Diphallia: literature review and proposed surgical classification system

REVIEW ARTICLE

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Diphallia: literature review and proposed surgical classification system

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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 base 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.[1] 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).[2]

Embryology

Previously, Cecil[3] 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.

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Fig. 1. Pompeian Lupanar showing Priapus with a double phallus. Image from Galassi *et al.* accessed January third, 2021.

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]

Treatment

Treatment of diphallia is typically approached on an individual basis, as no two patients within the literature are the same. The associated anomalies are a major cause of mortality in diphallia patients, and treatment is usually done in a stepwise manor, with corrections aimed at the anomalies, excision of the more abnormal phallus, excision of the urethra in instances of duplicate urethra, and excision or correction of either duplicate bladders or bladder exstrophy.[1] It may also be difficult to discern which phallus to excise, thus with the proposed classification system, a label can be used to drive the treatment approach, allowing a decision to be made based on the more normal phallus, with the more normal urethral pathway and normal bladder configuration.

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, psuedophallia 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

Schneider[80] 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) Pseudodiphalia 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

Table 1 Literature review of diphallia, associated anatomy and classifications

Author, Year	Age at definitive treatment	Number of corpora cavernosa	Hypospadias/ Epispadias	Scrotum	Testes	Penoscrotal Transposition	Urethra	Bladder	Other anomalies	Classification
Acimi[5]	3 yrs.	Phallus 1: 2 Phallus 2: 2	Hypospadias	Normal	Descended bilaterally	No	Duplicate	Single	Imperforate anus	1Aα
Acimi[6]	4 mo.	Phallus 1: 2Phallus 2: 2	Hypospadias	Bifid	Descended bilaterally	No	Duplicate, one functional	Single	Unilateral Kidney agenesis Lumbosacral meningocele Ureterovesical duplication	1Bα
Adair and Lewis[7]	1 yr.	Phallus 1: 2 Duplicate Glans	N/A	Normal	Descended unilaterally	No	Single	Single	Umbilical hernia Unilateral Kidney agenesis	7Aα
Al-Herbish and Al-Samarrai[8]	1 mo.	Phallus 1: 2 Phallus 2: 2	Hypospadias	Bifid	Descended bilaterally	No	Duplicate with bifurcation of one urethra	Single	Unilateral Kidney agenesis Musculoskeletal anomalies Hemivertebra and absent first rib	1Aα
Aihole, 2015[9]	1 yr.	Phallus 1: 2 Duplicate Glans	N/A	Normal	Descended bilaterally	No	Duplicate	Single	Pre-axial polydactyly Solitary Kidney Atrial septal defect	7Bα
Akgül *et al.*[10]	4 yrs.	Phallus 1: 2 Phallus 2: 2	N/A	Normal	Descended bilaterally	No	Duplicate	Duplicate	Anal atresia Duplicate rectum, colon, cecum, appendix and terminal ileum Rectovesical fistula	1Aβ
Aleem[11]	2 mo.	Phallus 1: 1 Phallus 2: 1	Epispadias	Bifid	Descended bilaterally	No	Single	Single	Wide diastasis of the pubic bones and partial sacral agenesis.	1Dα
Ali[12]	19 yrs.	Phallus 1: 1 Phallus 2: 2	N/A	Normal	Descended bilaterally	No	Single	Single	N/A	2Aα
Arya *et al.*[13]	31 yrs.	Phallus 1: 2 Phallus 2: 2	Epispadias	Bifid	Descended bilaterally	No	Duplicate, no urethral plates	Exstrophy	N/A	1Dγ
	3 mo.	Phallus 1: 2 Phallus 2: 2	N/A	Bifid	Descended bilaterally	No	Duplicate	Single	N/A	1Aα
Bakheet and Refaei[14]	Neonate	Phallus 1: 2 Phallus 2: 2	N/A	Duplicate	Duplicate, descended bilaterally	No	Duplicate	Single	Duplicate colon, rectum, anus	1Aα
Bhat *et al.*[15]	1 yr.	Phallus 1: 2 Phallus 2: 2	Hypospadias	Bifid	Descended bilaterally	No	Duplicate, opening normally and into lateral wall of bladder	Single	N/A	1Cα
Blanco[16]	18 yrs.	Phallus 1: 2 Phallus 2: 2	Hypospadias	Normal	Descended bilaterally	No	Duplicate	Single	N/A	1Aα
Cernach *et al.*[17]	11 mo.	Phallus 1: 2 Phallus 2: 2	N/A	Bifid	Descended bilaterally	No	Duplicate	Duplicate	Duplicate anus, one imperforatehemivertebrae and diastasis of pubic symphysis	1Aβ
Chadha *et al.*[18]	Neonate	Phallus 1: 2 Phallus 2: 2	N/A	Bifid	Descended into lateral compartments	No	Bifurcation at prostatic urethra	Single	N/A	1Aα
de Oliveira *et al.*[19]	5 yrs.	Phallus 1: 2 Phallus 2: 2	N/A	Normal	Descended bilaterally	No	Duplicate, unilateral stenosis	Single	N/A	1Bα
Deshpande[20]	2 yrs.	Phallus 1: 2	N/A	Bifid		No	Duplicate	Single	N/A	1Aα

Table 1 Continued

Author, Year	Age at definitive treatment	Number of corpora cavernosa	Hypospadias/ Epispadias	Scrotum	Testes	Penoscrotal Transposition	Urethra	Bladder	Other anomalies	Classification
		Phallus 2: 2			Descended unilaterally					
Dewan *et al.*[21]	7 yrs.	Phallus 1: 2 Phallus 2: 2	N/A	Bifid	Descended unilaterally	No	Duplicate, opening normally and into lateral wall of bladder	Single	Imperforate anus	1Aα
Djordjevic and Perovic[22]	15 mo.	Phallus 1: 2 Phallus 2: 2	Hypospadias	Bifid	Descended bilaterally	No	Duplicate	Duplicate	Symphyseal diastasis	1Aβ
Dunn *et al.*[23]	3 yrs.	Phallus 1: 2 Phallus 2: 2	N/A	Bifid	Descended bilaterally	No	Duplicate	Duplicate	N/A	1Aβ
Dutta *et al.*[24]	Neonate	Phallus 1: 2 Phallus 2: 2	N/A	Bifid	Descended bilaterally	No	Duplicate, pneumaturia	Duplicate	Pelvic kidney Anorectal atresia	1Aβ
Elsawy *et al.*[25]	1 mo.	Phallus 1: 2 Phallus 2: 2	N/A	Normal	Descended bilaterally	No	Duplicate, one hypoplastic	Single	Inguinal hernia Absent right thumb	4Aα
Frollo *et al.*[26]	84 yrs.	Phallus 1: 2 Phallus 2: 2	Hypospadias	Normal	Descended bilaterally	No	No urethra in either phallus	Single	N/A	1Cα
Gavali *et al.*[27]	5 yrs.	Phallus 1: 2 Duplicate Glans	Hypospadias	Normal	Descended bilaterally	No	Single	Single	Single Kidney agenesis	7Bα
Ghafoori *et al.*[28]	5 yrs.	Phallus 1: 1 Phallus 2: 1	N/A	Normal	Descended bilaterally	No	Duplicate	Single	N/A	3Aα
Goad *et al.*[29]	13 yrs.	Phallus 1: 2 Phallus 2: 2	Hypospadias	Normal	Descended bilaterally	No	Single	Single	N/A	1Aα
Gupta and Virdi[30]	10 yrs.	Phallus 1: 2 Phallus 2: 2	N/A	Normal	Descended bilaterally	No	Duplicate	Single	N/A	1Aα
Gyftopoulos *et al.*[31]	Neonate	Phallus 1: 2 Phallus 2: 2	Hypospadias	Bifid	Undescended bilaterally	No	Duplicate	Duplicate	Horseshoe kidney Partial duplication of distal colonVentricular septum defect Hypoplasia of the right leg due to agenesis of the fibula Cloacal opening at the perineum	1Aβ
	Neonate	Phallus 1: 2 Phallus 2: 2	N/A	Normal	Descended bilaterally	No	Duplicate, one with blind ending	Duplicate	Urethrorectal Y-fistula	3Aβ
Hanine *et al.*[32]	8 yrs.	Phallus 1: 1 Phallus 2: 1	N/A	Normal	Descended bilaterally	No	Single	Single	N/A	2Aα
Hollowell *et al.*[13]	5 mo.	Phallus 1: 2 Phallus 2: 2	Hypospadias	Bifid	Descended bilaterally	No	Duplicate	Duplicate	Imperforate anus Perineal fistula Meningocele	1Aα
	50 yrs.	Phallus 1: 2 Phallus 2: 2	Hypospadias	Bifid	Descended into lateral compartments	No	Duplicate	Single	Prolapse of the rectum Absence of perineal Midline infraumbilical musculature Bilateral inguinal hernias	1Cα
Jesus *et al.*[33]	20 yrs.	Phallus 1: 2 Duplicate Glans	Hypospadias	Normal	Descended bilaterally	No	Duplicate	Single	N/A	7Cα

Table 1 Continued

Author, Year	Age at definitive treatment	Number of corpora cavernosa	Hypospadias/ Epispadias	Scrotum	Testes	Penoscrotal Transposition	Urethra	Bladder	Other anomalies	Classification
Johnson *et al.*[34]	17 yrs.	Phallus 1: 2 Duplicate Glans	N/A	Normal	Descended bilaterally	No	Duplicate	Single	N/A	7Aα
Karagöz *et al.*[35]	15 yrs.	Phallus 1: 2 Duplicate Glans	N/A	Normal	Descended bilaterally	No	Single	Single	N/A	7Aα
Kardasevic *et al.*[36]	Neonate	Phallus 1: 2 Phallus 2: 2	Hypospadias	#NAME?	Descended bilaterally	No	Duplicate	Duplicate	Atrial and ventricular septal defect Duplicate right ureter Triple spleen Duplicate right kidney	1Aβ
Karna and Kapur[37]	Neonate	Phallus 1: 2 Phallus 2: 2	N/A	Bifid	Undescended bilaterally	No	Duplicate	Single	Omphalocele Tracheoesophageal fistula Imperforate anus Single Kidney agenesis Duplicate distal ileum, cecum, appendix and colon Musculoskeletal anomalies	1Aα
Kaufman *et al.*[38]	15 yrs.	Phallus 1: 2 Phallus 2: 2	Hypospadias	Normal	Descended bilaterally	No	Duplicate, with third perineal urethra	Single	Imperforate anus	1Aα
Keckler[39]	Neonate	Phallus 1: 2 Phallus 2: 2	N/A	Normal	Descended bilaterally	No	Duplicate	Single	N/A	1Aα
Kendrick *et al.*[40]	3 mo.	Phallus 1: 2 Phallus 2: 1	N/A	Normal	Descended bilaterally	Partial	Duplicate, opening normally and into lateral wall of bladder	Single	N/A	2Bα
Khorramirouz *et al.*[41]	6 yrs.	Phallus 1: 2 Duplicate Glans	N/A	Normal	Descended bilaterally	No	Duplicate, unilateral stenosis	Duplicate	Horseshoe Kidney	7Bβ
Kırlı *et al.*[42]	Neonate	Phallus 1: 2 Phallus 2: 2	Epispadias	Normal	Descended bilaterally	No	Duplicate	Exstrophy	Inguinal hernia	1Dγ
Kundal *et al.*[43]	3 yrs.	Phallus 1: 2 Phallus 2: 2	Epispadias	Normal	Descended bilaterally	No	Duplicate	Single	N/A	1Aα
Landy *et al.*[44]	Neonate	Phallus 1: 2 Phallus 2: 2	N/A	Normal with accessory scrotum	Descended bilaterally	No	Duplicate	Single	Imperforate anus	1Aα
Larsen[45]	14 yrs.	Phallus 1: 1 Phallus 2: 1	N/A	Normal	Descended unilaterally	Partial	Duplicate	Single	Atrophic leg	3Aα
Leite *et al.*[46]	2 yrs.	Phallus 1: 1 Phallus 2: 1	N/A	Normal	Descended bilaterally	No	Duplicate, one with blind ending	Single	Short oesophagus	3Aα
Maher *et al.*[47]	Neonate	Phallus 1: 2 Phallus 2: 2	N/A	Duplicate	Descended bilaterally	No	Duplicate	Duplicate	Separated natal clefts with no anal orifice Hydronephrotic left kidney Duplicated colon Caudal duplication syndrome	1Aβ
Mandal and Sahi[48]	Neonate	Phallus 1: 2	N/A	Duplicate		No	Duplicate	Single	N/A	1Aα

Table 1 Continued

Author, Year	Age at definitive treatment	Number of corpora cavernosa	Hypospadias/ Epispadias	Scrotum	Testes	Penoscrotal Transposition	Urethra	Bladder	Other anomalies	Classification
		Phallus 2: 2			Duplicate, descended bilaterally					
Marti-Bonmati *et al.*[49]	Neonate	Phallus 1: 2 Phallus 2: 2	Hypospadias	Ectopic scrotal tissue	Descended unilaterally	No	Duplicate, one with blind ending	Exstrophy	Imperforate anus	1Aγ
Maruyama *et al.*[50]	Neonate	Phallus 1: 1 Phallus 2: 1	Hypospadias	Bifid	Descended unilaterally	No	Duplicate	Single	Tracheoesophageal fistula with oesophageal atresia Imperforate anus Bilateral radial limb defects Cleft palate Patent ductus arteriosus, Single umbilical artery Right megaureterLeft multicystic dysplastic kidney	3Cα
Matsumoto *et al.*[51]	12 mo.	Phallus 1: 1 Phallus 2: 2	Hypospadias	Bifid	Descended bilaterally	Complete	Duplicate	Single	Atrial septal defect	2Aα
Melekos *et al.*[52]	8 yrs.	Phallus 1: 1 Phallus 2: 1	Hypospadias	Normal	Descended bilaterally	No	Duplicate	Single	Horseshoe Kidney	6Cα
Mingazzini[53]	36 yrs.	Phallus 1: 1 Phallus 2: 1	N/A	Normal	Descended bilaterally	No	Duplicate, unilateral stenosis	Single	Umbilical hernia	3Aα
Mirshemirani *et al.*[54]	Neonate	Phallus 1: 2 Phallus 2: 2	Hypospadias	Bifid	Descended bilaterally	No	Duplicate	Duplicate	Imperforate anus Duplicate colon	1Cβ
	14 yrs.	Phallus 1: 1 Phallus 2: 1	Hypospadias	Bifid	Descended bilaterally	No	Duplicate	Single	Imperforate anus Duplicate sigmoid colon	3Cα
	4 yrs.	Phallus 1: 2 Phallus 2: 2	N/A	Normal	Descended bilaterally	No	Duplicate	Duplicate	Right inguinal hernia	1Aβ
	12 yrs.	Phallus 1: 2 Phallus 2: 2	Epispadias	Bifid	Descended bilaterally	No	Duplicate	Exstrophy	Single kidney Hemi-Vertebra	4Dγ
	1 yr.	Phallus 1: 2 Phallus 2: 2	N/A	Bifid	Descended bilaterally	No	Duplicate	Duplicate	N/A	1Aβ
	9 mo.	Phallus 1: 2 Phallus 2: 2	N/A	Bifid	Descended bilaterally	No	Duplicate	Single	Imperforate anus	1Aα
Mukunda *et al.*[55]	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.*[56]	9 yrs.	Phallus 1: 2 Phallus 2: 1	N/A	Normal	Descended bilaterally	No	Bifurcation at prostatic urethra	Single	Rotational anomaly of right kidney Left ureter duplication	2Eα
Nunez *et al.*[57]	Neonate	Phallus 1: 2 Duplicate Glans	Hypospadias	Normal	Descended bilaterally	No	Duplicate	Single	Anorectal malformation	7Cα
Peris[58]	17 yrs.	Phallus 1: 2 Phallus 2: 0	N/A	Normal	Descended bilaterally	No	Single	Single	N/A	2Aα
Priyadarshi[59]	1 yr.	Phallus 1: 1 Phallus 2: 1	Epispadias	Bifid	Descended bilaterally	No	Duplicate	Single	Ectopic bowel segment	3Dα
Rajarajan[60]	23 yrs.	Phallus 1: 2 Phallus 2: 2	N/A	Normal	Descended bilaterally	No	Duplicate	Single	Anorectal anomalies and colonic duplication	4Cα
Rao and Chandrasekharam[61]	3 mo.	Phallus 1: 2 Duplicate Glans	Epispadias	Normal	Descended bilaterally	No		Single	N/A	7Dα

Table 1 Continued

Author, Year	Age at definitive treatment	Number of corpora cavernosa	Hypospadias/ Epispadias	Scrotum	Testes	Penoscrotal Transposition	Urethra	Bladder	Other anomalies	Classification
							Duplicate, one with blind ending			
Remzi[62]	14 yrs.	Phallus 1: 2 Phallus 2: 2	Hypospadias	Duplicate	Duplicate, descended bilaterally	No	Duplicate	Exstrophy	hemivertebrae, lumbosacral scoliosis	1Cγ
Rock and Gearheart[63]	1 yr.	Phallus 1: 2 Phallus 2: 2	N/A	Bifid	Descended bilaterally	No	Duplicate	Duplicate	Hypoplastic kidney Widely separate pubic diastasis Sacral insufficiency	1Aβ
Rodriguez[64]	Neonate	Phallus 1: 2 Phallus 2: 2	HypospadiasEpispadias	Duplicate	Duplicate, descended bilaterally	No	Duplicate	Single (bilobated)	Duplicate umbilical cord	1Cα
Rossete-Cervantes and Villegas-Muñoz[65]	83 yrs.	Phallus 1: 2 Phallus 2: 2	N/A	Normal	Duplicate, descended bilaterally	No	Duplicate	Single	N/A	1Aα
Savir *et al.*[66]	31 yrs.	Phallus 1: 2 Phallus 2: 2	N/A	Normal	Descended bilaterally	No	Duplicate	Single	N/A	4Aα
Sharma *et al.*[67]	Neonate	Phallus 1: 2 Phallus 2: 2	N/A	Normal	Descended bilaterally	No	Duplicate, one with blind ending	Single	Duplicate colon	1Aα
Sharma *et al.*[68]	Neonate	Phallus 1: 2Phallus 2: 2	N/A	Normal	Descended bilaterally	No	Duplicate, one with blind ending	Single	N/A	1Aα
Sina *et al.*[69]	2 mo.	Phallus 1: 2 Phallus 2: 1	N/A	Normal with accessory scrotum	Descended bilaterally	No	Duplicate	Single	N/A	2Aα
Smith and Sherer[70]	18 yrs.	Phallus 1: 2 Duplicate Glans	N/A	Normal	Descended unilaterally	No	Duplicate	Exstrophy	N/A	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	Hypospadias	Bifid	Descended bilaterally	No	Duplicate, both blind ending	Exstrophy	Hypoplastic kidney Omphalocele Bilateral inguinal hernia Imperforate anus Colovesical fistula	1Aγ
	14 yrs.	Phallus 1: 2 Duplicate Glans	Epispadias	Bifid	Descended bilaterally	No	Duplicate	Exstrophy	Agenesis left upper extremity, and a web deformity of the left popliteal region. Rectoperineal fistula	7Cγ
	Neonate	Phallus 1: 2 Phallus 2: 2	N/A	Normal	Descended bilaterally	No	Duplicate	Exstrophy	Vesicoureteral reflux Aplastic kidney	2Dγ
	17 yrs.	Phallus 1: 1 Phallus 2: 1	N/A	Normal	Descended bilaterally	No	Duplicate	Duplicate	Atrophic right kidney	6Aβ
Tepeler *et al.*[73]	14 yrs.	Phallus 1: 2 Phallus 2: 1	N/A	Bifid	Descended bilaterally	No	Duplicate	Single	N/A	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	N/A	1Aα

Table 1 Continued

Author, Year	Age at definitive treatment	Number of corpora cavernosa	Hypospadias/ Epispadias	Scrotum	Testes	Penoscrotal Transposition	Urethra	Bladder	Other anomalies	Classification
Vilanova and Raventos[76]	30 yrs.	Phallus 1: 2 Phallus 2: 1	N/A	Normal	Descended bilaterally	No	Single	Single	N/A	2Aα
Wojewski and Kossowski[77]	6 yrs.	Phallus 1: 2 Phallus 2: 2	N/A	Duplicate	Duplicate, descended bilaterally	No	Duplicate	Duplicate	Talipes equinovarus Ureter stenosis	1Aβ
Zhang *et al.*[78]	23 yrs.	Phallus 1: 2 Duplicate Glans	N/A	Normal	Duplicate, descended bilaterally	No	Single	Single	N/A	7Aα
Zolfaghari *et al.*[79]	4 mo.	Phallus 1: 2 Phallus 2: 2	Hypospadias	Normal	Descended bilaterally	No	Duplicate, one with blind ending	Single	Bilateral congenital hip dislocation, severe right talipes equinovarus and hypotrophy of the right lower limb. Inguinal hernia	1Aα

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

Phallus	
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
1Cα	4	4.6
3Aα	4	4.6
7Aα	4	4.6
1Aγ	2	2.3
1Dα	2	2.3
1Dγ	2	2.3
3Cα	2	2.3
4Aα	2	2.3
7Bα	2	2.3
7Cα	2	2.3
1Bα	2	2.3
1Cβ	1	1.1
1Cγ	1	1.1
1Eα	1	1.1
2Bα	1	1.1
2Dγ	1	1.1
2Eα	1	1.1
3Aβ	1	1.1
3Dα	1	1.1
4Cα	1	1.1
4Dγ	1	1.1
6Aα	1	1.1
6Aβ	1	1.1
6Cα	1	1.1
7Aα	1	1.1
7Aγ	1	1.1
7Bβ	1	1.1
7Cγ	1	1.1
7Dα	1	1.1

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 4 Most common anatomical varriants for diphallia

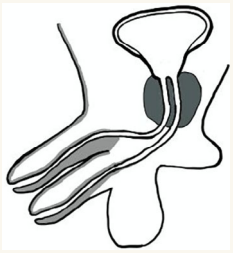
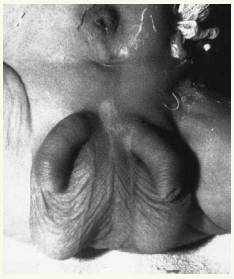
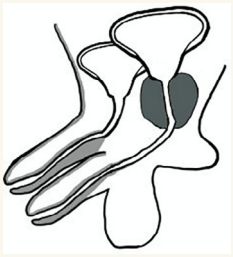
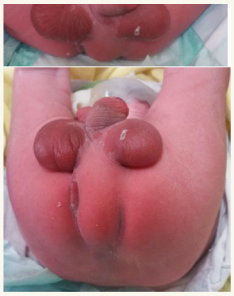
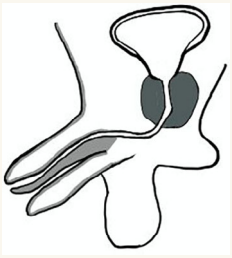
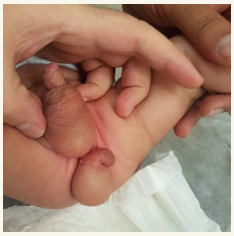
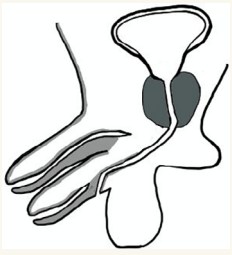
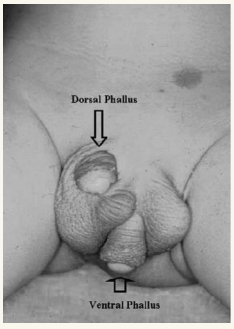
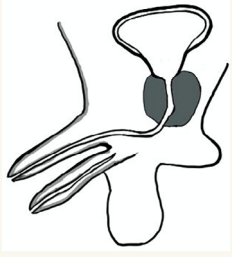
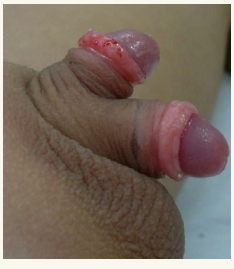
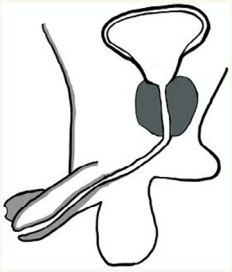
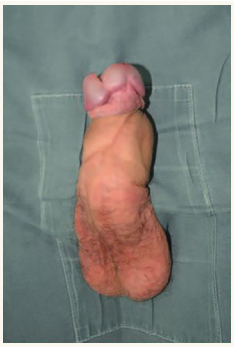
Classification	Diagram	Gross anatomy	References
1Aα			Chadha *et al.*[18]
1Aβ			Maher *et al.*[47]
2Aα			Sina *et al.*[69]
1Cα			Bhat *et al.*[15]
3Aα			Leite *et al.*[46]

Table 4 Continued

Classification	Diagram	Gross anatomy	References
7Aα			Zhang *et al.*[78]

most normal urethra and the bladder configuration. Of the reviewed literature with sufficient information, our classification showed that diphallia patients categorized as 1Aα 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 2Aα configuration was the next most seen classification within the literature, appearing in 7% or 8.0%, followed by configurations 1Cα, 3Aα, 7Aα, 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[10,13,17,22–24,31,36,41,47,54,63,72] or bladder exstrophy,[13,42,49,62,70,72,77] as well as duplicate ureters,[36,56] vesicoureteral reflux,[47,72] and ureteric stenosis.[77] Further anomalies within the urinary system include issues and malformations pertaining to the kidneys, which include single kidney agenesis,[7–9,37,54,63,72] duplicate unilateral kidney,[36] pelvic kidney[24] and horseshoe kidney.[31,41,52] Fistulas pertaining to the urinary system were also a common association with rectovesical and urethrorectal[10,31,72] 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.[5,13,17,24,37,38,44,47,49,50,54,72] There can also be further duplication of various aspects of the gastrointestinal system associated with diphallia, which includes duplication of the rectum, colon, cecum, appendix, and terminal ileum.[10,14,31,37,47,54,60,67] Along with the mentioned fistulas mentioned above, perineal[13] and tracheoesophageal fistula[37,50] 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,[7,25,41,53] however inguinal hernias[54,72,79] are also cited within the literature. Further gastrointestinal anomalies then include ectopic bowel segments,[59,74] omphalocele[37,72] and Meckel diverticulum.[55]

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

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
Muskuloskeletal		
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 hemivertebra[8,54,62] and a wide diastasis of the pubic bone[11,17,22,63] with further musculoskeletal malformations comprising of menigocele[6,13] and talipes equinovarus[77,79] and less commonly pre-axial polydactyly,[9] partial sacral agenesis,[11] agenesis and hypotrophy of digits or limbs,[25,31,45,72,79] or bilateral congenital hip dislocations.[79] Cardiovascular malformations are more rare, however present in multiple patients, which include abnormalities such as atrial septal defects.[9,51] 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*.[81] 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.

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