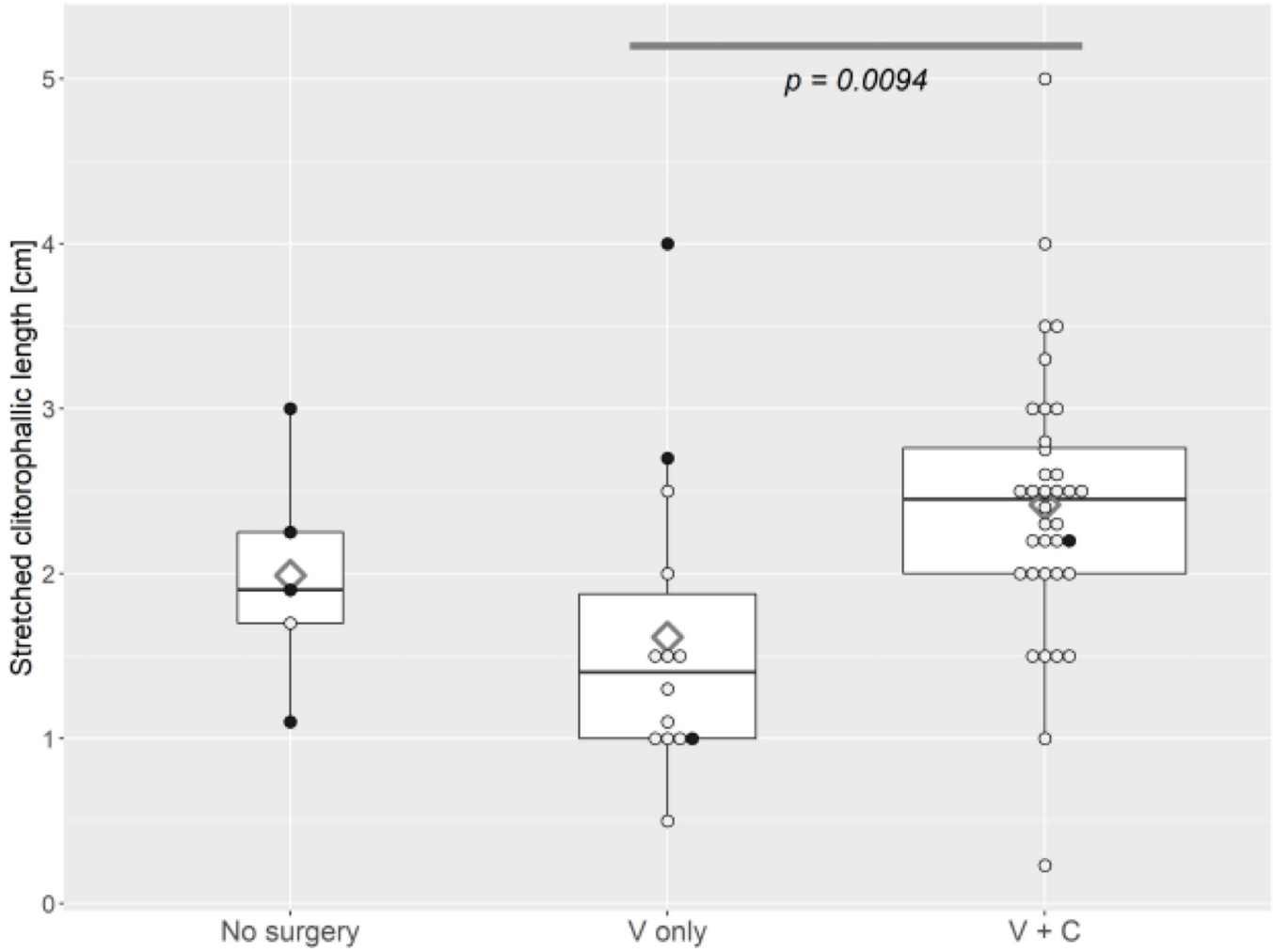

Figure 2.

Stretched clitorophallic length in each surgical group. Darker horizontal lines are medians; diamonds are means; circles are individual data points (open circles denote patients with CAH and closed circles denote patients with a diagnosis other than CAH). Boxes represent the interquartile ranges. Whiskers (vertical lines) indicate the 5th and 95th percentiles of the distribution. V only, vaginoplasty only; V + C, vaginoplasty and clitoroplasty.

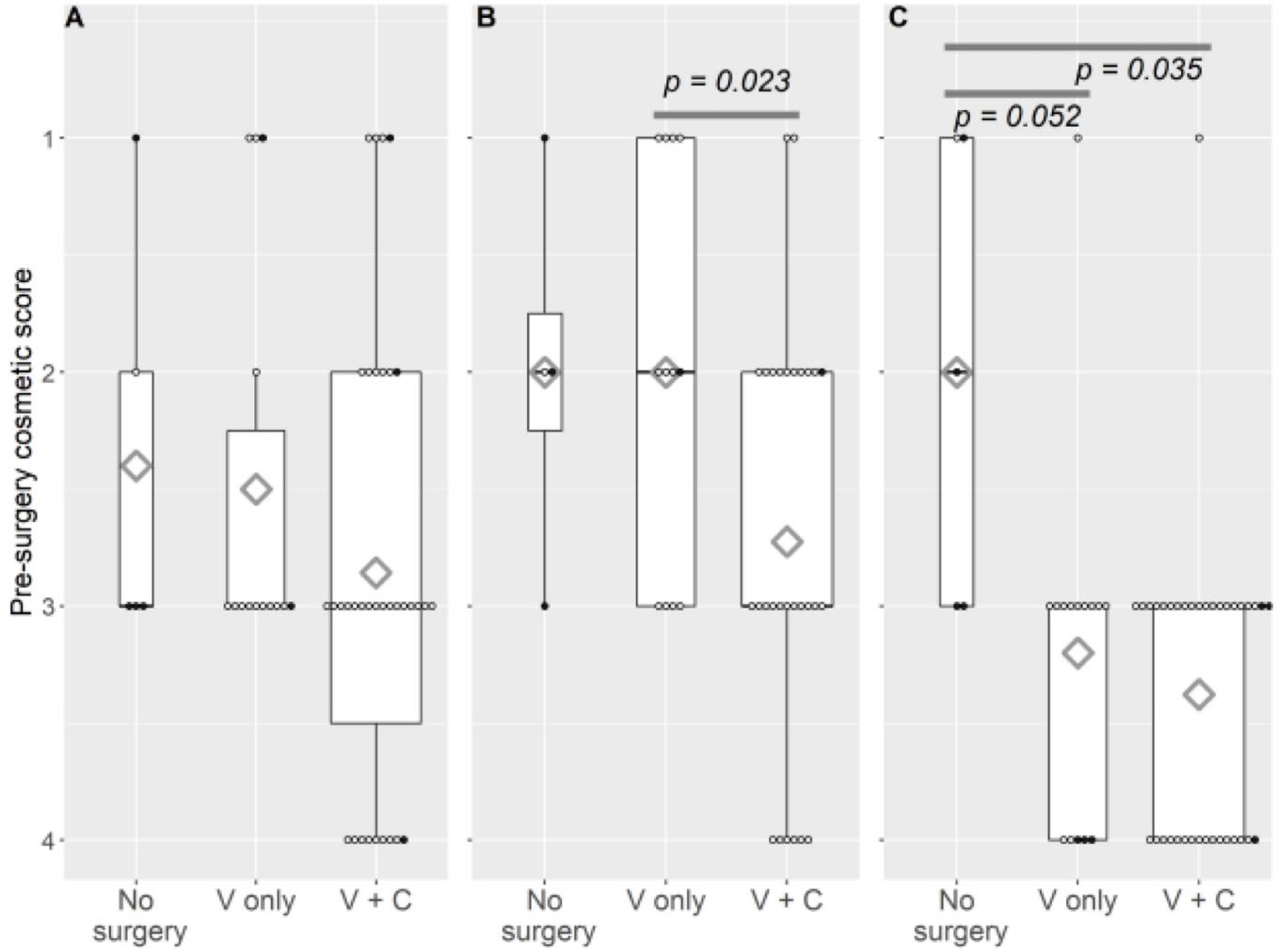

Figure 3.

Pre-surgery ratings of the cosmetic appearance of the child's genitals, rated by A) mothers, B) fathers, and C) urologists. Higher scores indicate less satisfaction. Darker horizontal lines are medians; diamonds are means; circles are individual data points (open circles denote patients with CAH and closed circles denote patients with a diagnosis other than CAH). Boxes represent the interquartile ranges. Whiskers (vertical lines) indicate the 5th and 95th percentiles of the distribution. V only, vaginoplasty only; V + C, vaginoplasty and clitoroplasty.

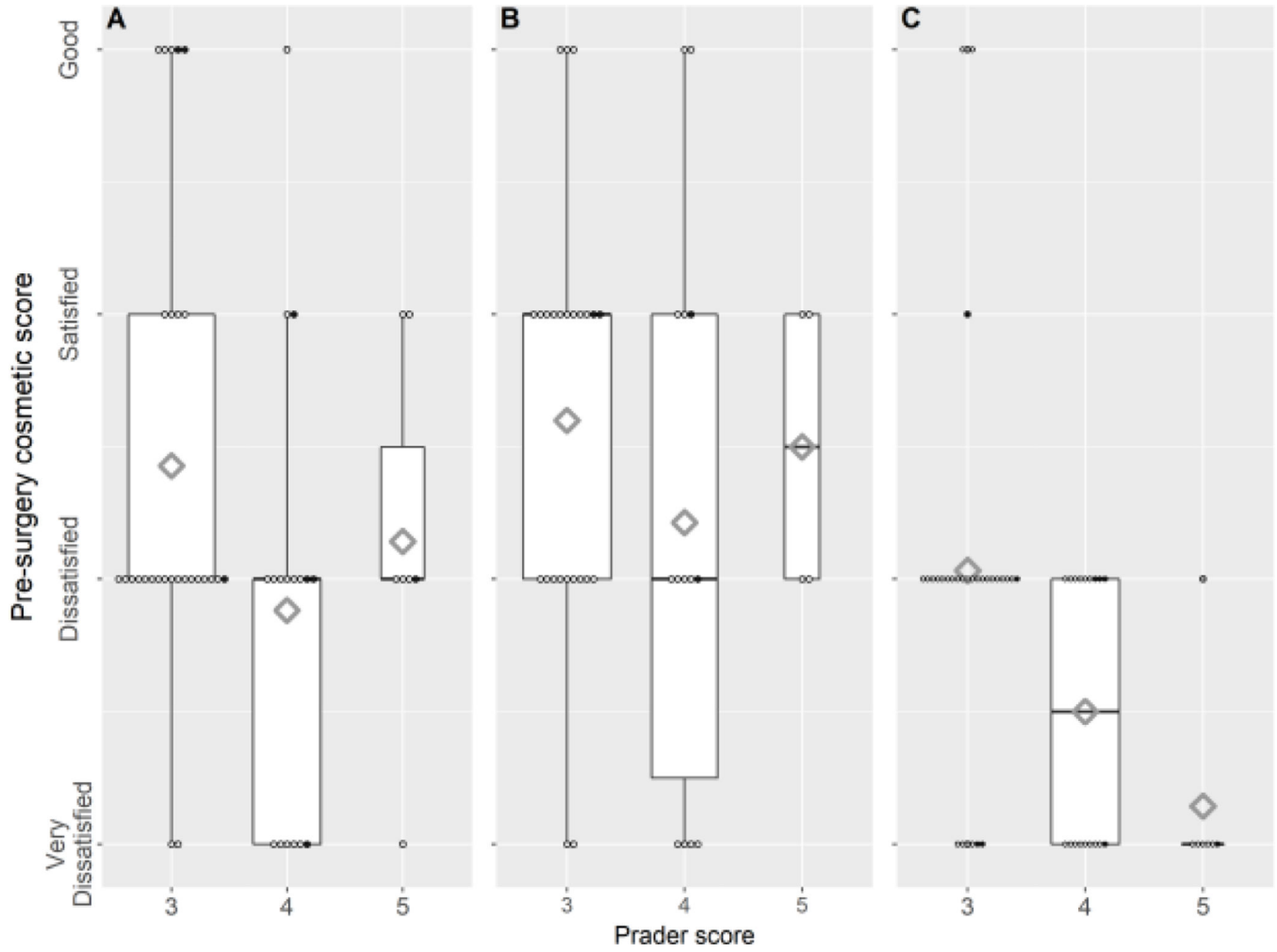

Figure 4.

Relationship between Prader score and pre-surgery cosmetic score of A) mothers, B) fathers, and C) urologists. Higher scores indicate less satisfaction. Darker horizontal lines are medians; diamonds are means; circles are individual data points (open circles denote patients with CAH and closed circles denote patients with a diagnosis other than CAH). Boxes represent the interquartile ranges. Whiskers (vertical lines) indicate the 5th and 95th percentiles of the distribution.

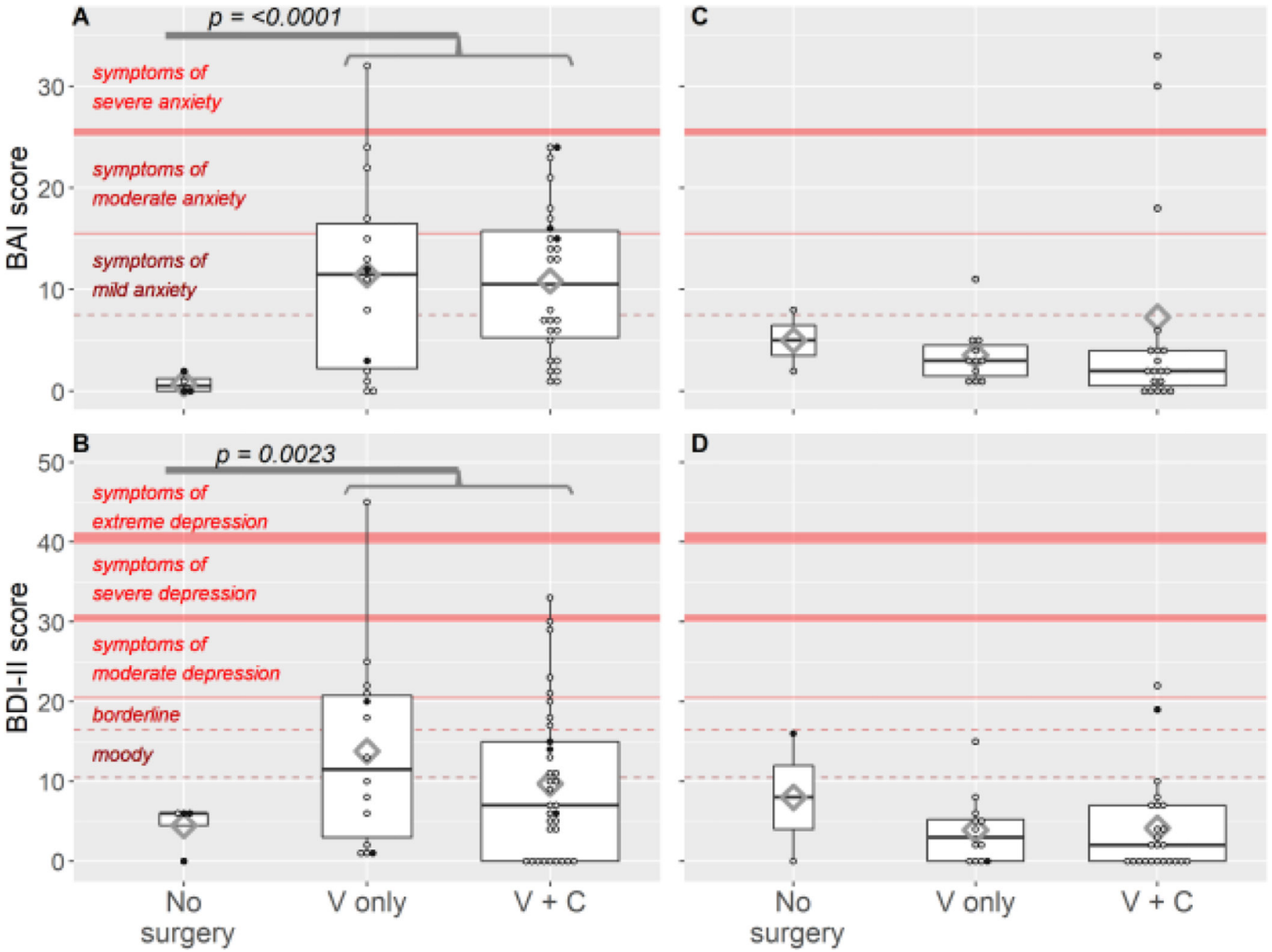

Figure 5.

Beck Anxiety Inventory (BAI) and Beck Depression Inventory (BDI-II) scores for mothers (A & B, respectively) and fathers (C & D, respectively). Darker horizontal lines are medians; diamonds are means; circles are individual data points (open circles denote patients with CAH and closed circles denote patients with a diagnosis other than CAH). Boxes represent the interquartile ranges. Whiskers (vertical lines) indicate the 5th and 95th percentiles of the distribution. V only, vaginoplasty only; V + C, vaginoplasty and clitoroplasty.

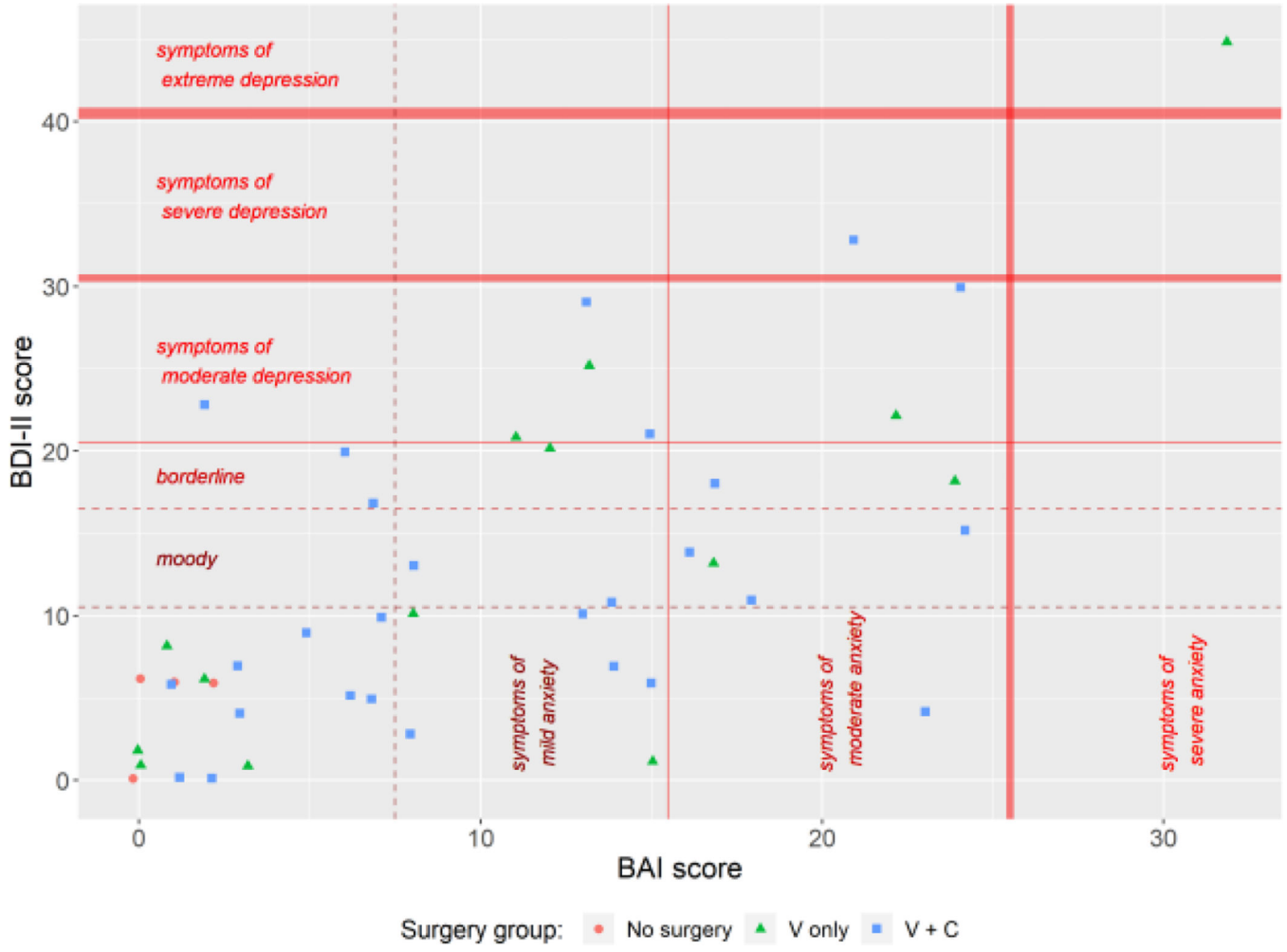

Figure 6.

Correlation between maternal symptom scores for anxiety and depression. BAI, Beck Anxiety Inventory; BDI-II, Beck Depression Inventory; V only, vaginoplasty only; V + C, vaginoplasty and clitoroplasty.