ACT Government
Chief Minister, Treasury and Economic Development

Freedom of Information Publication Coversheet

The following information is provided pursuant to section 28 of the *Freedom of Information Act 2016*.

FOI Reference: CMTEDDFOI 2023-110

Information to be published	Status
1. Access application	Published
2. Decision notice	Published
3. Documents and schedule	Published
4. Additional information identified	No
5. Fees	N/A
6. Processing time (in working days)	7
7. Decision made by Ombudsman	N/A
8. Additional information identified by Ombudsman	N/A
9. Decision made by ACAT	N/A
10. Additional information identified by ACAT	N/A

From: no-reply@act.gov.au
To: CMTEDD FOI
Subject: Freedom of Information request
Date: Wednesday, 22 March 2023 8:25:10 PM

Please find online enquiry details below. Please ensure this enquiry is responded to within fourteen working days.

Your details

All fields are optional, however an email address OR full postal address must be provided for us to process your request. An email address and telephone contact number will assist us to contact you quickly if we need to discuss your request.

Title:
First Name:
Last Name:
Business/Organisation:
Address:
Suburb:
Postcode:
State/Territory:
Phone/mobile:
Email address:

Request for information

(Please provide as much detail as possible, for example subject matter and relevant dates, and also provide details of documents that you are not interested in.)

Under the Freedom of Information Act 2016 I want to access the following document/s (*required field):

This request is in regard to the Variation in Sex Characteristics (Restricted Medical Treatment) Bill 2023 and we want the documents that provide answers to the below questions. - How the consultation process occurred, to include hypospadias in the draft bill. This includes what consultation occurred with stakeholders (including those with hypospadias) and who was consulted as part of the process? - How the decision to exclude hypospadias occurred after the draft consultation process had finished, and how the decision to still include proximal hypospadias with undescended testicles was decided. What peer reviewed evidence supports this decision? And if there was limited evidence to support this decision then how and why was this decision made, and who by? - Were parents with children with proximal hypospadias with undecided testes targeted as part of the decision making process that occurred after the public consultation on the draft report? If not, why not? - Did the ACT government reach out to the public who had provided feedback as part of the initial consultation for the bill (specifically hypospadias), and if so how did this occur? If not, why not? - Have parents who have children diagnosed with these conditions been notified by the ACT

	government of the Bill or will they have to find this information out for themselves? - What support is being provided to parents as the bill is enacted over the next 12 months?
I do not want to access the following documents in relation to my request::	N/A

Thank you.
Freedom of Information Coordinator

ACT Government
Chief Minister, Treasury and Economic Development

Our ref: CMTEDDFOI2023-110

FREEDOM OF INFORMATION REQUEST

I refer to your application under section 30 of the *Freedom of Information Act 2016* (the Act), received by the Chief Minister, Treasury and Economic Development Directorate (CMTEDD) on 22 March 2023 in which you sough access to information relating to information relating to the ***"Variation in Sex Characteristics (Restricted Medical Treatment) Bill 2023".***

Specifically, you sought access to the documents that provide answers to the below questions:

1. *How the consultation process occurred, to include hypospadias in the draft bill. This includes what consultation occurred with stakeholders (including those with hypospadias).*
2. *How the decision to exclude hypospadias occurred after the draft consultation process had finished, and how the decision to still include proximal hypospadias with undescended testicles was decided. What peer reviewed evidence supports this decision? And if there was limited evidence to support this decision then how and why was this decision made, and who by?*
3. *Were parents with children with proximal hypospadias with undecided testes targeted as part of the decision making process that occurred after the public consultation on the draft report? If not, why not?*
4. *Did the ACT government reach out to the public who had provided feedback as part of the initial consultation for the bill (specifically hypospadias), and if so how did this occur? If not, why not?*
5. *Have parents who have children diagnosed with these conditions been notified by the ACT government of the Bill or will they have to find this information out for themselves?*
6. *What support is being provided to parents as the bill is enacted over the next 12 months?*

On 3 April 2023 you had a conversation with me and confirmed that you would be amenable to receiving the information in the form of a letter rather than a series of documents.

On 12 April 2023, you clarified the scope of your request, amending question 1 to exclude who was consulted as part of the consultation process.

Authority

I am an Information Officer appointed by the Director-General under section 18 of the Act to deal with access applications made under Part 5 of the Act.

Timeframes

In accordance with section 40 of the Act, CMTEDD was required to provide a decision on your access application by 21 April 2023.

Decision on access

Searches were completed for relevant information, and the information contained in **Attachment B** falls within the scope of your request. I have decided to grant full access to this information and provided a schedule of the relevant documents as **Attachment A** outlining my access decisions.

In accordance with section 54(2) of the Act a statement of reasons outlining my decisions is below.

Statement of Reasons

As a decision maker, I am required to determine whether the information within scope is in the public interest to release. To make this decision, I am required to:

- assess whether the information would be contrary to public interest to disclose as per Schedule 1 of the Act
- perform the public interest test as set out in section 17 of the Act by balancing the factors favouring disclosure and factors favouring non-disclosure in Schedule 2

The public interest information **under schedule 2 of the Act**

The Act has a presumption in favour of disclosure. As a decision maker I am required to decide where, on balance, public interest lies. As part of this process, I must consider factors favouring disclosure and non-disclosure.

Taking into consideration the information found to be within the scope of your request, I have identified that the following public interest factors are relevant to determine if release of the information contained within these documents is within the 'public interest'.

Factors **favouring disclosure** in the public interest:

(a) disclosure of the information could reasonably be expected to do any of the following:

(i) promote open discussion of public affairs and enhance the government's accountability.

(ii) contribute to positive and informed debate on important issues or matters of public interest.

(viii) reveal the reason for a government decision and any background or contextual information that informed the decision.

I have put substantial weight on the above factors favouring disclosure. The release of this information can reasonably be expected to provide information that will inform the community and inform debate on important issues or matters of interest.

I did not identify any factor favouring nondisclosure and have decided to release this information to you in full.

Charges

Processing charges are not applicable for this request because the number of pages released to you is below the charging threshold of 50.

Online publishing – Disclosure Log

Under section 28 of the Act, CMTEDD maintains an online record of access applications called a disclosure log. Your original access application and my decision will be published on the CMTEDD disclosure log. Your personal contact details will not be published. You may view CMTEDD disclosure log at https://www.cmtedd.act.gov.au/functions/foi/disclosure-log-2023

Ombudsman Review

My decision on your access request is a reviewable decision as identified in Schedule 3 of the Act. You have the right to seek Ombudsman review of this outcome under section 73 of the Act within 20 working days from the day that my decision is published in CMTEDD disclosure log, or a longer period allowed by the Ombudsman.

We recommend using this form *Applying for an Ombudsman Review* to ensure you provide all of the required information. Alternatively, you may write to the Ombudsman:

The ACT Ombudsman
GPO Box 442
CANBERRA ACT 2601

Via email: actfoi@ombudsman.gov.au

ACT Civil and Administrative Tribunal (ACAT) Review

Under section 84 of the Act, if a decision is made under section 82(1) on an Ombudsman review, you may apply to the ACAT for review of the Ombudsman decision. Further information may be obtained from the ACAT at:

ACT Civil and Administrative Tribunal
Level 4, 1 Moore St
GPO Box 370
Canberra City ACT 2601
Telephone: (02) 6207 1740
http://www.acat.act.gov.au/

Should you have any queries in relation to your request please contact me by telephone on 6207 7754 or email CMTEDDFOI@act.gov.au.

Yours sincerely,

Katharine Stuart
Information Officer
Chief Minister, Treasury and Economic Development Directorate

12 April 2023

ACT Government
Chief Minister, Treasury and Economic Development

FREEDOM OF INFORMATION REQUEST SCHEDULE

WHAT ARE THE PARAMETERS OF THE REQUEST	Reference NO.
"Variation in Sex Characteristics (Restricted Medical Treatment) Bill 2023" 1. *How the consultation process occurred, to include hypospadias in the draft bill. This includes what consultation occurred with stakeholders (including those with hypospadias).* 2. *How the decision to exclude hypospadias occurred after the draft consultation process had finished, and how the decision to still include proximal hypospadias with undescended testicles was decided. What peer reviewed evidence supports this decision? And if there was limited evidence to support this decision then how and why was this decision made, and who by?* 3. *Were parents with children with proximal hypospadias with undecided testes targeted as part of the decision making process that occurred after the public consultation on the draft report? If not, why not?* 4. *Did the ACT government reach out to the public who had provided feedback as part of the initial consultation for the bill (specifically hypospadias), and if so how did this occur? If not, why not?* 5. *Have parents who have children diagnosed with these conditions been notified by the ACT government of the Bill or will they have to find this information out for themselves?* 6. *What support is being provided to parents as the bill is enacted over the next 12 months?*	**CMTEDDFOI 2023-110**

Ref No	Page number	Description	Date	Status	Reason for Exemption	Online Release Status
1	1-8	Letter response from		Full		Yes
2	9-23	Attachment A - Internal Brief on Hypospadias for external consultation 24 Aug 2021		Full		Yes
3	24-27	Attachment B - Defining Hypospadias in Prescribed List		Full		Yes

Total No of Docs
3

ACT Government
Chief Minister, Treasury and Economic Development

Our ref: CM2023/1729

[redacted]

Questions regarding the Variation in Sex Characteristics (Restricted Medical Treatment) Bill 2023

I write regarding your Freedom of Information (FOI) request submitted to the Directorate on 30 March 2023 which included a number of questions about development of the *Variations in Sex Characteristics (Restricted Medical Treatment) Bill 2023* (the Bill).

Given the nature of the questions you are seeking to have answered, I understand you have agreed to the Directorate initially providing written answers directly addressing your specific questions. Information regarding each of your separate questions has been provided below plus two attachments to this letter.

1. **How the consultation process occurred, to include hypospadias in the draft bill. This includes what consultation occurred with stakeholders (including those with hypospadias) and who was consulted as part of the process?**

From commencement in 2019, the reform process has had in scope people born with sex characteristics (such as genitals, gonads or chromosome patterns) that do not fit typical binary notions of male or female bodies. More information about the formal definition used in the Bill is included in the response to question 2 below. Hypospadias, like all other variations, has always been within this scope, as it meets this general definition.

The focus of these reforms has been on how to provide protections to all people with variations, though in the course of those consultations we heard from some stakeholders who wanted certain variations included or excluded.

The overarching consultation process for the project on deferrable medical interventions on people with variations in sex characteristics is documented in the listening reports from the three phases of public consultations, available on the Office of LGBTIQ+ Affairs (the Office) website.[1] This included:

- A December 2020 Discussion Paper on key issues in the prohibition of deferrable medical interventions on intersex people.

[1] https://www.cmtedd.act.gov.au/policystrategic/the-office-of-lgbtiq-affairs/variations-in-sex-characteristics-bill

- An April 2021 legal issues workshop, participated in by expert lawyers from across Australia.
- A June 2021 Options Paper released for public comment.
- An Exposure Draft of the Bill and Regulation released for public comment from May to July 2022.

At all stages, consultation was with stakeholders who work across or represent the range of variations in sex characteristics, which include hypospadias.

During consultation on the draft Bill in May-July 2022, a survey was set up that could be completed by members of the community. The Directorate received 14 survey responses (nine who stated they were from the ACT, five from other states) from people who said they were parents of children with hypospadias. Some of those responses talked specifically about views regarding that particular variation in sex characteristics. Most of these responses expressed concern about limitations being placed on the role of parents in providing care to their child. Most indicated they did not consider hypospadias to be a variation in sex characteristics, or should not be covered by the legislation. This is discussed under the response to question 2 below.

The Directorate did not ask individuals or organisational representatives who provided input to specify what variation in sex characteristics that they or their child may have. The Directorate only had information about a person's variation in the minority of cases where a person chose to disclose this information.

2. **How the decision to exclude hypospadias occurred after the draft consultation process had finished, and how the decision to still include proximal hypospadias with undescended testicles was decided. What peer reviewed evidence supports this decision? And if there was limited evidence to support this decision then how and why was this decision made, and who by?**

The objective of these reforms, including the Bill, is to provide support to each person who has a variation in sex characteristics and their family. Excluding a variation potentially reduces the protections for those people and could expose them to risks of treatment that has been inadequately considered for the range of risks it might present. Accordingly, the Bill extends protection to everyone[2] who has a variation, and then creates a mechanism for a variation to be exempted from requiring treatment plan approval, where there is a reason to do so.

[2] Everyone who lacks capacity to provide their own consent to the treatment being proposed: see Bill section 9.

The mechanism that allows this to occur is in the definition of variations in sex characteristics, which is defined in the Bill using commonly accepted language in medical and legal settings. That definition is in section 7 and says:

> (1) *variation in sex characteristics—*
>
> *(a) means a congenital condition that involves atypical sex characteristics; and*
>
> *(b) includes a condition prescribed by regulation; but*
>
> *(c) does not include a condition prescribed by regulation not to be a variation in sex characteristics.*
>
> *(2) In this section:*
>
> *sex characteristics—*
>
> *(a) means a person's chromosomal, gonadal or anatomical sex; and*
>
> *(b) includes—*
>
> *(i) the person's hormones that are related to sex; and*
>
> *(ii) the sexual and reproductive parts of the person's anatomy; and*
>
> *(iii) the person's secondary physical features emerging as a result of puberty.*

Subsection 1(c) is the mechanism that can allow individual bodily variations to be explicitly excluded from scope through the making of a regulation.[3]

Given the Directorate heard from some parents who did not want the Bill to apply to hypospadias, it had to consider those views, alongside the views of other stakeholders, published evidence and other reviews in the field. The goal remained how best to support people with hypospadias and their families.

Among the comments received from parents, many of the supporting reasons provided in those comments were based on one or more incorrect beliefs about the reform:

- An assumption that if a variation was not associated with ambiguity in sex of rearing, it was not relevant to the reform (whereas the reform's aim was always broader than this and concerns normalising medical treatments affecting the sex characteristics, performed without personal consent);
- A misunderstanding of the extent to which the Bill would limit parents' roles (which has been clarified in the revised Bill and Explanatory Statement);
- An assumption there would be no additional services available to families (whereas the 2022-23 ACT Budget funded a new psychosocial care unit that is currently being set up); and/or
- A belief that the Bill would prevent treatments to address health needs such as penile pain or urinary tract infection (whereas it will not).

[3] Note that a variation does not need to be listed in a regulation under subsection 1(b) to be covered – that subsection is solely for offering clarity to those relying on the law. This is different to how the draft Bill released for community consultation worked.

Peer reviewed evidence has been relied upon throughout the project, including in relation to hypospadias. Attachment A is a paper prepared to support project team discussion regarding hypospadias, which reviewed some of that literature as of mid-2021. Some of the peer reviewed evidence considered in the assessment of how to treat hypospadias within this reform are:

- The frequency of occurrence of hypospadias (around 1 in 150 male births),[4] and the fact that most of these are distal hypospadias[5];
- Advice received from health professionals during consultations, that appears consistent with most literature, that complications are more common in the more complex hypospadias cases, such as proximal hypospadias, than in distal hypospadias;
- High rates of complexity and complication for some hypospadias surgery,[6] which may indicate an area of treatment in which decision regret may be significant;
- A survey-based study of men who had had hypospadias surgery showing only slightly under half agreed it should be done in infancy;[7]
- Lack of evidence of psychosocial harm arising from delayed surgery;[8]
- Review of the medical literature that showed a distinction within health professional opinion and practice that proximal hypospadias with cryptorchidism can be considered a variation in sex characteristics and should be approached differently to distal hypospadias and/or hypospadias without cryptorchidism. This literature is summarised in Attachment B.

Synthesising this information, and considering all the available views, the Government's decision was to, in the draft regulation, exclude hypospadias other than proximal hypospadias with cryptorchidism.

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[4] Tee K, Croaker D and Rampersad R. 2020. "Increasing incidence of hypospadias in the Australian Capital Territory, 1987-2016", *Journal of Pediatric Urology* 16: S39.

[5] van der Horst HJR and de Wall LL. 2017. "Hypospadias, all there is to know" *European Journal of Paediatrics* 176(4): 435-441

[6] E.g. Nguyen, S., Durbin-Johnson, B. and Kurzrock, E.A., 2021. Reoperation after hypospadias repair: long-term analysis. *The Journal of urology*, 205(6), pp.1778-1784; Long CJ and Canning DA. 2016. "Hypospadias: Are we as good as we think when we correct proximal hypospadias?", *Journal of Pediatric Urology* 12: 196.e1-196.e5; Long CJ, Chu DI, Tenney RW, Morris AR, Weiss DA, Shuklas AR, Srinivasan AK, Zderic SA, Kolo TF, Canning DA. 2017. "Intermediate-Term Follow-up of Proximal Hypospadias Repair Reveals High Complication Rate" *Journal of Urology* 197: 852-858; Carmack A, Notini L, and Earp B. 2015. "Should Surgery for Hypospadias Be Performed Before an Age of Consent?" *Journal of Sex Research* 00: 1-12.

[7] Bennecke, E., Bernstein, S., Lee, P., van de Grift, T.C., Nordenskjöld, A., Rapp, M., Simmonds, M., Streuli, J.C., Thyen, U. and Wiesemann, C., 2021. Early genital surgery in disorders/differences of sex development: patients' perspectives. *Archives of Sexual Behavior*, 50(3), pp.913-923.

[8] Roen K, and Hegarty B. 2018. "Shaping parents, shaping penises: How medical teams frame parents' decisions in response to hypospadias" *British Journal of Health Psychology* 23(4): 967-981; Schonbucher VB, Weber DM, and Landolt M. 2008. "Psychosocial Adjustment, Health-Related Quality of Life, and Psychosexual Development of Boys with Hypospadias: A systematic review" *Journal of Pediatric Psychology* 33(5): 530, 534

The Government is going to continue to monitor treatment in this area, including through reporting mechanisms built into the new scheme. The Bill contains a requirement that the operation of the new scheme be reviewed after two years. It expects this to include consideration of whether the exclusion of some hypospadias has been consistent with the protections that the Bill is intended to provide.

3. **Were parents with children with proximal hypospadias with undecided testes targeted as part of the decision making process that occurred after the public consultation on the draft report? If not, why not?**

During meetings with health professionals at different points in the process, the Directorate verbally indicated that it would welcome additional input from people with different variations in sex characteristics. The Directorate explained that, beyond contacting support organisations, the main means by which people would be made aware of the project would be either through public website contact, or through health professionals telling their patient populations about the project.

The Directorate does not hold personal information about individuals with variations in sex characteristics, nor has access to confidential medical databases, that would allow it to target communication to people with a particular variation in sex characteristics, or their parents. It therefore could not directly target this subset of people. The Directorate relied primarily on the published literature, as discussed in more detail in response to question 2, above.

4. **Did the ACT government reach out to the public who had provided feedback as part of the initial consultation for the bill (specifically hypospadias), and if so how did this occur? If not, why not?**

We understand this question to refer to whether the Government sought subsequently to contact community members who had provided feedback during May-July 2022 in response to the draft Bill.

In August 2022, a listening report was released that summarised the input received about the Bill, which included detailed description of divergent stakeholder views regarding hypospadias.[9]

On 15 December 2022, stakeholder organisations and individuals who provided written submissions were sent an email inviting them to participate in a webinar that was held on 24 January 2023. The webinar was also advertised on the project website, and some stakeholder organisations promoted it through their networks. Participants were invited to provide written questions before or after that webinar.

[9] https://www.cmtedd.act.gov.au/policystrategic/the-office-of-lgbtiq-affairs/variations-in-sex-characteristics-bill

Those questions contributed to the content of the Frequently Asked Questions that are available on the project webpage.[10]

Due to an administrative error that came to light during preparation of these answers, that invitation did not get sent to anyone who was:

1. Not on the list of stakeholders the Directorate maintained during 2022; *and*
2. Contacted the Directorate during the Bill consultation process and provided verbal input but did not provide an emailed submission.

We believe this would have affected one family of a child with a variation in sex characteristics. Staff in the Directorate were upset to find the oversight and will offer an apology to anyone affected by it.

The online survey administered as part of the consultation process was designed to be anonymous to afford privacy to respondents. As a result the Directorate does not know who the respondents are, and had no mechanism to contact those participants to invite them to the webinar. The Directorate provided information to ACT health professionals, including surgical doctors involved in the provision of care for hypospadias within the ACT, about the process.

Though the survey did not ask respondents to indicate how they were made aware of the survey, comments some respondents provided suggest some were made aware by their ACT surgical specialist. This is consistent with the Directorate's request to health professionals that they encourage patients to engage with the project.

As set out in more detail in response to question 2, the main basis for the decision of how to define the hypospadias exclusion was a weighing up of published literature and opinions provided during the different consultation stages.

5. Have parents who have children diagnosed with these conditions been notified by the ACT government of the Bill or will they have to find this information out for themselves?

The Bill is currently under consideration by the Legislative Assembly and has not yet been debated or passed. Assuming it is debated and passed, the Bill, together with a Regulation, will be publicly notified.

The Bill as currently drafted will not take effect until six months after it is passed. The provisions in the Bill, requiring treatment plans to be in place for restricted medical treatments on prescribed people who have variations in sex characteristics, will not become mandatory for an additional twelve months. We are still therefore some time away from the Bill's protections taking effect.

[10] Ibid

It is not expected that families will have to find out for themselves about the new scheme, including the legislation, the expanded peer support and the psychosocial care unit. During the period before the Bill takes full effect, the Government will be organising and delivering training for health care professionals, establishing the statutory authority that implements the legislation, and delivering information to raise community awareness of the changes.

Health professionals will be responsible for compliance with the law, where they are proposing a restricted medical treatment for a child under the law. As such, we expect them to make parents who have children diagnosed with variations covered by the Bill aware of the reform and its requirements.

We expect that all health professionals working with children with variations in sex characteristics and their families will be providing referrals to peer support services, and we expect that peer support organisations will be reaching out to communities, which will contribute to increased awareness of the supports available for, and the needs of, people with variations.

6. What support is being provided to parents as the bill is enacted over the next 12 months?

The implementation timeline for the Bill is expected to be 18 months, which does not commence until the Bill passes (see answer to question 5, above). We expect that treating health professionals will continue to provide support and care for patients with variations in sex characteristics and their families. This includes making families aware of the reform and how new treatment pathways will operate, and continuing to provide care during the transition period.

Canberra Health Services is currently establishing the Variations in Sex Characteristics Psychosocial Support Unit to support the wholistic care of people with variations in sex characteristics and their families. Psychosocial professionals will assist children and/or their families to understand and emotionally process a child or young person's intersex variation and the impact on their life, health and wellbeing.

These psychosocial professionals will become a mandatory part of care once the legislation comes into effect, and will ensure that patients and their families have the information and supports available to them to assist with informed decision-making processes regarding available treatments (including any deferral of treatment options) for the patient's particular variation.

In the 2022-23 Budget, the Government also committed additional funding to bolster peer support services offered by non-government community partners, for people born with variations in sex characteristics. This is expected to include overarching peer support organisations, as well as variation specific organisations where they exist.

Referral to peer supports is expected to occur through the Psychosocial Support Unit, as well as through treating practitioners once training is complete. This will be a vital part of

supports provided as it will connect patients and families to people with lived experience, providing a broader range of perspectives, as well as an accepting environment. These organisations already exist and can accept referrals made during the transition period to the Bill's commencement, with the additional funding commitment coming into effect from the 2023-24 Financial Year (i.e. 1 July 2023).

Thank you for raising these questions with the Directorate and your consideration of the responses provided. If you consider the information provided in this response does not answer your questions, we would be happy to provide further written responses about these reforms and the process through which they were developed, or to produce specific documents in line with FOI processes.

Yours sincerely

Andrew Mehrton

Andrew Mehrton
Executive Branch Manager, Social Policy Branch
Chief Minister, Treasury and Economic Development Directorate

12 April 2023

Hypospadias – evidence regarding rationales for surgery and timing – a brief review

1. This briefing note provides an overview of research undertaken by the Office of LGBTIQ+ Affairs about the issues involved in surgical interventions that respond to hypospadias, generally called hypospadias repair. It is intended to support discussion about these issues in the design of a regulatory framework for medical interventions for people with variations in sex characteristics.
2. Medical consensus largely views that surgical hypospadias repairs are best carried out in early childhood (prior to 18 months of age).[1] These are therefore patients who cannot provide input or provide personal informed consent[2] to any procedure. Given this, parents are asked to make a decision on their child's behalf regarding this surgery.
3. Because these interventions modify sex characteristics and occur without personal consent, there is debate about whether hypospadias and hypospadias repair should be included in the scope of legislation that safeguards the rights of people with variations in sex characteristics to consent to medical intervention on their sex characteristics.
4. The Victorian government has recently commissioned a discussion paper that proposes hypospadias be included in the scope of such legislation.[3] Medical groups, such as the Australasian Paediatric Endocrine Group, have argued in the past that outcome data of undertaking hypospadias repair surgery in early childhood is positive and that the medical consensus is that these surgeries should not be deferred.[4]
5. This brief arises from the need to better understand hypospadias repair and review the existing literature on outcomes of these procedures. Key questions explored in the brief:
    a. What is the evidence that surgery in early childhood produces better health outcomes than later surgery with personal consent?
    b. If surgery is deferred, is there evidence that deferral produces negative health outcomes?

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[1] van der Horst HJR and de Wall LL. 2017. "Hypospadias, all there is to know" *European Journal of Paediatrics* 176(4): 435-441.

[2] In this paper, "personal consent" is used to distinguish the consent of the individual from the consent of a parent or guardian.

[3] Equality Australia. 2021. A Victorian Intersex Oversight scheme: a consultation paper on a legal scheme to protect people from medical interventions on their sex characteristics without personal consent. https://equalityaustralia.org.au/wp-content/uploads/2021/07/Consultation-paper-on-intersex-reform-in-Victoria-final-1.pdf.

[4] Australasian Paediatric Endocrine Group. 2013. *Submission of the Australasian Paediatric Endocrine Group to the Senate Inquiry into the Involuntary or Coerced Sterilization of People with Disabilities in Australia: Regarding the Management of Children with Disorders of Sex Development.*

Hypospadias Overview

6. Hypospadias is a genital variation in which the urethra opening (meatus) forms on the underside of the penis, scrotum or, in a smaller number of cases the perineum, rather than on the tip of the penis glans.
7. Hypospadias occurs in approximately 1 in 200-300 male births,[5] and is typically detected at birth or early childhood, though more mild forms may not be identified or noticed by the individual.[6]
8. Individuals with hypospadias are assigned male at birth, and identity as male/men. No studies were identified that discussed gender dysphoria or transgender identification among hypospadias populations. In the absence of such research, it can reasonably be assumed that the prevalence of gender dysphoria or transgender identification in those with hypospadias is no different to that of the general population as a whole.
9. Hypospadias can have wide variation in presentation, with two primary forms described as distal, in which the urethra opening is located on or near the glans of the penis, and proximal, in which the urethra opening is on the penis shaft, scrotum or perineum.
10. Distal hypospadias is more prevalent, reported in a number of studies to make up 70% of all hypospadias occurrence.[7] The Office of LGBTIQ+ Affairs has heard there may be a higher prevalence, closer to 90-95%, seen in the ACT; however more severe presentations have slowly increased in Canberra.[8]
11. Distal hypospadias is mostly a cosmetic issue rather than a functional one,[9] and is often referred to as 'mild'.
12. Proximal hypospadias is more likely to be accompanied by other variations in genital development, such as excessive downward curvature of the penis (chordee), or underdeveloped foreskin.[10] In some hypospadias cases one or both testes may be undescended. Depending on the extent of these genital variations there may be issues related to normative function, such as the inability for the person to urinate standing upright.

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[5] 1 in 150 for the ACT: Tee K, Croaker D and Rampersad R. 2020. "Increasing incidence of hypospadias in the Australian Capital Territory, 1987-2016", *Journal of Pediatric Urology* 16: S39.
[6] Bouty A, Ayers KL, Pask A, Heloury Y, and Sinclair AH. 2015. "The Genetic and Environmental Factors Underlying Hypospadias" *Sexual Development* 9: 239-259.
[7] Bouty et al., 2015; van der Horst and de Wall, 2017.
[8] Tee et al. 2020.
[9] Vavilov S, Smith G, Starkey M, Pockney P, and Deshpande AV. 2020. "Parental decision regret in childhood hypospadias surgery: A systematic review" *Journal of Paediatrics and Child Health* 56 Issue 10: 1514-1520. https://doi.org/10.1111/jpc.15075.
[10] Carmack et al., 2015.

13. Hypospadias is controversially a variation in sex characteristics (also known as intersex variations). Hypospadias can co-occur with other intersex variations, but it commonly occurs in individuals who have no other identified variations in sex characteristics. Whether the presence of hypospadias alone should be considered as an intersex variation is contested, with the medical community generally not viewing it as an intersex variation while intersex communities and academic observers commenting that it should be.[11] It is included in the definition used in the Australian reference work *Disorders/Differences of Sex Development*,[12] which also makes the distinction between 'simple' hypospadias and hypospadias that is symptomatic of another variation in sex characteristics.[13]
14. One of the arguments for its inclusion is that the treatment is, in the majority of cases, predominantly cosmetic and not necessary in order to manage physical health. As a result, it has been proposed that surgical repair of hypospadias should be included in the scope of reforms that prevent surgery on sex characteristics without an individual's consent.[14]
15. A range of methods and techniques were discussed in the literature with the primary aims of surgery to relocate the meatus to the tip of the glans, sometimes accompanied with repair of the glans, foreskin reconstruction, correction of curvature and/or other procedures intended to make a hypospadic penis function like, and resemble, an unaffected penis.[15]
16. Depending on the method and techniques employed, hypospadias repair may be a single stage surgery, or it may be planned as a multi-stage procedure.[16]

<u>Reasons for hypospadias surgery</u>

17. The predominant reasons for hypospadias repair, particularly for milder cases, are based on normative views about male bodies and men, primarily that men should be able to stand to urinate and assumptions about the impact the inability to do this will have on psychosocial

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[11] Griffiths, DA. 2020. "Hypospadias and the Performative, Psychological and Perfect Penis" in *Talking Bodies Vol. II: Bodily Languages, Selfhood and Transgressions*, edited by Bodie A. Ashton, Amy Bonsall and Johnathan Hay. London: Palgrave Macmillian.
[12] O'Connell MA, Hutson JM and Grover SR. 2020. "Medical Management of DSD", in Hutson JM et al (eds), *Disorders/Differences of Sex Development,* Springer nature: Singapore, p.194.
[13] Eg. Hutson JM and Grover SR. 2020. "DSD Later in Childhood", in Hutson JM et al (eds), *Disorders/Differences of Sex Development,* Springer Nature: Singapore, p.168.
[14] Griffiths, 2020; interACT: Advocates for Intersex Youth. 2021. "FAQ: What is intersex" https://interactadvocates.org/faq/.
[15] American Urology Association. 2021. "Hypospadias" *Urology Care Foundation*. https://www.urologyhealth.org/urology-a-z/h/hypospadias.
[16] Mallenahalli S, Fang AH, Tong CMC, and Dangle PP. 2021. "A Review of Literature on Long-Term Outcomes of Proxmial Hypospadias – Urinary, Sexual, and Psychosocial" *Current Sexual Health Reports* 13: 38-44.

development. There are also assumptions about desirable sexual function. The range of reasons given for surgery, often in combination, include:[17]

a. To ensure the ability to urinate standing upright and to easily direct the flow of urine
b. To ensure a normative penile appearance
c. To ensure that penetrative sexual intercourse is achievable
d. To ensure that insemination through heterosexual intercourse is achievable.

18. A range of reasons for surgery in infancy, as opposed to adolescence or adulthood, are reported:
    a. Psychosocial reasoning, relating to a belief that growing up with a hypospadic penis will create stigma, shame and/or bullying due to not being easily able to urinate standing up and/or not having a normative penile appearance.[18]
    b. Faster penile growth in adolescence than in infancy may impair recovery and healing.[19]
    c. Younger children don't experience the same post-surgical pain levels as older children.[20]
    d. Younger children will not remember the procedure.[21]
19. Carmack et al. note that given surgery is mostly performed in infancy, surgery is typically performed on the basis of assumed future problems, not extant physical and/or psychosocial problems.
20. There are only relatively rare instances of surgery in adolescents or adults, where a present problem has led the individual to seek treatment.[22]

Methodological issues

21. In evaluating the outcomes of hypospadias repair in early childhood and the timing of surgery, a number of issues and methodological difficulties present in the literature can be noted.
22. While the majority of studies used the description of distal and proximal, the definition of these categories does not appear to have uniformity, and there are other classification systems employed in some studies (for example anterior, midshaft and posterior). This lack

[17] Carmack et al., 2015; Roen K, and Hegarty B. 2018. "Shaping parents, shaping penises: How medical teams frame parents' decisions in response to hypospadias" British Journal of Health Psychology 23(4): 967-981.
[18] Roen and Hegarty, 2018.
[19] Carmack et al., 2015.
[20] Carmack et al., 2015.
[21] Carmack et al., 2015.
[22] Carmack et al., 2015.

of uniformity impacts on how results in the literature are interpreted, and affects how they are considered in designing any potential legislation.

23. Some studies focus on either distal or proximal hypospadias (or use another classification system), while some studies report data for hypospadias without distinguishing or identifying the form.
24. In evaluating the outcomes of surgical repair, most published studies define outcomes as normative appearance and function with the aim of comparing between different surgical techniques.[23]
25. While there is also research that evaluates psychological health, psychosexual health, and/or quality of life, there are a number of different systems and questionnaires employed to assess these (such as the Hypospadias Objective Scoring System or the Paediatric Quality of Life Inventory, among others), with no standardisation in outcome evaluation.[24] This makes comparison between studies difficult.
26. Given the medical consensus promotes early childhood intervention there are by far fewer surgeries performed on adults, and thus a relatively limited evidence base on the deferment of hypospadias repair. For example, one study commented on by Carmack et al. reported outcomes for 1140 males who had hypospadias repair surgery, only 69 of whom had urethroplasty surgery (one of the primary surgeries addressing hypospadias) during adulthood.[25]
27. No research study has been identified that compared outcomes for an adult group who underwent early childhood repair to an adult group of men with hypospadias who had not had any surgical repair.
28. Only two studies were identified that compared outcomes for those who underwent surgery at different ages, discussed in relevant sections below.[26] Beyond this, where comparison or control groups are used these were made up of men without hypospadias, presenting a methodological issue when assessing outcomes related to the timing of surgery.

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[23] Long et al., 2017.

[24] Braga LH, Lorenzo AJ, Bagli DJ, Salle JLP and Caldamone A. 2016. "Application of the STROBE statement to the hypospadias literature: Report of the international pediatric urology task force on hypospadias" *Journal of Pediatric Urology* 12(6): 367-380; van der Horst and de Wall, 2017.

[25] Carmack A, Notini L, and Earp B. 2015. "Should Surgery for Hypospadias Be Performed Before an Age of Consent?" *Journal of Sex Research* 00: 1-12. http://dx.doi.org/10.1080/00224499.2015.1066745.

[26] Wang WW, Deng CH, Chen LW, Zhao LY, Mo JC, Tu XA. "Psychosexual adjustment and age factors in 130 men undergone hypospadias surgery in a Chinese hospital" *Andrologia* 2010 42(6): 384-388. Snodgrass W, Villanueva C, and Bush N. 2014. "Primary and Reoperative Hypospadias Repair in Adults – Are results different than in children?" *Journal of Urology* 192(6): 1730-1733

29. The majority of published research on outcomes are from single-surgeon or single-centre case series with relatively limited follow-up. One review of published studies found follow-up time ranges of between 6 weeks and 9 years.[27] Some of the impact and potential complications of hypospadias repair (especially related to sexual outcomes) will not be evident for more than a decade from the time of surgery, however there is very limited research on longer-term outcomes, especially post-pubertal.[28]
30. Further, in evaluating longer-term psychological health, psychosexual health, and/or quality of life outcomes, what evidence there is in recently published literature relates to surgeries that may have been performed 10-20 years ago, or more, with different techniques than are in use today. The longer terms outcomes of today's surgical methods may not be fully evident for another 10-20 years. This makes it impossible to assess and judge the impact of hypospadias repair taking place in the present.
31. Most papers in the field report a number of study limitations. In addition to those discussed above, there are issues with respondent bias, adequacy of survey response rates, and almost all studies being retrospective. There have been issues with underestimated rates of surgical complications because publication has occurred before sufficient length of time for follow-up has been established.[29] In conducting the review we also noted that a large number of studies appeared to involve treating specialists reporting or evaluating their own interventions or those of others in their own treating teams, raising significant issues around independence, confirmation bias and interviewer bias. While most types of study limitation were identified within the papers reviewed, this type was seldom addressed.

<u>Evidence relating to function and normative penile appearance</u>

32. There is evidence that the ability to urinate effectively standing up provided a basis for sex assignment in historical cases of ambiguous genitalia in Europe prior to the invention of surgery.[30] Upright urination thus appears to be a longstanding cultural component of sex difference.

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[27] Springer A. 2014. "Assessment of outcome in hypospadias surgery – a review" *Frontiers in Pediatrics*. https://doi.org/10.3389/fped.2014.00002.

[28] Rynja SP, de Jong TPVM, Bosch JLHR, de Kort LMO. 2011. "Functional, cosmetic and psychosexual results in adult men who underwent hypospadias correction in childhood." *Journal of Pediatric Urology* 7(5): 504-515; Long CJ, Chu DI, Tenney RW, Morris AR, Weiss DA, Shuklas AR, Srinivasan AK, Zderic SA, Kolo TF, Canning DA. 2017. "Intermediate-Term Followup of Proximal Hypospadias Repair Reveals High Complication Rate" *Journal of Urology* 197: 852-858.

[29] Long CJ and Canning DA. 2016. "Hypospadias: Are we as good as we think when we correct proximal hypospadias?", *Journal of Pediatric Urology* 12: 196.e1-196.e5

[30] Mak, G. 2013. *Doubting Sex: Inscriptions, bodies and selves in nineteenth-century hermaphrodite case histories*. Manchester: Manchester University Press.

33. However, Roan and Hegarty report that there is no known research that establishes whether it is important to psychosocial health for males to urinate standing.[31] Given this, it is difficult to evaluate whether an inability to urinate standing will have an impact on a boy as he grows up.
34. One study which compared a control group of men who did not have hypospadias with a group of men who had hypospadias (but had not had surgical repair) found that some men sit to urinate, whether or not they had hypospadias, and that doing so does not appear to bother men who sit to urinate, again whether or not they had hypospadias.[32]
35. That surgery both doesn't always prevent, and can itself be the cause, of a range of psychosocial issues is noted in resources produced by the Hypospadias and Epispadias Association (HEA), a USA based support group. They report that everything from bullying to poor self-esteem and lack of genital confidence can be experienced by those with hypospadias who have had repair surgery.[33] HEA does not have a position on surgery, instead aiming to provide a range of information to assist in decision making by both parents and those considering surgery for themselves.
36. A meta-analysis of the wellbeing of boys with hypospadias, which examined 13 studies, found that 'no conclusion can be drawn with regard to the importance of corrective surgery itself for the children's psychosocial and psychosexual development'. 'Furthermore, the review pointed out that the guidelines for surgical treatment are partly based on psychological assertions that have not been empirically confirmed.'[34]
37. In considering how adults viewed their childhood hypospadias repair, one review found that patient perceptions of cosmetic outcome were generally positive in the majority of patients, however the number of patients reporting satisfaction fell below 50% for those who had proximal and more complex forms of hypospadias.[35]

<u>Evidence relating to sexual outcomes</u>

38. In considering reasoning for surgery related to sexual intercourse and insemination it is important to again emphasise that surgery is overwhelming being performed in early

[31] Roan and Hegarty, 2018.
[32] Schlomer B, Breyer B, Copp H, Baskin L, and DiSandro M. 2014. "Do adult men with untreated hypospadias have adverse outcomes? A pilot study using a social media advertised survey" *Journal of Pediatric Urology* 10: 672-679.
[33] Hypospadias and Epispadias Association. 2017. "Hypospadias: An Overview". http://heainfo.org/index.php/2017/07/08/hypospadias-an-overview/
[34] Schonbucher VB, Weber DM, and Landolt M. 2008. "Psychosocial Adjustment, Health-Related Quality of Life, and Psychosexual Development of Boys with Hypospadias: A systematic review" *Journal of Pediatric Psychology* 33(5): 530, 534.
[35] Rynja et al., 2011.

childhood, and thus on a patient who will likely not become sexually active for more than a decade from the time of surgery. Given this, long-term follow up studies are important.

39. However due to issues in long-term follow up studies, especially those that might cover multiple decades, it is very difficult to evaluate sexual functioning and fertility outcomes for childhood hypospadias repair.[36]
40. There is one study identified that compared groups who had surgery at different ages and which investigated psychosexual status and sexual function. This consisted of 130 patients who had surgery between 1988 and 2007, with one group of those who had hypospadias repair under age 10, one who had repair age 10-18, and one group who had repair after age 18, in addition to a control group of men without hypospadias.[37]
41. Overall, those who had hypospadias repair had worse outcomes than the control group with regard to 'penis development' (described as penis length and circumference), however there was no significant difference found between the control groups and those who had hypospadias repair with regard to libido strength, penile erectile function and overall sexual satisfaction, nor was there are any significant difference between the different age of surgery groups.[38]
42. Regarding fertility and insemination, some population-based studies have found lower paternity rates in men with hypospadias, however Gul et al. noted in their review of this literature that whether this is due to fertility issues or functional factors or other reasons was not clear and required more research.[39] Likewise the impact of hypospadias repair on fertility and insemination is not clear.
43. Further there are assisted reproductive technologies (ART) that may assist couples who are not able to achieve insemination through intercourse, and that are used by many different kinds of couples for a wide range of reasons, not just hypospadias. It is not clear what the prevalence of ART is among hypospadias populations and how this compares to the general population.

<u>Evidence base relating to timing of surgery</u>

44. With regards to reasons for surgery during early childhood, the claim that younger children don't experience the same post-surgical pain levels as older children is disputed. There is

[36] Gul M, Hildorf S, and Silay MS. 2021. "Sexual functions and fertility outcomes after hypospadias repair" *Your Sexual Medicine Journal* 33: 149-163.
[37] Wang WW, Deng CH, Chen LW, Zhao LY, Mo JC, Tu XA. "Psychosexual adjustment and age factors in 130 men undergone hypospadias surgery in a Chinese hospital" *Andrologia* 2010 42(6): 384-388.
[38] Wang et al., 2010.
[39] Gul et al., 2021

evidence that this is not the case as Carmack et al. summarise in their review of relevant literature: [40]

> It is now recognized that even very young infants have a well-developed capacity to experience pain and that developmental differences in expression of pain, including an infant's lack of ability to use language, seriously complicate such an assessment (see, e.g., Fitzgerald, 1998; Franck, Greenberg, & Stevens, 2000; Frisch & Simonsen, 2015; Johnston, Stevens, Craig, & Grunau, 1993).[41]

45. Regarding memory and hypospadias repair, those who did not recall their surgery were likely to have positive body image and be satisfied.[42] However, memory formation is complex, and memory of events can be impacted by parents and a wide range of other factors, so the link between memory of surgery and positive outcomes should be approach cautiously.[43]
46. There is also an argument put forward that growth is slower during infancy and that this aids healing. However, as Carmack et al. point out, penile growth is even slower after puberty since the penis has reached its full size.[44] Further, pubertal growth itself might affect the final cosmetic and functional aspects of hypospadias repair and in some cases necessitate further surgery.[45]
47. Some earlier studies indicated a possible correlation of later repair with more complications, but more recent reviews have found no association between age of initial repair and the number of complications. One of these recent studies, by Snodgrass et al., reviewed over a thousand surgeries performed by the same surgeon using the same technique. [46] It found no correlation between age and complication rates. This could indicate that earlier findings were due to differences in surgeon or technique, rather than the patient's age.
48. A British study found parental decision regret declined with increasing age at first surgery, though this was not a dominant factor when all variables were subject to multivariate analysis.[47]
49. Similarly, one issue that may be present with surgery at a later age is that because the large majority of hypospadias repairs are performed by paediatric surgeons and urologists, there

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[40] Carmack et al., 2015.
[41] Carmack et al., 2015: 4.
[42] van der Horst and de Wall, 2017; Carmack et al., 2015.
[43] Carmack et al., 2015.
[44] Carmack et al., 2015.
[45] Rynja et al., 2011.
[46] Snodgrass et al., 2014.
[47] Bethell GS, Chhabra S, Shalaby MS, Corbett H and Kenny SE. 2020. "Parental decisional satisfaction after hypospadias repair in the United Kingdom", *Journal of Pediatric Urology* 16(2): 164.e1-164.e7.

may be a lack of expertise in hypospadias repair for surgeons who work with older populations.[48]

Surgical Risks and Complications

50. Physical health risks of hypospadias repair surgeries include wounds on the penis (fistulas), scar tissue at the opening or inside the urethra that may cause a blockage (stenosis or stricture), and chronic pain and infections at the operation site, among other complications. These are in addition to the general risks that all surgery carries.[49]
51. Complications can occur anywhere between shortly after surgery and up to decades after surgery.[50] Most studies focus on the 1-to-2-year period following a surgery, and reports on longer-term complication prevalence are less common.[51]
52. Some complications require further surgical intervention, which themselves may result in further complication and further surgical repair. There are anecdotal accounts of individuals having up to 17 surgeries associated with their hypospadias.[52]
53. In considering complication rates it should be noted that some studies focus on either distal or proximal (or use another classification system), while some report data for all hypospadias repair without distinguishing. This can make it difficult to evaluate or compare claimed rates.
54. In general, complication rates appear higher for interventions on proximal (urethra opening on the shaft, or scrotum) than the milder distal (urethra opening on or near the glans) hypospadias.[53]
55. The American Urological Association reports that less than 1 in 10 distal repairs have complications. No evidence is provided for this rate.[54] Two recent surveys of relevant literature found a range of reports on long-term complications,[55] which call into question the low complication rate stated by the American Urological Association.

[48] Hypospadias and Epispadias Association, 2017.
[49] Carmack et al., 2015; American Urological Association, 2021.
[50] Tack LJW, Springer A, Riedl S, Tonnhofer U, Hiess M, Weninger J, Mahmoud A, Van Laeke E, Hoebeke, Cools M, and Spinoit AF. 2021. "Adolescent and Young Adult Urogenital Outcome following Childhood Hypospadias Repair: Perfection Revisited" *Journal of Urology*. https://doi.org/10.1097/JU.0000000000001869; Long et al., 2017.
[51] Carmack et al., 2015
[52] Mosaic Science. 2016. "My life with hypospadias". https://mosaicscience.com/extra/my-life-hypospadias-14-mins/.
[53] Carmack et al., 2015.
[54] American Urological Association, 2021.
[55] Carmack et al., 2015; Roan and Hegarty, 2018.

56. The percentage of proximal hypospadias patients requiring more than one hypospadias related surgery in their lifetime was reported by Camack et al. as ranging from about 25% up to 50%, depending on the report and research methods.[56] Long and Canning report complication rates of similar magnitudes.[57] Similarly, Long et al. report a 56% complication rate for proximal hypospadias repair within a 32 month follow up window, and recommend further follow up research to better understand and evaluate complication rates.[58]
57. One recent study found that hypospadias repair when performed on children younger than 12 months was associated with a higher rate of subsequent surgical intervention. This study also found an 39% rate of at least one addition surgical intervention, with some of these additional interventions performed decades after the initial repair.[59] Notably all participants in this study had pre-puberty initial intervention.
58. Rates of complication also vary with specific surgical technique.[60]
59. Carmack et al. state that 'The only conclusion that can safely be drawn at this time is that the complication rates are at least those reported in the literature and may very well be higher, given all of the barriers to collecting accurate long-term follow up data.'
60. Roan and Hegarty conclude that it is not clear whether rates of complications are well-known and understood by clinicians managing hypospadias or whether the risks of complications (and difficulty in evaluating these) are fully and accurately communicated to parents considering hypospadias repair for their children.[61]

<u>Non-surgical care</u>

61. Parents choosing to defer surgery can sometimes be framed by surgeons as 'doing nothing', when there a number of non-surgical options that may address some of the potential issues surgical repairs aims to prevent.[62]
62. Further, these non-surgical options might help and be pathways to decision-making about surgery as an individual grows up and becomes able to provide input or consent. The aim of deferring surgery is not that hypospadias repair never takes place, but that individuals are able to participate in the decision to have surgery.

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[56] Carmack et al., 2015.
[57] Long CJ and Canning DA. 2016. "Hypospadias: Are we as good as we think when we correct proximal hypospadias?", *Journal of Pediatric Urology* 12: 196.e1-196.e5
[58] Long et al., 2017.
[59] Tack et al., 2021.
[60] Rampersad R, Nyo, YL, Hutson, J. et al. 2017. "Foreskin reconstruction vs circumcision in distal hypospadias". *Pediatr Surg Int* 33: 1131–1137.
[61] Roan and Hegarty, 2018.
[62] Roan K. 2008. "'But we have to do something': Surgical 'Correction' of Atypical Genitalia", *Body & Society* 14(1): 47-66.

63. Counselling and psychosocial support for parents and/or for the child may help with focusing on the parent-child relationship, rather than on a child's anatomical difference. Roan and Hegarty note that this can assist in framing bodies with hypospadias as 'a body that is loveable, and a body that belongs to someone who deserves to be allowed to make his own decisions when he is old enough.'[63]
64. Bioethicist Alice Drager has commented that the use devices or prosthetics that might assist someone with hypospadias in standing to urinate has the potential to assist the individual in confirming whether surgery is necessary for them and/or what specific surgical outcomes are desired thus helping to identify the specific procedures and method.[64] No discussion of such devices or their use was found in the reviewed literature about hypospadias.
65. Dreger also related an anecdotal story about a family who decided against childhood surgical repair for their child's hypospadias. In an effort to support his child, the father and other male family members decided to sit to urinate so that his child grew up in an environment where this was normalised; 'Rather than getting that little boy to come to their norm, they decided to go to his.'[65]
66. An individual, and/or their parents, may go on to decide that surgery is in their best interests, but there are a number of non-surgical options that might be explored before considering surgery. The framing of deferring surgery as doing nothing is both inaccurate, and itself may be contributing to parents being more inclined to choose surgery without exploring non-surgical options that still constitute care and intervention.[66]
67. Roan and Hegarty observe that hypospadias tended to be considered and treated by a surgeon alone, or potentially with other surgeons, rather than with a Multi-Disciplinary Team (MDT). They suggest that one benefit of an MDT evaluating proposed medical interventions is that it might increase inclusion of non-surgical supports such as psychological support and counselling.[67]

<u>Conclusions</u>

68. There appears little if any evidence in the reviewed literature that surgery for hypospadias in early childhood produces better outcomes than surgery undertaken later with personal consent. This is largely due to a lack of long-term outcome results for those undergoing

[63] Roan and Hegarty, 2018: 977.
[64] Alice Dreger. 2017. "Do you have to pee standing up to be a real man?" *Pacific Standard, The Science of Society*. https://psmag.com/social-justice/pee-standing-real-man-73133.
[65] Dreger, 2017.
[66] Roen and Hegarty, 2018.
[67] Roen and Hegarty, 2018.

childhood surgical intervention, and a lack of evidence about surgical intervention in adolescence or adulthood. During the current policy development project, some stakeholders have suggested that deferring surgery would represent an "experiment", yet some practitioners have referred to current early intervention practices in the same terms.[68]

69. Two papers were identified that suggested age at first surgery was not a predictor of long-term outcomes.
70. If hypospadias, whether in its entirety or only in complex forms, is captured within the scope of any potential regulatory framework, it would be desirable systematically to collect data about both surgical and non-surgical interventions and their outcomes. The inclusion of hypospadias (or other variations in sex characteristics) in the scope of a regulatory response could be reviewed at appropriate intervals and could be removed in future if there is evidence to suggest that deferment of surgery is leading to negative outcomes.
71. Lastly, while there is inconclusive evidence for the benefit of childhood hypospadias repair, neither is it currently possible to argue that deferring surgery to adolescence or adult hypospadias repair will definitely produce better outcomes. What deferring surgery would do is create an opportunity for individuals to make an informed choice about a surgical intervention performed on their bodies.

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[68] Liao, L-M, Wood D and Creighton SM. 2015. "Parental choice on normalising cosmetic genital surgery", *BMJ* 351: h5124.

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Defining Hypospadias in Prescribed List

Ministers have indicated that only more complex hypospadias cases should be in the scope of the legislation. This will require the regulation to describe this for both medical accuracy and legal certainty. This note summarises literature on this issue and recommends a definition.

Source	Description	Notes
Hughes, I.A., Houk, C., Ahmed, S.F., Lee, P.A. and Society, L.W.P.E., 2006. Consensus statement on management of intersex disorders. Journal of pediatric urology, 2(3), pp.148-162.	"severe hypospadias"	Severe hypospadias is included as an example of an 46,XY DSD. The paper goes on to note that isolated perineal hypospadias, or mild hypospadias with undescended testis are criteria that may suggest a DSD is present though does not state or imply that these *are* a DSD themselves.
Lee, P.A., Nordenström, A., Houk, C.P., Ahmed, S.F., Auchus, R., Baratz, A., Dalke, K.B., Liao, L.M., Lin-Su, K., Looijenga 3rd, L.H. and Mazur, T., 2016. Global disorders of sex development update since 2006: perceptions, approach and care. Hormone research in paediatrics, 85(3), pp.158-180.	"Among patients with hypospadias and cryptorchidism, currently the diagnosis of specific DSD conditions is generally limited to those with proximal hypospadias with cryptorchidism."	Cryptorchidism is the absence of at least one testicle from the scrotum.
Johnson, E.K., Jacobson, D.L., Finlayson, C., Yerkes, E.B., Goetsch, A.L., Leeth, E.A. and Cheng, E.Y., 2020. Proximal hypospadias—isolated genital condition or marker of more?. The Journal of Urology, 204(2), pp.345-352.	"Practically the DSD designation has been largely limited to boys with proximal hypospadias who also have undescended testis, as affirmed in the 2016 consensus update."	Testis is the singular, meaning that have just one undescended testicle would qualify as per the 2016 update, above.
Snodgrass, W., Macedo, A., Hoebeke, P. and Mouriquand, P.D., 2011. Hypospadias dilemmas: a round table. Journal of Pediatric Urology, 7(2), pp.145-157.	Hoebeke – "Consensus here that those patients who are suspected to have DSD (because of associated genital anomalies, most often nonscrotal gonad) should be evaluated." Snodgrass - "By definition, neither hypospadias nor undescended testes is considered a disorder of sexual development, even though they	This roundtable discussion demonstrated a variety of views among senior paediatric urologists regarding the question of hypospadias and DSD designation. The discussion also explored the question of evaluating the 'severity' of hypospadias, noting that

	may represent part of the spectrum that includes 46XY DSD, and occasional patients with the combination of hypospadias and undescended testis have ovotesticular DSD or mixed gonadal dysgenesis. Ultimately, all classifications of biologic systems are somewhat arbitrary, yet necessary for scientific communication."	severity can be difficult to fully gauge pre-operatively. However, 'severity' is a different classification system than that of meatal location description, proximal or distal. The paper noted that these descriptions are still valid classifications even though they are not wholly standardised.
Australian Human Rights Commission Report	"The most common referrals to the Differences of Sex Development (DSD) forum were for timing and need for gonadectomy for non-functioning gonads with malignant potential and hypospadias surgery for boys with complex hypospadias associated with other genital variations (eg, undescended testes)." "Specialist clinicians from Royal Children's Hospital in Melbourne said that the cohort they see includes complex hypospadias"	At least one submission reported to the AHRC that their DSD MDT sees and treats cases of hypospadias with other genital variations. The report further noted that "Some clinical stakeholders stated that they do not see people with certain specific variations or those with 'simple' hypospadias. It was suggested that MDT review is generally for those characterised as more complex cases." The AHRC report uses 'testes' implying that both testes must be undescended to meet the criteria. It is not clear if this is drawn directly from submissions, or a result of language used in the submission for ease of comprehension.
Kalfa, N., Gaspari, L., Ollivier, M., Philibert, P., Bergougnoux, A., Paris, F. and Sultan, C., 2019. Molecular genetics of hypospadias and cryptorchidism recent developments. *Clinical Genetics*, *95*(1), pp.122-131.	"…posterior and perineal hypospadias with micropenis and major chordee are one of the most severe 46,XY DSDs with undetermined sex at birth."	This paper uses the classification system of 'posterior and perineal', rather than proximal.
Wong, Y.S., Tam, Y.H., Pang, K.K.Y. and Yau, H.C., 2018.	"Most cases encountered in clinical practice are distal	This paper provides both a definition of proximal and a

Incidence and diagnoses of disorders of sex development in proximal hypospadias. Journal of Pediatric Surgery, 53(12), pp.2498-2501.	hypospadias while a minority of patients have proximal hypospadias with more severe hypoplasia of ventral tissue beyond a proximal division of corpus spongiosum." "Patients presenting with proximal hypospadias and one or more of the coexisting anomalies of micropenis, undescended/impalpable testes, and penoscrotal transposition/bifid scrotum should warrant DSD evaluation. Presence of bilaterally descended testes in scrotum does not preclude the possibility of DSD."	discussion of hypospadias relation to a DSD designation. It also implicitly assumes that hypospadias indicates a possible DSD, but is not itself a DSD.
Kearsey, I. and Hutson, J.M., 2017. Disorders of sex development (DSD): not only babies with ambiguous genitalia. A practical guide for surgeons. Pediatric surgery international, 33(3), pp.355-361.	"Use of the word, 'hypospadias', not only implies the neonate is a male, but also that the anomaly is 'just a minor problem in urethral development', when in fact it may be a complex global anomaly of sexual development of both internal as well as external genitalia. To avoid making this error, doctors need a simple algorithm to use at the bedside to enable them to distinguish 'simple hypospadias' from a serious DSD as the appearances can overlap. The simplest rule to use is to assume the baby may have a DSD if the 'hypospadias' is associated with either of the following: (1) bifid scrotum (i.e. nonfused labioscrotal folds) or (2) impalpable testis (unilateral or bilateral) as when the testis is missing how can we know that there is a testis at all? Conversely, 'hypospadias' can be diagnosed if the urethral anomaly on the phallus is associated with a fused scrotum containing two descended	

	testes, effectively excluding a generalised anomaly of androgenic function.”	

Recommendation:

“Proximal hypospadias with cryptorchidism” is an appropriate description for inclusion in the prescribed list.