World Journal of Pediatric Surgery

# Predictors of surgical complications in boys with hypospadias: data from an international registry

Kathryn Scougall,[1] Jillian Bryce,[1] Federico Baronio,[2] Rachel L Boal,[3] Jose Roberto Castera,[4] Sebastián Castro,[4] Tim Cheetham,[3] Eduardo Corrêa Costa,[5] Feyza Darendeliler,[6] Justin H Davies,[7] Mirjam Dirlewanger,[8] Gabriella Gazdagh,[7] Evgenia Globa,[9] Gil Guerra-Junior,[10] Tulay Guran,[11] Gloria Herrmann,[12] Paul-Martin Holterhus,[13] Ahsen Karagözlü Akgül,[14] Renata Markosyan,[15] Kenneth McElreavey,[16] Marcio Lopes Miranda,[10] Anna Nordenstrom,[17] Stuart O'Toole,[18] Sukran Poyrazoglu,[6] Gianni Russo,[19] Valerie Schwitzgebel,[8] Marianna Stancampiano,[19] Michael Steigert,[20] S Faisal Ahmed,[1] Angela K Lucas-Herald [1]

**To cite:** Scougall K, Bryce J, Baronio F, *et al*. Predictors of surgical complications in boys with hypospadias: data from an international registry. *World J Pediatr Surg* 2023;**6**:e000599. doi:10.1136/wjps-2023-000599

Received 24 March 2023
Accepted 25 August 2023



For numbered affiliations see end of article.

**Correspondence to**
Dr Angela K Lucas-Herald; angela.lucas-herald@glasgow.ac.uk

## ABSTRACT

**Background** Complications are frequently reported after hypospadias repair and there is a need to understand the factors that influence their occurrence.

**Methods** Data from boys with hypospadias born between 2000 and 2020 were obtained from the International Disorders of Sex Development (I-DSD) Registry. Logistic regressions, fisher's exact tests and spearman's correlation tests were performed on the data to assess associations between clinical factors and complication rates.

**Results** Of the 551 eligible boys, data were available on 160 (29%). Within the cohort, the median (range) External Masculinization Score (EMS) was 6 (2, 9). All presented with one or more additional genital malformation and 61 (38%) presented with additional extragenital malformations. Disorders of androgen action, androgen synthesis and gonadal development were diagnosed in 28 (18%), 22 (14%) and 9 (6%) boys, respectively. The remaining 101 (62%) patients were diagnosed as having non-specific 46,XY Disorders of Sex Development. Eighty (50%) boys had evidence of abnormal biochemistry, and gene variants were identified in 42 (26%). Median age at first hypospadias surgery was 2 years (0, 9), and median length of follow-up was 5 years (0, 17). Postsurgical complications were noted in 102 (64%) boys. There were no significant associations with postsurgical complications.

**Conclusions** Boys with proximal hypospadias in the I-DSD Registry have high rates of additional comorbidities and a high risk of postoperative complications. No clinical factors were significantly associated with complication rates. High complication rates with no observable cause suggest the involvement of other factors which need investigation.

### WHAT IS ALREADY KNOWN ON THIS TOPIC

⇒ Surgical correction of hypospadias is often associated with high rates of complications.
⇒ It is not clear what the predictors are for surgical complications.

### WHAT THIS STUDY ADDS

⇒ Nearly two-thirds of boys with proximal hypospadias in the International Disorders of Sex Development Registry have postoperative complications.
⇒ No clinical factors were significantly associated with complication rates.

### HOW THIS STUDY MIGHT AFFECT RESEARCH, PRACTICE OR POLICY

⇒ High complication rates with no observable cause suggest the involvement of other factors which need investigation.
⇒ Further studies are needed to identify novel clinical factors which may be associated with complication rates in this complex group.

## INTRODUCTION

Hypospadias, a congenital defect of the male external genitalia, occurs in 1 in 300 live births.[1] Surgical correction is the only treatment option, with postoperative complication rates reported to range from 8% to 56%.[2 3] These disparities are largely explained by variability in hypospadias phenotypes. A urethral opening at the distal end of the penis is classed as a mild hypospadias and accounts for 70% of all hypospadias cases, while an opening at the mid or proximal end of the penis is classed as severe hypospadias and accounts for the remaining 30% of cases.[4] Severe forms are often found in combination with a smaller glans, less penile skin, greater curvature, and poor urethral plate quality, which are associated with more challenging surgeries and with poorer postsurgical outcomes.[5] In addition, severe hypospadias may represent differences/disorders of sex development (DSD), a term used to describe a wide range of

complex congenital disorders in which genetic, chromosomal, gonadal or anatomical sex is abnormal,[6] resulting in atypical formation of the reproductive system. These boys often present with additional genital defects (cryptorchidism, bifid scrotum and/or micropenis) and non-genital defects.[7 8]

Postsurgical complications can negatively impact long-term functional, cosmetic, and psychological outcomes. Although various clinical factors, such as phenotype, age at operation, the surgeon's experience, surgical technique and length of follow-up have all been associated with postoperative complications,[9] they do not fully explain complication rates. It has been hypothesized that novel clinical factors may be associated with postsurgical complications and could therefore be used as predictive tools. Previous studies have indicated that boys with a genetically confirmed androgen receptor (*AR*) mutation or a co-occurring extragenital congenital malformation, are at increased risk of postsurgical complications.[8 10] However, further investigation is required to confirm these associations. By characterizing this population and identifying novel predictive factors of postsurgical outcomes, alterations in both diagnosis and management can be made to optimize patient care and minimize the chances of complications occurring, thus improving short-term and long-term outcomes. As such, the aim of this study was to assess complication rate in an international cohort and identify if any clinical factors can predict surgical outcomes for boys with hypospadias.

## METHODS

Retrospective data were collected using the internationalDSD (I-DSD) registry based on the following inclusion criteria: XY boys raised as male, with hypospadias, born between 2000 and 2020. Data were collected on year of birth, birth weight, gestational age, biochemistry at first presentation, presence of co-occurring genital malformations (bifid scrotum, micropenis, unilateral or bilateral undescended testes), presence of co-occurring extragenital malformations, karyotype, presence of genetic mutations, number of hypospadias surgeries, type of surgical technique used, age at first surgery, number of re-do operations, presence of postsurgical complications, details of postsurgical complications and length of surgical follow-up.

Distal hypospadias were categorized by a meatal opening in the glanular and coronal regions. Mid hypospadias were categorized by a meatal opening in the subcoronal and midshaft regions. Proximal hypospadias were categorized by a meatal opening in the proximal shaft, penoscrotal, scrotal or perineal region. Using the information collected, the External Masculinization Score (EMS)[11] was calculated for each boy. Biochemical results were categorized as 'high', 'low', 'normal' or 'unknown'. Micropenis was defined as a phallus length below 2.5 cm at birth or 2.5 standard deviation (SD) below the average length at time of presentation. Chordee was reported as per the surgeon's opinion with no specific definition. Single-staged procedures were defined as a repair intended to be completed in a single operation and staged procedures were defined as repairs intended to be completed in two or more operations. A re-do operation was defined as the need for any additional operation after previous hypospadias repair. Small for gestational age (SGA) was defined according to the reporting center's local practice. A complication was defined as anything that would require medical or surgical intervention as a direct consequence of the hypospadias surgery.

Statistical analysis was performed using R Studio (V.4.2.0) and Graphpad Prism V.8.0. Logistic regressions were performed to assess whether endocrine abnormalities, disorder type, procedure type, age at first surgery, the presence of extragenital malformations, presence of genetic variants and EMS could predict the occurrence of postsurgical complications. To account for the effects of collinearity, separate logistic regressions were run for each variable (ie, the independent variable). Multivariate logistic regression was undertaken and Spearman's correlation tests were performed to assess the correlation between numeric variables. Fisher's exact tests were performed to further assess the association between complication rate and subgroups of clinical factors. P values <0.05 were deemed statistically significant.

All patients had to provide written consent to be included in a pseudo-anonymised format on the I-DSD Registry. Full details regarding the Standard Operating Procedures (SOP) of the I-DSD Registry can be found at: home.i-dsd.org.

## RESULTS

Of the 53 eligible centers on the I-DSD registry, 16 (30%) were able to provide data on 243 boys with an additional 25 obtained from the surgical records of one participating center (figure 1).

### Cohort characteristics

The characteristics of the cohort are summarized in table 1. At the time of the study, the median age of the cohort was 9 years old (2, 22).

Of the 160 boys, 101 (63%) received a clinical diagnosis of non-specific 46,XY DSD. Of the remaining 59 (37%) with a specific disorder type, genetic variants were found in 33 (56%). Sixteen out of 59 (27%) boys were diagnosed based on abnormal biochemistry alone, while 10 (17%) were diagnosed through clinical observation alone. The median EMS of the cohort was 6 (2, 9), indicating a general trend of severe under masculinization.

Of the 160 boys included in the study, 100% had one or more genital malformation. Of the 160 boys, 30 (19%), 34 (21%), 58 (36%) and 25 (16%) had 1, 2, 3

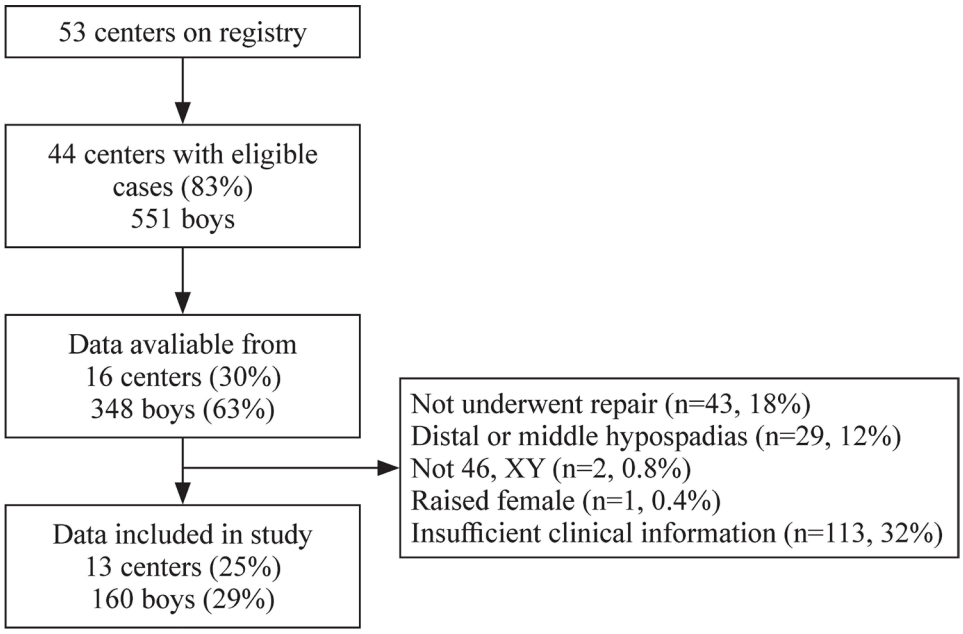


**Figure 1** Consort diagram of included data.

and 4 additional genital malformations present, respectively. The remaining 13 (8%) has ≥2 additional genital malformations; due to missing data, the exact number was unknown. One hundred and thirty-nine (87%) patients had chordee, 101 (63%) presented with micropenis, 74 (46%) had undescended testes and 57 (36%) had a bifid scrotum. Of those with undescended testes, 46 (62%) had bilateral undescended testes and 28 (38%) had unilateral undescended testes.

In terms of extragenital malformations, 63 (39%) presented with at least one additional extragenital malformation, most commonly SGA (figure 2).

### Endocrine and genetic testing

Of the 142 (89%) boys who underwent endocrine testing, 80 (56%) had abnormal biochemistry and 62 (44%) had normal biochemistry. Of the eight different endocrine evaluations requested in this study, 89 (63%) boys received four or more of these tests. Of the 160 boys, 135 (84%) received genetic testing. Variations in 17 genes were identified in 42 (31%) boys (table 2).

### Hypospadias surgery

Median age at first hypospadias surgery was 2 years (0, 9 years) and the median length of clinical follow-up following first hypospadias surgery was 5 years (0,17). Staged operations were the planned surgical method for 102 (64%) boys. The median number of re-do operations was 1 (1, 7) and re-do operations were performed on 75 (47%) of boys.

Of the 160 boys, 102 (64%) experienced at least one complication. The median number of complications within the cohort was 1 (0, 7). Following the first hypospadias repair, the first documented complication arose after a median of 1 year (0, 11 years). Forty-five percent of these complications occurred 2 or more years after first surgery. The most common complications were fistulae (n=45, 44%), abnormal voiding (n=25, 25%), and abnormal meatal shape or position (n=23, 23%).

No single clinical factor investigated was significantly associated with postsurgical complication risk (table 3). In total, those with a known disorder had a complication rate of 62%. No specific disorder (Disorder of Androgen Synthesis (DAS), Disorder of Androgen Action (DAA), Disorder of Gonadal Development (DGD)) was associated with complication development. EMS was also not significantly correlated with number of complications (p=0.8).

Of the 64 boys with ≥1 additional extragenital malformation, 46 (72%) presented with at least one postsurgical complication. There was no association between development of complications and particular extragenital malformations. In addition, the number of extragenital malformations had no association with the number of complications.

Complications occurred in 4 out of 7 (57%), 82 out of 119 (69%) and 16 out of 33 (48%) of those receiving their first surgery at <12 months, 12–24 months and >24 months, respectively. Of the 102 boys that received a staged operation, 69 (68%) experienced at least one postsurgical complications. Of the 58 boys who underwent a single operation, 33 (57%) experienced at least one postsurgical complication. Multivariate logistic regression also did not demonstrate any significant predictors of complication development (table 4).

Complication rates between centers varied from 50% to 100%. The highest complication rates were generally associated with the centers, which provided fewest boys. To establish trends between complication rates and income, centers were categorized by their

Table 1 Characteristics of study cohort

| Variables, n (%) | Whole cohort (n=160) |
|---|---|
| Associated genital malformations | |
| Yes | 160 (100) |
| No | 0 (0) |
| Additional extragenital malformations | |
| Yes | 63 (39) |
| No | 97 (61) |
| Age at first operation | |
| <12 months | 7 (4) |
| 12–24 months | 119 (74) |
| >24 months | 33 (21) |
| Unknown | 1 (1) |
| Type of procedure | |
| Single stage repair | 58 (36) |
| Staged repair | 102 (64) |
| Reoperation | |
| Yes | 75 (47) |
| No | 85 (53) |
| Genetic evaluation | |
| Yes | 135 (84) |
| No | 20 (13) |
| Unknown | 5 (3) |
| Endocrine evaluation | |
| Yes | 142 (88) |
| Basal testosterone | 136 (96) |
| Basal LH/FSH | 104 (73) |
| Testosterone post hCG stimulation | 87 (61) |
| AMH | 81 (57) |
| LHRH test | 55 (39) |
| Inhibin B | 10 (7) |
| No | 9 (6) |
| Unknown | 9 (6) |
| Family history | |
| Yes | 22 (14) |
| No | 138 (86) |
| Disorder type | |
| Non-specific XY DSD | 101 (63) |
| Disorders of androgen synthesis | 30 (19) |
| 5α-RD2 deficiency | 12 (40) |
| 3β-HSD deficiency | 6 (20) |
| 17β HSD3 deficiency | 5 (17) |
| POR deficiency | 4 (13) |
| 17α-OHD deficiency | 2 (7) |
| 17,20 lyase deficiency | 1 (3) |
| Disorders of androgen action | 22 (12) |
| PAIS | 22 (100) |
| Disorders of gonadal development | 9 (6) |
| PGD | 9 (100) |

AMH, anti-Müllerian hormone; DSD, disorders of sex development; FSH, follicle stimulating hormone; hCG, human chorionic gonadotrophin; LH, luteinizing hormone; LHRH, luteinizing hormone releasing hormone; PAIS, partial androgen insensitivity syndrome; PDG, partial gonadal dysgenesis; POR deficiency, cytochrome P450 oxidoreductase deficiency; 17α-OHD deficiency, 17-alphahydroxylase deficiency; 5α-RD2 deficiency, 5-alpha-reductase type 2 deficiency; 3β-HSD deficiency, 3-beta-hydroxysteroid dehydrogenase deficiency; 17β HSD3 deficiency, 17-beta-hydroxysteroid dehydrogenase type 3 deficiency.

country's income status (high, upper middle, lower middle or low income) according to the World Bank classifications.[12] Eight (62%) centers were categorized as high income, while five (38%) centers were categorized as upper-middle income. No centers that participated in the study were from lower middle-income or low-income counties. Complication rates between high and upper middle-income countries were similar at 39% and 38%, respectively. Of the 13 centers that provided eligible data, 8 (62%) are part of Europe, 3 (23%) are part of South America and 2 (15%) are part of Asia/transcontinental.

## DISCUSSION

The complication rate of 64% observed in this cohort is high and exceeds that of 56% previously reported in boys with proximal hypospadias.[2] Prior to the former study by Long *et al*, lower complication rates for proximal repairs were often reported, ranging from 13% to 23%.[13 14] As nearly half of complications seen in this cohort arose 2years after first hypospadias repair, it is possible that past studies lacked adequate follow-up periods, partially accounting for the comparatively high rates seen in the current study. Long-term prospective studies regarding complications in hypospadias are required to truly determine complication rates over time.

As previously described, additional genital malformations may be associated with poor surgical outcomes, likely secondary to smaller glans, less penile skin, greater degrees of chordee, and poor urethral plate quality.[8] In this study, 87% of boys presented with chordee, which has been associated with a 55% increase in complication rates.[15] Additionally, as 63% of the boys presented with micropenis, tissue availability and glans quality would likely be suboptimal in many cases, requiring the utilization of more complex surgical methods during hypospadias correction. Indeed, as the data for this study were

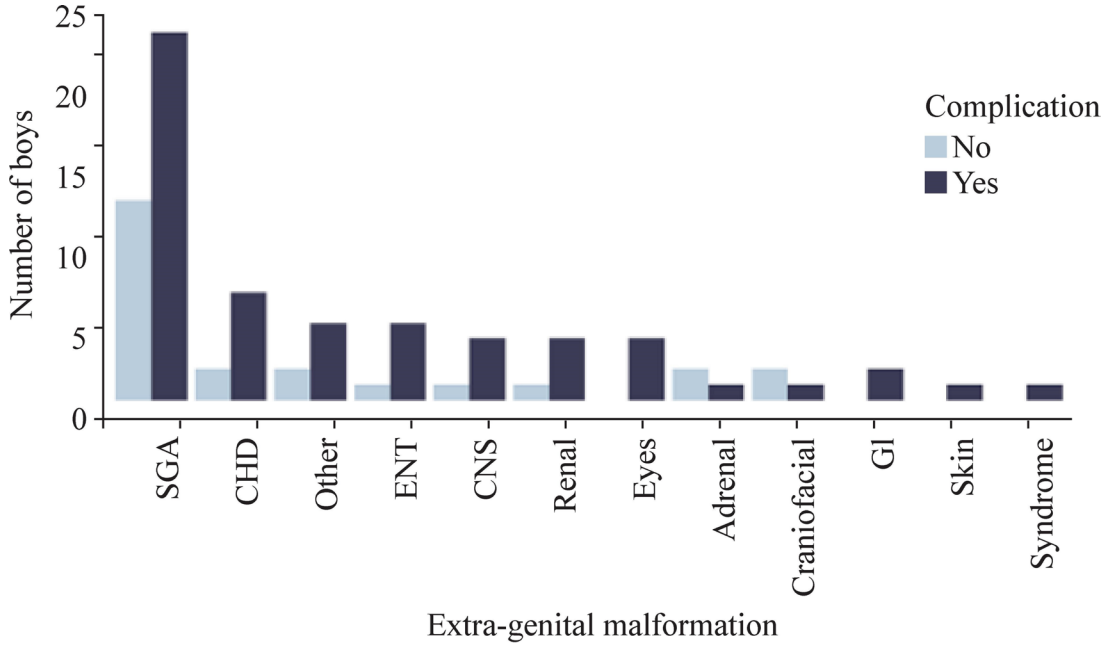


**Figure 2** Frequency of complication according to additional extragenital malformation. CHD, congenital heart defect; CNS, central nervous system; ENT, ear nose and throat; GI, gastrointestinal; MS, musculoskeletal; SGA, small for gestational age.

obtained from the I-DSD Registry, inherently, this cohort may be at a greater risk of complications due to the suboptimal anatomy associated with severe undermasculinization and DSD. Due to the retrospective nature of this study, however, it was not possible to collect detailed information on the quality of the urethral plate, angle of penile curvature or glans width. We would recommend that these should be introduced as aspects of standard data collection during hypospadias repairs. In addition, more detailed standard data collection regarding the precise type of surgical procedure would be useful for any future studies into complication rates.

**Table 2** Genetic variants identified in the cohort

| Gene, n (%) | Number of variants in cohort (n=42) |
|---|---|
| AR | 8(20) |
| NR5A1 | 7(18) |
| SRD5A2 | 7 (17) |
| HSD17B3 | 5(12) |
| HSD3B2 | 4(10) |
| *CHD7* | 2 (5) |
| *DHCR7* | 1 (2) |
| *DMD* | 1 (2) |
| *FGF8* | 1 (2) |
| *HSD17B* | 1 (2) |
| *MAMLD1* | 1 (2) |
| *PROK2* | 1 (2) |
| *RLIMB1* | 1 (2) |
| *RPGR* | 1 (2) |
| *WDR11* | 1 (2) |

Within this study cohort, median age at first surgery was older than the 6–18 months recommended by the American Academy of Paediatrics (AAP) and the European Association of Urology (EAU).[16 17] This may be due to characteristics associated with DSD postponing hypospadias surgery. For example, malformations such as undescended testes may be corrected first (via orchidopexy) due to the risk of malignancy or functional impairment. Furthermore, due to high rates of SGA and micropenis, it is possible that some hypospadias surgeries were postponed in order to permit penile growth and reduce anesthetic risk.

SGA was the most prevalent extragenital malformation in this cohort and was seen in 23% of all boys, which may therefore contribute to the high rates of complications observed. A recent study has reported higher hypospadias reoperation rates in boys with SGA.[18] That said, in our cohort, SGA alone was not sufficient to predict postsurgical complication rates as a single risk factor. This again confirms that several mechanisms may lead to complications.

No single clinical factor or combination of risk factors was significantly associated with postsurgical complication rates in this cohort. It was previously reported that boys with hypospadias and an identified AR variant were more likely to require additional operations than those with no identified AR variant.[10] In our study, however, while there was a high complication rate, the presence of additional extragenital malformations, or a genetic diagnosis were not associated with risk of complication. Previous studies have excluded patients with genetic variants in their analysis, which may explain differences in reported complication rates.[18] It is plausible that the high rates of extragenital malformations within

Table 3 Odds of complication according to risk factor

| Variables | n (%) | OR (95% CI) |
|---|---|---|
| Associated genital malformation | | |
| No | 0 (0) | NA |
| Yes | 160 (100) | NA |
| Extragenital malformation | | |
| No | 96 (60) | 1 |
| Yes | 64 (40) | 1.686 (0.867, 3.349) |
| Endocrine evaluation | | |
| Normal | 62 (44) | 1 |
| Abnormal | 80 (56) | 0.869 (0.434, 1.726) |
| Genetic evaluation | | |
| No variant found | 93 (69) | 1 |
| Variant found | 42 (31) | 0.528 (0.255, 1.090) |
| Disorder | | |
| Non-specific 46,XY DSD | 65/101 (64) | 1.01 (0.517, 1.967) |
| Known disorder | 37/59 (62) | 1 |
| Disorder of androgen synthesis | 16/28 (57) | |
| Disorder of androgen action | 14/22 (64) | |
| Disorder of gonadal development | 7/9 (78) | |
| Operation | | |
| Staged | 102 (64) | 1 |
| Single stage | 58 (36) | 0.631 (0.324, 1.230) |
| EMS | | 1.780 (0.750, 4.332) |
| Age at first surgery | | 0.778 (0.587, 1.010) |

CI, confidence interval; DSD, disorders of sex development; EMS, External Masculinization Score; OR, odds ratio.

the current cohort prevented an association being observed and that discrepancies in the results obtained from this study may be due to variations in genetic and biochemical testing internationally. It is possible that severe forms of hypospadias, such as those which may be reported by specialists in the care of DSD, may be part of a more complex syndrome that affects systems out-with the urogenital system, highlighting the need for multidisciplinary care in hypospadias and other DSD conditions.[19]

Table 4 Multivariate logistic regression for presence of complications

| | Effect | 95% CI | P value |
|---|---|---|---|
| Other genital anomalies | -0.2 | -0.6 to 0.1 | 0.2 |
| EMS | -0.09 | -0.2 to 0.05 | 0.2 |
| SGA | -0.05 | -1.1 to 1.7 | 0.9 |
| Age at first operation (years) | -0.4 | -0.7 to 0.1 | 0.06 |
| Genetic diagnosis | -0.1 | -0.3 to 0.5 | 0.6 |
| Staged procedure | -0.4 | -0.9 to 0.25 | 0.3 |

CI, confidence interval; EMS, External Masculinization Score; op, operation; SGA, small for gestational age.

This study has several strengths. It is one of the largest of its type, offering valuable insight into the characteristics and complication risks of a rare group of individuals with hypospadias. Additionally, the use of international data allows the results gathered to be applied on a global basis, demonstrating the benefits of using networks such as the I-DSD Registry. Due to the rarity and heterogeneity of these conditions, data collected from single centers are typically limited and only applicable to a specific region. This study could therefore offer a guide for the creation of international management protocols. One limitation of this study is the variations in genetic and biochemical testing observed between clinics. Similarly, clinical definitions will differ internationally, for example, in how SGA is classified. Definition and classification variations may therefore alter result interpretation.

In conclusion, boys with proximal hypospadias in the I-DSD Registry have high rates of additional comorbidities and a high risk of postoperative complications. No clinical factors alone or in combination were significantly associated with complication rates. High

complication rates with no observable cause suggest the involvement of other factors which need investigation. Further studies are needed to identify novel clinical factors which may be associated with complication rates in this complex group.

**Author affiliations**
[1]University of Glasgow, Glasgow, UK
[2]IRCCS Azienda Ospedaliero-Universitaria di Bologna Policlinico S Orsola, Bologna, Italy
[3]Great North Children's Hospital, Newcastle Upon Tyne, UK
[4]Hospital de Niños Ricardo Gutiérrez, Buenos Aires, Argentina
[5]Hospital de Clinicas de Porto Alegre, Porto Alegre, Brazil
[6]Istanbul University, Istanbul, Turkey
[7]Southampton Children's Hospital, Southampton, UK
[8]University Hospital of Geneva, Geneva, Switzerland
[9]Ukrainian Research Center of Endocrine Surgery Endocrine Organs and Tissue Transplantation, Kyiv, Ukraine
[10]State University of Campinas, Campinas, Brazil
[11]Marmara University, Istanbul, Turkey
[12]University Medical Centre, Ulm, Germany
[13]University Hospital of Schleswig-Holstein Campus Kiel/Christian-Albrechts-University of Kiel, Kiel, Germany
[14]Marmara University Faculty of Medicine, Istanbul, Turkey
[15]Yerevan State Medical University Endocrinology Clinic, Yerevan, Armenia
[16]Institut Pasteur, Paris, France
[17]Karolinska University Hospital, Stockholm, Sweden
[18]Royal Hospital for Children, Glasgow, UK
[19]IRCCS Ospedale San Raffaele, Milano, Italy
[20]Graubunden Cantonal Hospital, Chur, Switzerland

**Twitter** Angela K Lucas-Herald @lucas_herald

**Acknowledgements** AKL-H is funded by an NES/CSO Clinical Lectureship. AKL-H is supported by the Joint NES/CSO Postdoctoral Clinical Lectureship Scheme. SFA and JB are supported by the European Union's Health Programme (2014–2020) on the EuRRECa project '777215/EuRRECa' and SFA is also supported by the European Union's Health Programme (2014–2020) on the EuRR-Bone project '946831/EuRR-Bone. The I-DSD Registry was developed using support from the Medical Research Council (G1100236), the Seventh European Union Framework Program (201444), the European Society for Paediatric Endocrinology Research Unit and is maintained through fees applied for data access and through its biennial symposiums.

**Contributors** AKL-H and SFA designed the study and supervised the work of KS. AKL-H is the guarantor for the work. JB, FB, KS, RLB, JRC, SC, TC, ECC, FD, JHD, MD, GG, EG, GGJ, TG, GH, PMH, AKA, RM, KME, MLM, AN, SOT, SP, GR, VS, MRS, MS and SFA were involved in data acquisition. KS conducted data analysis and interpretation. KS and AKL-H wrote the drafts of the manuscript. All authors contributed to writing and/or reviewing the manuscript and approved the final version.

**Funding** This study was funded by Chief Scientist Office (PCL/21/05).

**Competing interests** None declared.

**Patient consent for publication** Not applicable.

**Ethics approval** Ethics approval was granted by the I-DSD Registry Data Access Committee on the grounds that data sharing between organizations would adhere to the principles of the UK Data Protection Act (2018), the European Union General Data Protection Regulations (2018) and the 'Conditions of Ethical Approval' as stipulated by the West of Scotland Research Ethics Service (project ID 269776) on 10/01/19. Participants gave informed consent to participate in the study before taking part.

**Provenance and peer review** Not commissioned; externally peer reviewed.

**Data availability statement** Data are available upon reasonable request.



**ORCID iD**
Angela K Lucas-Herald http://orcid.org/0000-0003-2662-1684

## REFERENCES

1 Ahmed SF, Dobbie R, Finlayson AR, *et al*. Prevalence of hypospadias and other genital anomalies among singleton births, 1988-1997, in Scotland. *Arch Dis Child Fetal Neonatal Ed* 2004;89:F149–51.
2 Long CJ, Chu DI, Tenney RW, *et al*. Intermediate-term followup of proximal hypospadias repair reveals high complication rate. *J Urol* 2017;197:852–8.
3 Wu Y, Wang J, Zhao T, *et al*. Complications following primary repair of non-proximal hypospadias in children: a systematic review and meta-analysis. *Front Pediatr* 2020;8:579364.
4 van der Horst HJR, de Wall LL. Erratum to: hypospadias, all there is to know. *Eur J Pediatr* 2017;176:1443.
5 Keays MA, Dave S. Current hypospadias management: diagnosis, surgical management, and long-term patient-centred outcomes. *Can Urol Assoc J* 2017;11:S48–53.
6 Hughes IA, Houk C, Ahmed SF, *et al*. Consensus statement on management of intersex disorders. *Arch Dis Child* 2006;91:554–63.
7 Mendonca BB, Domenice S, Arnhold IJP, *et al*. 46,XY disorders of sex development (DSD). *Clin Endocrinol (Oxf)* 2009;70:173–87.
8 Al-Juraibah F, Lucas-Herald A, Nixon R, *et al*. Association between extra-genital congenital anomalies and hypospadias outcome. *Sex Dev* 2019;13:67–73.
9 Dokter EM, van der Zanden LF, Laumer SJ, *et al*. Development of a prediction model for postoperative complications after primary Hypospadias correction. *J Pediatr Surg* 2020;55:2209–15.
10 Lucas-Herald A, Bertelloni S, Juul A, *et al*. The long-term outcome of boys with partial androgen insensitivity syndrome and a mutation in the androgen receptor gene. *J Clin Endocrinol Metab* 2016;101:3959–67.
11 Ahmed SF, Khwaja O, Hughes IA. The role of a clinical score in the assessment of ambiguous genitalia. *BJU Int* 2000;85:120–4.
12 The World Bank. The world by income and region; 2021. Available: https://datatopics.worldbank.org/world-development-indicators/the-world-by-income-and-region.html
13 Snodgrass W, Bush N. Tubularized Incised plate proximal hypospadias repair: continued evolution and extended applications. *J Pediatr Urol* 2011;7:2–9.
14 Castagnetti M, Zhapa E, Rigamonti W. Primary severe hypospadias: comparison of reoperation rates and parental perception of urinary symptoms and cosmetic outcomes among 4 repairs. *J Urol* 2013;189:1508–13.
15 Mohammed M, Bright F, Mteta A, *et al*. Long-term complications of Hypospadias repair: a ten-year experience from northern zone of Tanzania. *Res Rep Urol* 2020;12:463–9.
16 Kass E, Kogan SJ, Manley C, *et al*. Timing of elective surgery on the genitalia of male children with particular reference to the risks, benefits, and psychological effects of surgery and anesthesia. *Pediatrics* 1996;97:590–4.
17 Riedmiller H, Androulakakis P, Beurton D, *et al*. EAU guidelines on paediatric urology. *Eur Urol* 2001;40:589–99.
18 Haid B, Tack LJW, Spinoit A-F, *et al*. Being born small for gestational age (SGA) might be associated with a higher reoperation rate in proximal hypospadias. *J Pediatr Urol* 2022;18:609.
19 Ahmed SF, Achermann J, Alderson J, *et al*. Society for endocrinology UK guidance on the initial evaluation of a suspected difference or disorder of sex development. *Clin Endocrinol (Oxf)* 2021;95:818–40.