Diphallia: literature review and proposed surgical classification system

Dylan John Kendrick D*† and Roy Mark Kimble*†‡

*School of Medicine, University of Queensland, Brisbane, Queensland, Australia
†Department of Paediatric Surgery, Urology, Burns & Trauma, Queensland Children’s Hospital, Brisbane, Queensland, Australia and
‡School of Health Sciences, Queensland University of Technology, Brisbane, Queensland, Australia

Key words
classification, congenital anomaly, diphallia, duplicate penis, urethral duplication.

Abstract
Background: Diphallia occurs once in 5–6 million births, with no two patients presenting with the same anatomical variation. Here we discuss a review of diphallia case reports, as well as present a new surgical classification system based on the soft tissue composition of the two phalluses, the anatomy of the urethra present within the most normal phallus and the bladder configuration.

Methods: Eighty-seven diphallia case reports were collected and analysed, excluding those presented in animals and articles that were non-English, with the results compiled to provide an in-depth reference of the specific anatomy found in diphallia patients and the associated abnormalities.

Results: Our proposed classification system was then applied to each patient and the most common configuration based on our classification system presented, along with commonly seen associated anomalies.

Conclusion: The reviewed cases represent a subset of the most unique diphallia patients; thus, several cases may be left unreported. Future reports can then be categorized, aiding as a reference, and potentially building on the classification, should the patient not fit into a specific group, leading to an expansion of the classification system.

Introduction
Diphallia or duplicate penis is an extremely rare embryological anomaly with a wide range of anatomic appearances ranging from small accessory tissues to complete duplications of the phallus, glans, urethras, and bladders, as well as an extensive list of associated abnormalities. The phalluses are usually unequal in size and positionally, can lie side by side, stacked on top of the other in the sagittal plane, or with little association to each other. This paper assesses patients within the literature to summarize diphallia variants, as well as build on the existing classification of diphallia to include a more specific categorization of the phallus soft tissue, the urethral anatomy, and the bladder configurations to create a surgical classification system.

Background
Diphallia is estimated to occur in 1 out of 5–6 million births, with around 100 patients being reported within the literature. The oldest published instance of diphallia was reported in 1609 by Johannes Jacob Wecker; ‘in Bologna during public dissections the cadaver of a man who had a double penis’, however the earliest pictorial record of diphallia dates back to 1862 in the Lupanar (Latin for brothel) in Pompeii, in which a painting on the wall depicted a completely diphallic man (Fig. 1).3

Embryology
Previously, Cecil3 submitted four embryological explanations for diphallia. The first includes the bladder, the prostatic urethra, and the penis being derived from a bilateral anlagen, which normally gives a single end product by fusion, meaning diphallia is a product of an incompletely fused anlagen. Next, diphallia may be an atavism, as snakes and lizards normally possess double penis or possibly represents a teratoid structure. Cecil also suggested that it may be a minor degree of duplication, much like supernumery digits, of the individual.
Hollowell et al. however affirm that these explanations are incomplete and suggests that that embryologically, the diphallia anomaly occurs in the fetus between the third and seventh week of gestation, in which an insult hampers normal functioning of the caudal cell mass of the fetal mesoderm at the time of the urogenital sinus separating from the genital tubercle into the penis. Complete diphallia may then stem from longitudinal duplication of the infraumbilical cloacal, with the subsequent mesodermal migration leading to the formation of two separate and complete sets of genital tubercles, genital folds, and genital swellings.3,4

Methods
For this paper, a review of published diphallia patients was conducted by one reviewer. Google scholar and PubMed were analysed using key phrases diphallia, double penis, pseudophallia and bifid glans giving a total of 518 articles. Articles were screened based on title and abstract, making sure to include relevant case reports in humans, excluding articles in animals, duplicate articles and non-English articles. Following the screening of relevant inclusion and exclusion criteria, a total of 76 articles were analysed, which totalled 87 relevant diphallia patients found within the literature. Each patient was then analysed and presented in Table 1. From each patient, the unique anatomical variant for each was then examined. This included first addressing the age of the patient at the time of presentation, followed by categorizing the soft tissue structure of each phallus, the anatomy of the scrotum and testicles, as well as if there was a penoscrotal transposition. The reports were also analysed for their urethral anatomy, and if there was a hypospadias or epispadias present, as well as the bladder configuration. Lastly, the associated abnormalities were summarized based on the reviewers best clinical judgement. Each patient was then classified based on the proposed classification system below.

Classification
Schneider80 has previously classified diphallia into four main categories:
(1) Duplication of the glans alone
(2) Bifid diphallia
(3) Complete diphallia with each penis having two corpora cavernosa and a corpus spongiosum
(4) Pseudodiphallia in which there is a rudimentary accessory atrophic penis existing independently of the normal penis

Our proposed classification is based on anatomical variants found within the literature on diphallia, which builds on Schneider’s classification in order to provide a more specific description of the phallus soft tissue, as well as include a description of the pathway of the most normal urethra and the bladder configuration. The classification method has been proposed to be used to simplify the categorization of diphallia patients, using the system to classify the most intact phallus, the most normal urethra, and whether there is an additional surgical step regarding the bladder. The system will place the anatomy of diphallia into specific categories based on the structure of the phallus, the urethral anatomy and the bladder formations. Table 2 below displays the proposed categories.

Results
With a review of the literature, and application of the proposed classification system, common diphallia configurations can be displayed. While these individuals may still differ in terms of specific anatomy, such as the relation of the phalluses to one another, the
| Author, Year | Number of corpora cavernosa | Hypospadias/ Epispadias | Scrotum | Testes | Penoscrotal Transposition | Urethra | Bladder | Other anomalies | Classification |
|--------------|---------------------------|-------------------------|---------|-------|--------------------------|--------|--------|---------------|---------------|
| Acimi⁶       | Phallus 1: 2 Phallus 2: 2 | Hypospadias             | Normal  | Descended bilaterally    | No      | Duplicate               | Single  | Imperforate anus 1Aα |
| Acimi⁶       | Phallus 1: 2 Phallus 2: 2 | Hypospadias             | Bifid   | Descended bilaterally    | No      | Duplicate, one functional | Single  | Unilateral Kidney agenesis Ureterovesical duplication Umbilical hernia 1Bα |
| Adair and Lewis⁷ | Phallus 1: 2 Duplicate Glans Phallus 1: 2 | Hypospadias             | Normal  | Descended bilaterally    | No      | Single                  | Single  | Unilateral Kidney agenesis Unilateral Kidney agenesis Musculoskeletal anomalies 7Aα |
| Al-Herbish and Al-Samarrai⁸ | Phallus 1: 2 | Hypospadias             | Bifid   | Descended bilaterally    | No      | Duplicate with bifurcation of one urethra Duplicate | Single  | Hemivertebra and absent first rib 1Aα |
| Aihole, 2015⁹ | Phallus 1: 2 | N/A                     | Normal  | Descended bilaterally    | No      | Duplicate               | Duplicate | Pre-axial polydactyly Solitary Kidney Atrial septal defect Anal atresia Duplicate rectum, colon, cecum, appendix and terminal ileum Rectovesical fistula Wide diastasis of the pubic bones and partial sacral agenesis 1Bα |
| Akgül et al.¹⁰ | Duplicate Glans Phallus 1: 2 Phallus 2: 2 | N/A                     | Normal  | Descended bilaterally    | No      | Duplicate               | Duplicate | Wide diastasis of the pubic bones and partial sacral agenesis 1Bα |
| Aleem¹¹       | Phallus 1: 1 Phallus 2: 1 | Epispadias              | Bifid   | Descended bilaterally    | No      | Single                  | Single  | 1Dα |
| Ali¹²         | Phallus 1: 1 Phallus 2: 2 | N/A                     | Normal  | Descended bilaterally    | No      | Single                  | Single  | 2Aα |
| Arya et al.¹³ | Phallus 1: 2 Phallus 2: 2 | Epispadias              | Bifid   | Descended bilaterally    | No      | Duplicate, no urethral plates Exstrophy | N/A     | 1Dγ |
| Bakheet and Refaei¹⁴ | Phallus 1: 2 Phallus 2: 2 | N/A                     | Bifid   | Descended bilaterally    | No      | Duplicate               | Single  | 1Aα |
| Bhat et al.¹⁵ | Phallus 1: 2 Phallus 2: 2 | Hypospadias             | Bifid   | Descended bilaterally    | No      | Duplicate, opening normally and into lateral wall of bladder | Single  | Duplicate colon, rectum, anus 1Aα |
| Blanco¹⁶       | Phallus 1: 2 Phallus 2: 2 | Hypospadias             | Normal  | Descended bilaterally    | No      | Duplicate               | N/A     | 1Aα |
| Cernach et al.¹⁷ | Phallus 1: 2 Phallus 2: 2 | N/A                     | Bifid   | Descended bilaterally    | No      | Duplicate               | Duplicate | Duplicate colon, one imperforate hemivertebrae and diastasis of pubic symphyses 1Aβ |
| Chadha et al.¹⁸ | Phallus 1: 2 Phallus 2: 2 | N/A                     | Bifid   | Descended into lateral compartments | No      | Bifurcation at prostatic urethra | Single  | N/A     |
| de Oliveira et al.¹⁹ | Phallus 1: 2 Phallus 2: 2 | N/A                     | Normal  | Descended bilaterally    | No      | Duplicate               | Duplicate | N/A     |
| Deshpande²⁰   | Phallus 1: 2 | N/A                     | Bifid   | No                      | Phallus 2: 2 | Duplicate | Phallus 2: 2 | Duplicate | N/A     |
| Author, Year | Age at definitive treatment | Number of corpora cavernosa | Hypospadias/Epispadias | Scrotum | Testes | Penoscrotal Transposition | Urethra | Bladder | Other anomalies | Classification |
|--------------|-----------------------------|-----------------------------|------------------------|---------|-------|--------------------------|--------|--------|----------------|---------------|
| Dewan et al. | 7 yrs.                      | Phallus 1: 2                | N/A                    | Bifid   | Descended unilaterally   | No      | Duplicate | Single     | Imperforate anus | 1Aα           |
| Djordjevic and Perovic | 15 mo.                         | Phallus 1: 2                | Hypospadias            | Bifid   | Descended bilaterally   | No      | Duplicate | Duplicate | Symphyseal diastasis | 1Aβ           |
| Dunn et al.  | 3 yrs.                      | Phallus 1: 2, Phallus 2: 2  | N/A                    | Bifid   | Descended bilaterally   | No      | Duplicate | N/A        | Pelvic kidney      | 1Aβ           |
| Dutta et al. | Neonate                     | Phallus 1: 2, Phallus 2: 2  | N/A                    | Bifid   | Descended bilaterally   | No      | Duplicate | Single     | Single Kidney agenesis | 7Bα           |
| Elsawy et al.| 1 mo.                       | Phallus 1: 2, Phallus 2: 2  | N/A                    | Normal  | Descended bilaterally   | No      | Single    | Single     | Single Kidney agenesis | 7Bα           |
| Frollo et al.| 84 yrs.                     | Phallus 1: 2, Phallus 2: 2  | Hypospadias            | Normal  | Undescended bilaterally | No      | Duplicate | Duplicate | Horseshoe kidney | 1Aβ           |
| Gavali et al. | 5 yrs.                      | Phallus 1: 2                | Hypospadias            | Normal  | Descended bilaterally   | No      | Duplicate | Single     | Single Kidney agenesis | 7Bα           |
| Ghafoori et al. | 5 yrs.                     | Phallus 1: 2, Phallus 2: 1  | N/A                    | Normal  | Descended bilaterally   | No      | Duplicate | N/A        | 3Aα           |
| Goad et al.  | 13 yrs.                     | Phallus 1: 2                | Hypospadias            | Normal  | Descended bilaterally   | No      | Single    | Single     | 1Aβ           |
| Gupta and Virdi | 10 yrs.                   | Phallus 1: 2                | N/A                    | Normal  | Descended bilaterally   | No      | Duplicate | N/A        | 1Aβ           |
| Gyftopoulos et al. | Neonate                 | Phallus 1: 2                | Hypospadias            | Bifid   | Undescended bilaterally | No      | Duplicate | Duplicate | 1Aβ           |
| Hanine et al. | 8 yrs.                      | Phallus 1: 2                | N/A                    | Normal  | Descended bilaterally   | No      | Single    | Single     | 2Aα           |
| Hollowell et al. | 5 mo.                    | Phallus 1: 2                | Hypospadias            | Bifid   | Descended bilaterally   | No      | Duplicate | Duplicate | 1Aα           |
| Jesus et al. | 20 yrs.                     | Phallus 2: 2, Phallus 2: 2  | Hypospadias            | Normal  | Descended bilaterally   | No      | Duplicate | Single     | Bilateral inguinal hernias | 7Cα           |
| Author, Year | Age at definitive treatment | Number of corpora cavernosa | Hypospadias/Epispadias | Scrotum | Testes | Penoscrotal Transposition | Urethra | Bladder | Other anomalies | Classification |
|--------------|-----------------------------|----------------------------|------------------------|---------|-------|--------------------------|--------|--------|----------------|---------------|
| Johnson et al. | 17 yrs. | Phallus 1: 2 | N/A | Normal | Descended bilaterally | No | Duplicate | Single | N/A | 7Aα |
| Karagöz et al. | 15 yrs. | Phallus 1: 1 | N/A | Normal | Descended bilaterally | No | Single | Single | N/A | 7Aα |
| Kardasevic et al. | Neonate | Phallus 1: 2 | Hypospadias | #NAME? | Descended bilaterally | No | Duplicate | Duplicate | Atrial and ventricular septal defect | 1Aβ |
| Karna and Kapur | Neonate | Phallus 1: 2 | N/A | Bilid | Undescended bilaterally | No | Duplicate | Single | Musculoskeletal anomalies | 1Aα |
| Kaufman et al. | 15 yrs. | Phallus 1: 2 | Hypospadias | Normal | Descended bilaterally | No | Duplicate, with third perineal urethra | Single | Imperforate anus | 1Aα |
| Keckler | Neonate | Phallus 1: 2 | N/A | Normal | Descended bilaterally | No | Duplicate | Single | N/A | 1Aα |
| Kendrick et al. | 3 mo. | Phallus 1: 2 | N/A | Normal | Descended bilaterally | Partial | Duplicate, opening normally and into lateral wall of bladder | Single | N/A | 2Bα |
| Khorramirouz et al. | 6 yrs. | Phallus 1: 2 | N/A | Normal | Descended bilaterally | No | Duplicate, unilateral stenosis | Duplicate | Horseshoe Kidney | 7Bβ |
| Krif et al. | Neonate | Phallus 1: 2 | Epispadias | Normal | Descended bilaterally | No | Duplicate | Exstrophy | Inguinal hernia | 1Dy |
| Kundal et al. | 3 yrs. | Phallus 1: 2 | Epispadias | Normal | Descended bilaterally | No | Duplicate | Single | N/A | 1Aα |
| Landy et al. | Neonate | Phallus 1: 2 | N/A | Normal with accessory scrotum | Descended bilaterally | No | Duplicate | Single | Imperforate anus | 1Aα |
| Larsen | 14 yrs. | Phallus 1: 1 | N/A | Normal | Descended bilaterally | Partial | Duplicate | Single | Atrophic leg | 3Aα |
| Leite et al. | 2 yrs. | Phallus 1: 1 | N/A | Normal | Descended bilaterally | No | Duplicate, one with blind ending | Single | Short oesophagus | 3Aα |
| Maher et al. | Neonate | Phallus 1: 2 | N/A | Duplicate | Descended bilaterally | No | Duplicate | Duplicate | Separated natal clefts with no anal orifice | 1Aβ |
| Mandal and Sahu | Neonate | Phallus 1: 2 | N/A | Duplicate | Descended bilaterally | No | Duplicate | Single | N/A | 1Aα |
| Author, Year | Age at definitive treatment | Number of corpora cavernosa | Hypospadias/Epispadias | Scrotum | Testes | Penoscrotal Transposition | Urethra | Bladder | Other anomalies | Classification |
|--------------|----------------------------|----------------------------|------------------------|---------|-------|---------------------------|--------|--------|-----------------|----------------|
| Marti-Bonmati et al. | Neonate | Phallus 1: 2 Phallus 2: 2 | Hypospadias | Ectopic scrotal tissue | Descended bilaterally | No | Duplicate, one with blind ending | Single | Imperforate anus | 1Aγ |
| Maruyama et al. | Neonate | Phallus 1: 1 | Hypospadias | Bifid | Descended unilaterally | No | Duplicate | Single | Tracheoesophageal fistula with oesophageal atresia | 3Cα |
| Matsumoto et al. | 12 mo. | Phallus 1: 1 Phallus 2: 2 | Hypospadias | Bifid | Descended bilaterally | Complete | Duplicate | Single | Atrial septal defect | 2Aα |
| Melekos et al. | 8 yrs. | Phallus 1: 1 Phallus 2: 1 | Hypospadias | Normal | Descended bilaterally | No | Duplicate | Single | Horseshoe Kidney | 6Cα |
| Mingazzini | 36 yrs. | Phallus 1: 1 Phallus 2: 1 | N/A | Normal | Descended bilaterally | No | Duplicate, unilateral stenosis | Single | Umbilical hernia | 3Cα |
| Mirshemirani et al. | Neonate | Phallus 1: 2 Phallus 2: 2 | Hypospadias | Bifid | Descended bilaterally | No | Duplicate | Duplicate | Imperforate anus | 1Cγ |
| | 4 yrs. | Phallus 1: 1 Phallus 2: 1 | Hypospadias | Bifid | Descended bilaterally | No | Duplicate | Single | Duplicate sigmoid colon | 3Cα |
| | 12 yrs. | Phallus 1: 2 Phallus 2: 2 | Epispadias | Bifid | Descended bilaterally | No | Duplicate | Exstrophy | Single kidney | 4Dγ |
| | 9 mo. | Phallus 1: 2 Phallus 2: 2 | N/A | Bifid | Descended bilaterally | No | Duplicate | Duplicate | Hemi-Vertebra | 1Aλ |
| Mukunda et al. | Neonate | Phallus 1: 2 Phallus 2: 2 | N/A | Bifid | Descended bilaterally | No | Bifurcation at prostatic urethra | Single | Meckel’s diverticulum | 1Eα |
| Mutlu et al. | 9 yrs. | Phallus 1: 2 Phallus 2: 1 | N/A | Normal | Descended bilaterally | No | Bifurcation at prostatic urethra | Single | Rotational anomaly of right kidney | 2Eα |
| Nunez et al. | Neonate | Phallus 1: 2 Phallus 2: 1 | Hypospadias | Normal | Descended bilaterally | No | Bifurcation at prostatic urethra | Single | Anorectal duplication | 7Cα |
| Peris | 17 yrs. | Phallus 1: 2 Phallus 2: 0 | N/A | Normal | Descended bilaterally | No | Single | Single | N/A | 2Aα |
| Priyadarshi | 1 yr. | Phallus 1: 1 Phallus 2: 1 | Epispadias | Bifid | Descended bilaterally | No | Duplicate | Single | Ectopic bowel segment | 3Dα |
| Rajarajan | 23 yrs. | Phallus 1: 2 Phallus 2: 2 | N/A | Normal | Descended bilaterally | No | Duplicate | Single | Anorectal anomalies and colonic duplication | 4Cα |
| Rao and Chandrasekharan | 3 mo. | Phallus 1: 2 | Epispadias | Normal | Descended bilaterally | No | Single | N/A | Anorectal malformation | 7Dα |
| Author, Year | Age at definitive treatment | Number of corpora cavernosa | Hypospadias/Epispadias | Scrotum | Testes | Penoscrotal Transposition | Urethra | Bladder | Other anomalies | Classification |
|--------------|-----------------------------|-----------------------------|------------------------|---------|--------|--------------------------|---------|---------|----------------|---------------|
| Remzi⁶²       | 14 yrs.                     | Phallus 1: 2, Phallus 2: 2  | Hypospadias            | Duplicate | Duplicate, descended bilaterally | No       | Duplicate | Exstrophy, lumbosacral scoliosis | 1Cy          |
| Rock and Gearhart⁶³ | 1 yr.            | Phallus 1: 2, Phallus 2: 2  | N/A                    | Bifid    | Descended bilaterally         | No       | Duplicate | Hydroplastic kidney | 1A¹          |
| Rodríguez⁶⁴   | Neonate                    | Phallus 1: 2, Phallus 2: 2  | Hypospadias Epispadias| Duplicate | Duplicate, descended bilaterally | No       | Duplicate | Single (bilobated) | 1α            |
| Rossete-Cervantes and Villegas-Muñoz⁶⁵ | 83 yrs.        | Phallus 1: 2, Phallus 2: 2  | N/A                    | Normal   | Duplicate, descended bilaterally | No       | Duplicate | Single               | 1Aα           |
| Savir et al.⁶⁶ | 31 yrs.                    | Phallus 1: 2, Phallus 2: 2  | N/A                    | Normal   | Descended bilaterally         | No       | Duplicate | Single Duplicate colon | 1Aα           |
| Sharma et al.⁶⁷ | Neonate                  | Phallus 1: 2, Phallus 2: 2  | N/A                    | Normal   | Descended bilaterally         | No       | Duplicate, one with blind ending | Single N/A | 1A                 |
| Sharma et al.⁶⁸ | Neonate                  | Phallus 1: 2, Phallus 2: 2  | N/A                    | Normal   | Descended bilaterally         | No       | Duplicate | Single               | 1Aα           |
| Sina et al.⁶⁹  | 2 mo.                      | Phallus 1: 2, Phallus 2: 1  | N/A                    | Normal with accessory scrotum | Descended bilaterally | No       | Duplicate | Single               | 2Aα           |
| Smith and Sherer⁷⁰ | 18 yrs.                  | Phallus 1: 2, Duplicate Glands Phallus 1: 2, Phallus 2: 2 | N/A | Normal | Descended unilaterally | No       | Duplicate | Exstrophy             | 7Aγ           |
| Solomon et al.⁷¹  | Neonate                   | Phallus 1: 2, Phallus 2: 2  | N/A                    | Normal   | Descended unilaterally        | No       | Duplicate | Single Supernumerary kidney | 6Aα           |
| Sotiropoulos et al.⁷² | 12 yrs.               | Phallus 1: 2, Phallus 2: 2  | N/A                    | Normal   | Descended bilaterally         | No       | Duplicate | Exstrophy Hypoplastic kidney Omphalocele Bilateral inguinal hernia Imperforate anus Colovesical fistula Agenesis left upper extremity, and a web deformity of the left popliteal region Rectoperineal fistula Vesicoureteral reflux Aplastic kidney Atrophic right kidney | 1Aγ           |
| 14 yrs. Phallus 1: 2, Duplicate Glands | Epispadias | Bifid | Descended bilaterally | No | Duplicate | Exstrophy | 7Cγ |
| Neonate       | Phallus 1: 2, Phallus 2: 2  | N/A                    | Normal   | Descended bilaterally         | No       | Duplicate | Exstrophy | 2Dγ           |
| 17 yrs. Phallus 1: 2, Phallus 2: 1 | N/A | Normal | Descended bilaterally | No | Duplicate | Duplicate | 6Aβ |
| Tepeler et al.⁷³ | 14 yrs.                  | Phallus 1: 2, Phallus 2: 1  | N/A                    | Bifid    | Descended bilaterally         | No       | Duplicate | Single               | 2Aα           |
| Tirtayasa et al.⁷⁴ | 12 yrs.               | Phallus 1: 2, Phallus 2: 2  | Epispadias             | Bifid    | Descended bilaterally         | No       | Duplicate | Single Ectopic bowel segment | 1Dα           |
| Tu et al.⁷⁵    | Neonate                   | Phallus 1: 2, Phallus 2: 2  | N/A                    | Bifid    | Descended bilaterally         | No       | Duplicate | Single               | 1Aα           |
specific pathway of the urethras or the associated anomalies, the classification and the frequency in which they occur is based on the soft tissue make-up of the phalluses, the urethra as it pertains to the

Table 1

| Author, Year | Age at definitive treatment | Number of corpora cavernosa | Penis | Testes | Scrotum | Hypospadias/Epispadias | Bladder | Urethra | Other anomalies |
|--------------|---------------------------|----------------------------|------|--------|--------|------------------------|---------|--------|----------------|
| Vilanova and Raventos | 76 yrs. | N/A | Phallic 1: 2 | Normal | Descended bilaterally | Normal | Single | N/A | Normal Urethra |
| Wojewski and Kossowski | 77 yrs. | Phallic 1: 2 | Duplicate | Duplicate, descended bilaterally | Dupicate | Dupicate | N/A | N/A | Bilateral congenital hip dislocation, severe right talipes equinovarus, hypotrophy of the right lower limb, inguinal hernia |
| Zhang et al | 78 yrs. | N/A | Phallic 1: 2 | Normal | Dupicate, one with blind ending | N/A | Single | N/A | Normal Urethra |
| Zolfaghari et al | 79 yrs. | Phallic 1: 2 | Hypospadias | Normal | Descended bilaterally | N/A | Single | Bilateral congenital hip dislocation, severe right talipes equinovarus, hypotrophy of the right lower limb, inguinal hernia |

Table 2 Proposed classifications of phallus, urethra and bladder found in Diphallia

| Character | Type |
|-----------|------|
| Separate phalluses, 3 corpora each | 1 |
| Separate phalluses, 3 corpora in only one | 2 |
| Separate phalluses, neither contain 3 corpora | 3 |
| Phalluses contained within same shaft skin, 3 corpora each | 4 |
| Phalluses contained within same shaft skin, 3 corpora in only one | 5 |
| Phalluses contained within same shaft skin, neither contain 3 corpora | 6 |
| Bifid glans | 7 |

Urethra to most normal phallus

| Character | Type |
|-----------|------|
| Normal urethra present | A |
| Urethral stenosis | B |
| Hypospadias | C |
| Epispadias | D |
| Bifurcation | E |

Bladder

| Character | Type |
|-----------|------|
| Single | α |
| Double | β |
| Exstrophy | γ |

Table 3 Proportion of diphallia anatomical variants within the literature

| Classification | Count | Percent (%) |
|----------------|-------|-------------|
| 1Aα | 23 | 26.4 |
| 1Aβ | 11 | 12.6 |
| 2Aα | 7 | 8.0 |
| 1Aγ | 4 | 4.6 |
| 3Aα | 4 | 4.6 |
| 7Aα | 4 | 4.6 |
| 1Aγ | 2 | 2.3 |
| 1Dα | 2 | 2.3 |
| 1Dγ | 2 | 2.3 |
| 3Aγ | 2 | 2.3 |
| 4Aα | 2 | 2.3 |
| 7Bα | 2 | 2.3 |
| 1Bα | 2 | 2.3 |
| 1Bβ | 2 | 2.3 |
| 1Cβ | 1 | 1.1 |
| 1Cγ | 1 | 1.1 |
| 1Eα | 1 | 1.1 |
| 2Bα | 1 | 1.1 |
| 2Dα | 1 | 1.1 |
| 2Eα | 1 | 1.1 |
| 3Aβ | 1 | 1.1 |
| 3Dα | 1 | 1.1 |
| 4Cα | 1 | 1.1 |
| 4Eα | 1 | 1.1 |
| 6Aα | 1 | 1.1 |
| 6Aβ | 1 | 1.1 |
| 6Cα | 1 | 1.1 |
| 7Aα | 1 | 1.1 |
| 7Aγ | 1 | 1.1 |
| 7Bα | 1 | 1.1 |
| 7Cγ | 1 | 1.1 |
| 7Dα | 1 | 1.1 |
| Classification | Diagram | Gross anatomy | References     |
|----------------|---------|---------------|----------------|
| 1Aα            | ![Diagram](image1) | ![Gross Anatomy](image2) | Chadha et al.¹⁸ |
| 1Aβ            | ![Diagram](image3) | ![Gross Anatomy](image4) | Maher et al.⁴⁷ |
| 2Aα            | ![Diagram](image5) | ![Gross Anatomy](image6) | Sina et al.⁶⁹  |
| 1Cα            | ![Diagram](image7) | ![Gross Anatomy](image8) | Bhat et al.¹⁵  |
| 3Aα            | ![Diagram](image9) | ![Gross Anatomy](image10) | Leite et al.⁴⁶ |
most normal urethra and the bladder configuration. Of the reviewed literature with sufficient information, our classification showed that diphallia patients categorized as 1A contributed to the largest percentage of reports at 26.4%. The next most frequent pattern seen was 1Aβ where the soft tissue and urethra were structurally normal, however there was a duplicate bladder, which made up 12.6% of the patients reviewed. The 2A configuration was the next most seen classification within the literature, appearing in 7% or 8.0%, followed by configurations 1C, 3A, 7A, which each contributed to 4.6% of the patients. The remaining reports and the corresponding classifications can be reviewed in Table 3. Table 4 then depicts the most common configurations found within the literature and additional examples of diphallia, which shows a graphic of the corresponding anatomy, an example of the gross anatomy extracted from their corresponding reference and the patient that correspond to the detailed classification.

### Associated anomalies

Upon review of the literature, several diphallia patients present with no other associated anomalies. However, several abnormalities can be seen in other diphallia patients. These anomalies arise from both genitourinary and gastrointestinal systems, as well as some reports seeing musculoskeletal or cardiovascular anomalies. The majority of the malformations fall within the former two. Genitourinary abnormalities include the presence of either a duplicate bladder or bladder extrophy, as well as duplicate ureters, vesicoureteral reflux, and ureteric stenosis. Further anomalies within the urinary system include issues and malformations pertaining to the kidneys, which include single kidney agenesis, duplicate unilateral kidney, pelvic kidney, and horseshoe kidney. Fistulas pertaining to the urinary system were also a common association with rectovesical and urethrorectal being noted. Next, there is also a wide range of gastrointestinal abnormalities that appear to arise in patients with diphallia. The most seen is the presence of an imperforate anus and terminal ileum. Along with the mentioned fistulas mentioned above, perineal and tracheoesophageal fistula have been reported as associated abnormalities pertaining to the gastrointestinal tract. It was also noted that there were hernias associated with diphallia patients, mostly those occurring at the umbilicus, however inguinal hernias are also cited within the literature. Further gastrointestinal anomalies then include ectopic bowel segments, omphalocele and Meckel diverticulum.

In terms of musculoskeletal and cardiovascular abnormalities, there is a wide range of malformations shown in the literature that

### Table 4 Continued

| Classification | Diagram | Gross anatomy | References |
|----------------|---------|---------------|------------|
| 7Aα            | ![Diagram](image1.png) | ![Gross Anatomy](image2.png) | Zhang et al.78 |

### Table 5 Proportion of associated abnormalities

| Associated Abnormality                  | Count | Percent (%) |
|----------------------------------------|-------|-------------|
| **Gastrointestinal**                   |       |             |
| Imperforate anus                       | 12    | 13.8        |
| GIT duplication                        | 8     | 9.2         |
| Anorectal malformation                 | 5     | 5.7         |
| Ectopic bowel segments                 | 2     | 2.3         |
| Omphalocele                            | 2     | 2.3         |
| Oesophageal atresia with tracheoesophageal fistula | 2 | 2.3 |
| Meckel diverticulum                    | 1     | 1.1         |
| **Genitourinary**                      |       |             |
| Single renal agenesis                  | 7     | 8.0         |
| Horseshoe kidney                       | 3     | 3.4         |
| Duplicate ureters                      | 2     | 2.3         |
| Vesicoureteral reflux                  | 2     | 2.3         |
| Pelvic kidney                          | 1     | 1.1         |
| Duplicate kidney                       | 1     | 1.1         |
| Ureteric stenosis                      | 1     | 1.1         |
| **Musculoskeletal**                    |       |             |
| Limb agenesis/hypotrophy               | 5     | 5.7         |
| Wide diastasis of pubic bone           | 4     | 4.6         |
| Hemivertebra                           | 3     | 3.4         |
| Meningocele                            | 2     | 2.3         |
| Talipes equinovarus                    | 2     | 2.3         |
| Sacral agenesis                        | 1     | 1.1         |
| Bilateral hip dislocations              | 1     | 1.1         |
| **Cardiovascular**                     |       |             |
| Atrial septal defect                   | 2     | 2.3         |
| **Hernias**                            |       |             |
| Inguinal                                | 6     | 6.9         |
| Umbilical                              | 2     | 2.3         |
are associated with diphallia. Two common associations include both hemivertebrae and a wide diastasis of the pubic bone, with further musculoskeletal malformations comprising of meningocele, and talipes equinovarus. Cardiovascular malformations are more rare, however present in multiple patients, which include abnormalities such as atrial septal defects, a summary of associated abnormalities pertaining to specific patients is outlined in Table 1.

Table 5 then displays the most commonly seen abnormalities seen in concordance with diphallia. Most commonly, an imperforate anus was seen associated with diphallia patients which was shown in 13.8% of the reviewed patients. Next, 9.2% of the patients also saw further duplication along the gastrointestinal tract, with duplications of either the rectum, colon, cecum, appendix or terminal ileum being reported. Single kidney agenesis and inguinal hernias were then the next most seen abnormalities, arising in 8.0% and 6.9% of the patients, respectively. Lastly, the most common musculoskeletal anomaly associated with diphallia was limb agenesis or hypotrophy, which appeared in 5.7% of the diphallia cases.

Discussion
The paper highlights an overview of diphallia, illustrating different aspects of the anomaly including history, embryology and treatments. It also expands to provide a classification system that is built on previous works to give a system in which future patients can be categorized and compared. The results then feature the most common anatomical variations, showing that roughly 25% of the published instances have two phalluses with 3 corpora, at least a single normal urethra and a single bladder. The literature review and the published articles however may be biased as typically the more unique and interesting patients are presented, leaving a potentially large number of unpublished reports that could contribute to the current review and proposed classification system. The results regarding associated anomalies potentially hold a similar bias, in that unique abnormalities may have been focused on, leaving out seemingly minute associations. Lastly, as new patients may be presented, and different variants may arise in which the proposed classification system may or may not encompass, or penile anomalies such as triphallia or triple penis, as reported by Jabali et al. get reported, the current proposed classification may need to be modified.

Conclusion
Following a literature review, each diphallia patient is a unique variant with its own anatomical configuration and associated anomalies. This has led to the proposed classification system that builds on previous bodies of work to categorize each patient based on the most normal aspects of the diphallia. By classifying the structure of the phallus that is to be kept, the pathway of the urethra present within the most normal phallus and the bladder morphology, a surgical approach can be broached and executed to ensure a satisfactory functional goal, with preserved continence, erectile function, and cosmetic outcomes.

Conflict of interest
The review is not registered, and the protocol was not prepared. Support from the Paediatric Surgery and Urology Department at the Queensland Children’s Hospital. None declared.

Author contributions
Dylan John Kendrick: Conceptualization; data curation; formal analysis; investigation; methodology; visualization; writing - original draft; writing - review and editing. Roy Mark Kimble: Project administration; supervision; writing - review and editing.

References
1. Fahmy M. Congenital anomalies of the penis. Springer International Publishing Switzerland, 2017. Penile Duplication, pp. 73–8.
2. Galassi FM, Henneberg M, Habicht ME, Rühl FI. Diphallia in the ancient world: insights from a depictsan fresco (70–79 AD). Urology 2016; 22: 281–2.
3. Cecil AB. Anatomy, abnormalities and injuries of the penis, Vol. 162. H. Cabot. Philadelphia: Lea & Febiger, 1936.
4. Hollowell F Jr, Witherington R, Ballagas AJ et al. Embryological consideration of diphallus and associated anomalies. J. Urol. 1977; 111: 728–32.
5. Acimi S. Complete diphallia. Scand. J. Urol. Nephrol. 2004; 38: 446–7.
6. Acimi S. Complete diphallia. Scand. J. Urol. Nephrol. 2008; 42: 389–91.
7. Adair EL, Lewis EL. Ectopic scrotum and diphallia: report of a case. J. Urol. 1960; 84: 115–7.
8. Al-Herbish A, Al-Samaari AY. Diphallus associated with a third ectopic urethra. Pediatr. Surg. Int. 1996; 11: 189–90.
9. Aihole JS, Babu N, Shankar G. Glandular diphallus with urethral duplication: conventional technique for a rare congenital anomaly. Indian J. Urol. 2015; 31: 369–71.
10. Ağül AK, Uçar M, Çelik F, Krşţoğlu I, Kılıç N. Complete penile duplication with structurally normal penises: a case report. Balkan Med. J. 2018; 35: 340–3.
11. Aleem A. Diphallia: report of a case. J. Urol. 1972; 108: 357–8.
12. Ali MT. Case report: supernumerary penis (diphallia terrata). Int. J. Collab. Res. Int. Med. Public Health 2012; 4: 502–5.
13. Arya MC, Gandhi A, Singhal A, Vasudeo V, Singh R, Sonwal M. True diphallia: a report of two cases. Asian J. Rep. Urol. 2020; 1: 1–4.
14. Bakheet MA, Refaei M. Penile duplication and two anal openings; report of a very rare case. Iran. J. Pediatr. 2012; 22: 133–6.
15. Bhat HS, Sukumar S, Nair TB, Saheed CS. Successful surgical correction of true diphallia, scrotal duplication, and associated hypospadias. J. Pediatr. Surg. 2006; 41: e13–4.
16. Blanco S. Diphallus (double penis). J. Urol. 1945; 53: 786–90.
17. Cermach MC, Hayashi H, Soares D. Diphallia associated with malformation of hindgut derivatives. Urology 1989 Mar 1; 33: 209–10.
18. Chadhra R, Bagga D, Gupta S, Mahajan JK. Complete diphallia associated with features of covered extrophy. J. Pediatr. Surg. 2001; 36: 1–3.
71. Solomon AA, Rosenthal I, Linker MH. Double penis associated with supernumerary kidney. *J. Urol.* 1950; 64: 705–9.
72. Sotiropoulos A, Uson A, Lattimer JK. Duplication of external genitalia in men. *Urology* 1974; 4: 688–92.
73. Tepeler A, Karadag MA, Ozkuvanci U, Sarı E, Berberoğlu Y, Müslümanoğlu AY. Complete diphallus in a 14 years old boy.
74. Tirtayasa PM, Prasetyo RB, Rodjani A. Diphallia with associated anomalies: a case report and literature review. *Case Rep. Urol.* 2013; 8: 2013–4.
75. Tu YA, Su YN, Yang PK, Shih JC. Prenatal diagnosis of true diphallia and associated anomalies. *Obstetr. Gynecol.* 2014; 124: 416–8.
76. Vilanova, X, Ra Ventos A. Pseudodiphallia: a rare anomaly. *J. Urol.* 1954. 71, 338, 346.
77. Wojewski A, Kossowski W. Total diphallia: a case of plastic repair. *J. Urol.* 1964; 91: 84–6.
78. Zhang W, Yu N, Liu Z, Wang X. Pseudodiphallia: a rare kind of diphallia: a case report and literature review. *Medicine* 2020; 99: e21638.
79. Zolfaghari A, Pourissa M, Hajialilou S, Amjadi M. True complete Diphallia: case report. *Scand. J. Urol. Nephrol.* 1995; 29: 233–5.
80. Schneider P, Lamitter JK, Uson AC, Melicow AC. The male genital tract. In: Mustard WT, Ravitch MM, Snyder WH et al. (eds). *Pediatric Surgery*, 2nd edn. Chicago: Year Book Medical Publishers, 1969; 1263.
81. Jabali SS, Mohammed AA. Triphallia (triple penis), the first reported case in human. *Int. J. Surg. Case Rep.* 2020; 1: 198–200.