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DOI: https://doi.org/10.1016/j.epsc.2021.101814

Journal of Pediatric Surgery Case Reports 67 (2021) 101814

Contents lists available at ScienceDirect

Journal of Pediatric Surgery Case Reports

journal homepage: http://www.elsevier.com/locate/epsc

Anatomical features and challenges in separation and reconstruction of heteropagus twins

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ARTICLE INFO

Keywords:
Heteropagus twins
Parasitic twins
Asymmetrical conjoined twins
Surgical separation
Reconstructive surgical procedure

ABSTRACT

Heteropagus twins are an extremely rare condition and the term describes asymmetrical conjoined twins in which one twin ("autosite") has a mostly intact body and the other twin ("parasite") remains anatomically incomplete but is attached to and nourished by the other one. Autosites may show severe malformations themselves that are not associated with the twins' fusion. Beyond the challenge of surgical separation further complex reconstructions may be necessary. In this case series we want to demonstrate that despite complex anatomic features, good functional (e.g. gastrointestinal, genitourinary and motoric) and aesthetic results can be achieved.

1. Introduction

Heteropagus twins are asymmetrical conjoined monochorionic-monoamniotic twins often named "parasitic twins". They have an estimated incidence of less than one per one million live births [1] and represent only 1 to 2% of all conjoined twins [2]. The term "heteropagus" describes a situation in which one of the twins has a mostly intact body that is able to survive and is referred to as "autosite", whilst the counterpart twin, referred to as "parasite", is only rudimentarily developed, physically attached to and nourished by the autosite. The fusion point of heteropagus twins appears to be always in one of the same sites as of the more common symmetrical or "complete" conjoined twins [3,4]. Different variants are used to describe the location of the attachment, with the most frequently observed variant being the epigastric heteropagus, also called omphalopagus. Other less common presentations include ischiopagus, rachipagus and craniopagus [1]. The parasite can also be completely located within the body of the autosite, which is known as fetus-in-fetu syndrome [3,5]. Depending on the location and extent of the fusion, autosites may show severe anatomical abnormalities themselves and they often have additional malformations not associated with this fusion, such as heart defects or omphaloceles [3,4].

Whilst it was rarely possible to offer satisfactory surgical treatment options for seriously affected children in the past, good functional and aesthetic results can be achieved today. As a complex and rare condition, surgical approaches continue to be challenging and a multidisciplinary team is mandatory for planning and performing the demanding separation and reconstruction procedures [6]. After successful separation, patients may experience ongoing health impairments affecting the gastrointestinal tract or genitourinary system as well as orthopaedic problems. They require close follow-up and further surgical interventions are frequently necessary [7].

We hereby present three cases of heteropagus twins (two ischiopagus twins and one omphalopagus twin) that were treated by a

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https://doi.org/10.1016/j.epsc.2021.101814
Received 20 January 2021; Accepted 7 February 2021
Available online 14 February 2021

multidisciplinary team in a single centre and highlight the anatomical features of heteropagus twins as well as challenges encountered in gastrointestinal, genitourinary and orthopaedic separation and reconstruction. We also present the further clinical course and changes in quality of life for these patients.

2. Case 1

This male infant was born at 37 6/7 weeks of gestation by caesarean section. Prenatal imaging revealed an omphalocele. In addition, physical examination after birth showed a thoracoabdominal mass with rudimentary limbs and male genitalia, which was deemed to be an omphalopagus (Fig. 1).

Meconium was passed on time and micturition was normal. Furthermore, the baby showed normal spontaneous movements of all limbs, except for the rudimentarily developed parasitic limbs. Postnatal imaging demonstrated herniation of the left liver lobe, gallbladder and some intestinal loops of the autosite into the omphalocele. The parasite was proposed to consist of soft tissue and neither thoracic and visceral organs nor bone tissue could be found in imaging. Connections to the liver and pericardium of the autosite were suspected and a branch of the internal thoracic artery provided vascular supply to the parasite. Echocardiography showed an atrial aneurysm. Otherwise no further malformations were detected. Total resection of the parasite, reduction of the omphalocele as well as closure of the abdominal and thoracic wall by primary skin closure were performed on the first day of life without any complications. Intraoperatively, shared organs and organic connections of the heteropagus twins could be ruled out. However, part of the small intestine from the parasite was found in a separate compartment of the omphalocele. This bowel was resected. Later histological work-up revealed that the parasite did not only consist of soft tissue, but also of differentiated intestine and adrenal glands, rudimentary gonadal tissue, a rudimentary pelvis and rudimentary limbs without bony structures (Illustration 1).

The postoperative course was uneventful and the patient continues to have a normal development without any health impairments so far (actual age: seven years).

3. Case 2

This male infant was born at 37 2/7 weeks of gestation by caesarean section. Prenatal imaging revealed polymelia with four legs. Physical examination after birth showed a small omphalocele, an ischiopagus with four legs (one hypoplastic ventral pair of legs without any spontaneous movements and one normally developed dorsal pair of legs with spontaneous movements) as well as a pes equinovarus of the right foot and an imperforate anus. There were two sets of male genitalia symmetrically located between the twins' legs each with a hypoplastic empty medial scrotum, a normally developed lateral scrotum with palpable testis as well as a normal-sized penis on each side (Fig. 2). Micturition was regularly observed from both urethrae. Meconium was not passed due to the imperforated anus.

In addition to two normal kidneys located at the normal position, postnatal imaging (abdominal and pelvic MRI, voiding cystourethrogram) as well as cystourethroscopy and laparoscopy showed a third pelvic kidney with one ureter within the autosite which was presumed to belong to the parasite. This kidney along with the other two kidneys and ureters from the autosite drained urine into one common urinary bladder with two bladder necks and two urethrae. The contrast study revealed a normally configured bladder, no vesicoureteral reflux and spontaneous micturition from both urethrae, the right one being stronger than the left one. The content of the omphalocele consisted of intestinal loops belonging to the parasite with no connection to the autosite's intestine. The anorectal malformation was an imperforate anus without fistula. Additionally, fusion of two pelvises and bilateral hip dysplasia with a more severely affected right side were diagnosed. A branch of the left internal iliac artery provided vascular supply to the parasite (MRI 1; Illustration 2).

The first surgery was performed on the second day of life with resection of the omphalocele and parasitic intestine laying in a separate peritoneal cavity, closure of the abdominal wall defect and placement of a dismembered colostomy. Heteropagus separation took place in a second surgical intervention at the age of twelve months. The parasitic (hypoplastic) legs, pelvis, kidney, ureter and the two hypoplastic gonads were removed. Furthermore, the two external genitalia were ventrally approximated and a bilateral modified Salter osteotomy for pubic bone approximation was performed. Later histological work-up confirmed that the hypoplastic gonads contained testicular remnants.

At the age of 21 months, a pelvic magnetic resonance imaging was performed, showing a well-developed pelvic floor and the existence of sphincter muscles. The anorectal malformation was repaired by a laparoscopically assisted anorectal pull-through combined with an additional perineal access at the age of 25 months. The dismembered colostomy was taken down six weeks later. The postoperative course was complicated by a rectal stenosis that required continuous rectal dilatations for six months. Additionally, faecal continence could not be achieved. A caecostomy catheter was placed for the appliance of anterograde enemas at the age of three years and hence social faecal continence was achieved.

At the age of four years, the external genitalia were reconstructed by fusing the solitary corpus cavernosum and urethra from each penis in the

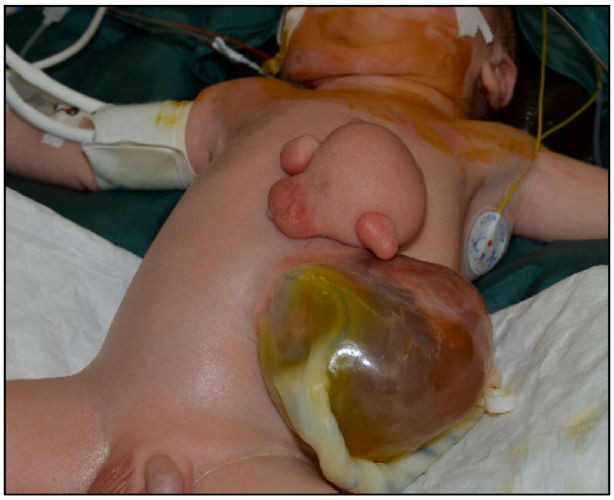


Fig. 1. Case 1 after birth showing omphalopagus and ompahlocele.

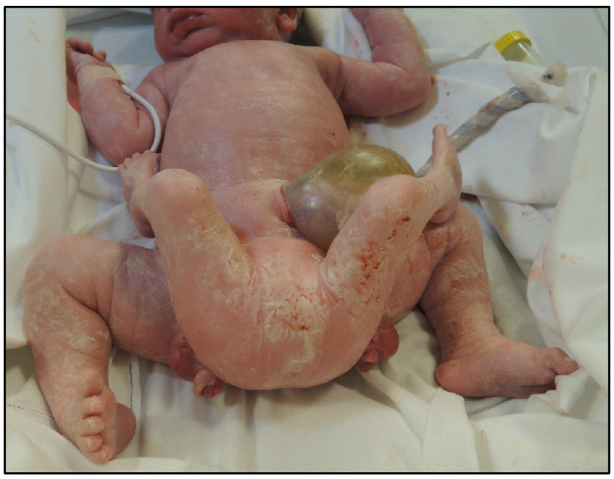


Fig. 2. Case 2 after birth showing omphalocele and ischiopagus.

midline and by reconstruction of the penile shaft skin (same session as the second osteotomy, see below). The anastomosis of the urethrae and the glans was not performed at this time because of the lengthy duration of the operation. Postoperatively, the cosmetic aspect of the external genitalia was significantly improved with the impression of a single penis in the midline. Micturition continued to be from both urethrae and the patient became continent. However, the external genitalia's position is still too perineal and a ventral translocation of the penis root together with the completion of the genital reconstruction is planned in the future.

Satisfactory clubfoot treatment results have been obtained by serial casting. Due to symphysis diastasis, a second bilateral modified Salter osteotomy and approximation of the symphysis pubis were performed at the age of four years (same session as the first step of the external genitalia reconstruction, see above). The postoperative course was complicated by right-sided sciatic nerve palsy, especially of the peroneal component, with dorsal extension deficit of the right foot. Adjustment of a drop foot orthesis was necessary. After six months, first signs of reinnervation of the peroneal nerve were observed which progressed to an almost complete reinnervation by 12 months. The patient was able to take his first steps at the age of 20 months. At present, at the age of seven years, he can walk without support and shows a fluent gait pattern. Radiological investigations demonstrate sufficient acetabular coverage of the femoral head on both sides but in combination with the ventral translocation of the penis root another pelvic osteotomy may be necessary.

4. Case 3

This female infant with complex malformations of her lower body was referred to our hospital for reconstructive surgeries at the age of 16 months. The malformations included an ischiopagus with four legs, two external female genitalia with two vaginal openings and an imperforate anus with two perineal fistulas (Fig. 3).

Fig. 3. Case 3 at admission showing ischiopagus.

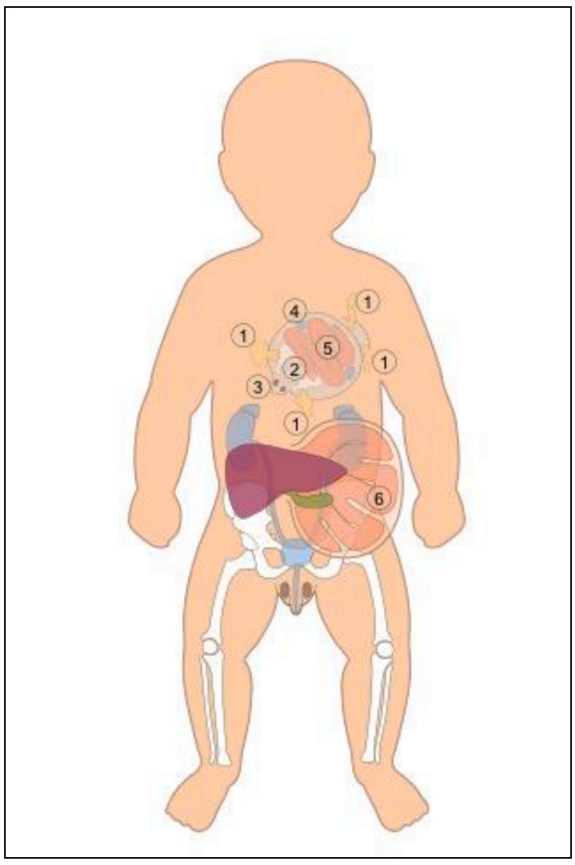


Illustration 1. (Case 1) Omphalopagus twins with ① rudimentary limbs, ② rudimentary pelvis, ③ rudimentary male genitalia, ④ differentiated adrenal glands, ⑤ differentiated intestine, ⑥ omphalocele with herniation of the left liver lobe and gallbladder oft the autosite, intestinal loops of the autosite and parasite.

The pregnancy had been unsupervised and the girl was born at term by normal spontaneous vaginal delivery. At admission, the patient was not able to stand up without support probably due to the weight of the parasite. Additionally, she experienced faecal incontinence with permanent defecations via the two perineal fistulas. Micturition was regular but urinary continence could not be assessed due to the patient's age. Recurrent urinary tract infections were not reported before admission, however one episode of pyelonephritis was diagnosed during the hospital stay. Detailed anatomical features were assessed by abdominal and pelvic MRI and CT, contrast enema, voiding cystourethrogram as well as vaginoscopy and were confirmed during surgery. They included orthopaedic/neurological malformations (four legs with two hypoplastic ventral legs, a second dysplastic pelvis, a partially split/double sacrum with intraspinal lipoma and tethered cord), urogenital malformations (fused renal ectopia on the right side with one orthotopic and one ectopic ureter to the urodynamically normal right bladder, hypodysplastic left kidney with one refluxing ureter to the left neurogenic bladder, both bladders with one urethra, double internal and external genitalia consisting of two hemiuteri with one ovary each and two vaginal openings), and colorectal malformations (imperforate anus, duplication of the colon with beginning at the hepatic flexure and each ending as a perineal fistula). Vascular supply to the parasite was provided by the left common iliac artery and by a retroperitoneal artery (CT 1; Illustration 3).

In the first surgery at the age of 17 months heteropagus separation was performed by resection of the hypoplastic legs, complex bilateral

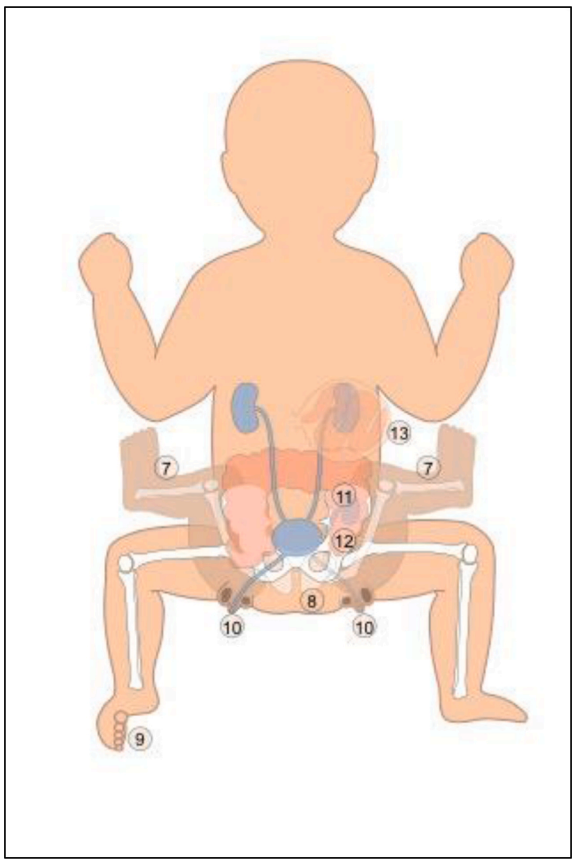


Illustration 2. (Case 2) Ischiopagus twins with ⑦ rudimentary limbs, ⑧ rudimentary pelvis and fusion with the autosite's pelvis, ⑨ clubfoot, ⑩ two sets of male genitalia, ⑪ third pelvic kidney, ⑫ imperforate anus (high type), ⑬ omphalocele with herniation of intestinal loops of the parasite.

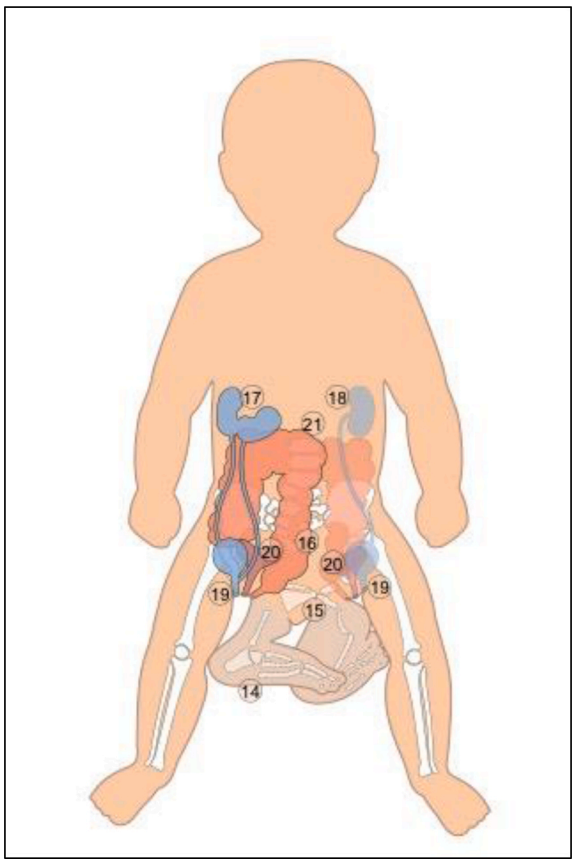
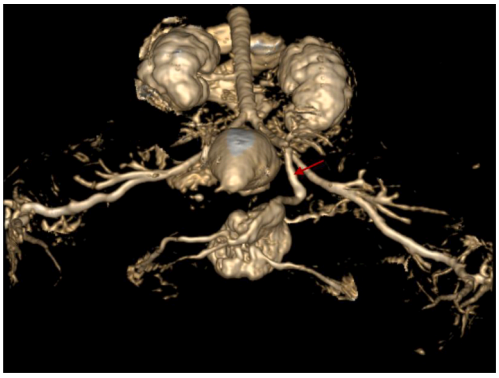


Illustration 3. (Case 3): Ischiopagus twins with ⑭ rudimentary limbs, ⑮ dysplastic second pelvis, ⑯ partially split/double sacrum, ⑰ fused renal ectopia with one orthotopic and one ectopic ureter, ⑱ third hypoplastic kidney, ⑲ two bladders, ⑳ double internal and external genitalia, duplication of the colon with imperforate anus and two perineal fistulas.

osteotomy of the ilium, one level on the right side and two levels on the left side as well as resection of the second dysplastic pelvis. Afterwards, anterior closure of the pelvic ring was performed and stabilized with external fixation technique. In the same session the two external genitalia were approximated in the midline.

The second operation was performed two months later at the age of 19 months and included resection of the left part of the colonic duplication, appendectomy, construction of a dismembered colostomy, an open colonic pull through combined with a posterior sagittal anorectoplasty (PSARP), resection of excessive pelvic parts as well as resection of the left hemiuterus, vagina, fallopian tube, urinary bladder and urethra. This step was necessary to achieve good functional and cosmetic results of the external and internal genital reconstruction. Additionally, reimplantation of the right ectopic ureter to the right bladder and end-to-side anastomosis of the left ureter to the ectopic ureter were performed. In a third and last operation three months later, the colostomy was successfully taken down.

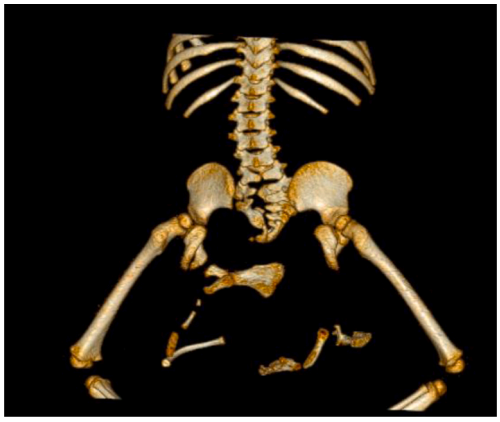
Postoperative course was uneventful. Currently, the patient shows full faecal continence with defecation about once daily. Vesicoureteral reflux on the left side is no longer detectable. An urodynamic examination before discharge demonstrated a low capacity bladder with normotensive filling, complete emptying and no evidence of neuropathic bladder dysfunction or urinary incontinence. But it cannot be excluded that the spinal cord pathology may have a negative impact on the bladder function in the future. A follow up examination including magnetic resonance imaging of the spinal column and urodynamic examination is planned in one year. An untethering may become necessary if clinical signs or symptoms of deterioration are observed. Concerning motor functions, the patient was able to stand safely and walk independently for longer distances at discharge.



MRI 1. (Case 2) MR angiography: a branch of the internal iliac artery (arrow) supplies the parasite.

5. Discussion

The aetiology of heteropagus twins is not fully understood and there is still a considerable debate over this issue. A widely accepted theory suggests an incomplete fission of one zygote, which also explains the origin of symmetrical conjoined twins [1]. An alternative hypothesis assumes the fusion of two separate zygotes [3,8,9]. A third theory proposes that ischemic atrophy may result, such that the parasite is the



CT 1. (Case 3) Pelvic CT: 3D reconstruction of the skeleton showing four legs with two hypoplastic ventral legs, a second dysplastic pelvis, a partially split/double sacrum.

consequence of a vascular compromise leading to an ischemic insult followed by the death and incomplete resorption of this twin as well as an attachment to the normally developed autosite [1,10]. In comparison to symmetrical conjoined twins, there is a male predominance in heteropagus twins and serious malformations also seem to be more common in males [3]. In view of the small number of cases we cannot confirm this gender predominance and we observed both genders being equally affected in regard to the severity of malformations.

The parasitic twins in our case series were all acardiac and acephalic. All of them consisted of supernumerary limbs and external genitalia and partially of some other organs such as intestine and components of the urinary system. These findings are comparable to the available literature. There are only a few reports of cardiac tissue found in parasitic twins that are usually acephalic and they mostly consist of limbs, genitalia and viscera [1,3,11].

Histological investigations of the parasites' limbs in our cases were not initiated with the exception of the first patient where only soft tissue was found in the rudimentarily developed limbs. The other two parasites showed almost normally developed limbs and radiological imaging demonstrated them to be composed of fat and bone tissue. In all cases spontaneous movements of the parasites limbs were not observed. This could be explained by the absence of neural innervation of the parasites' myoblasts leading to incomplete differentiation and skeletal muscular atrophy. Therefore, parasites' histology predominantly shows fat and bone tissue [1,2,12,13].

Autosites often have malformations themselves that are not associated to the fusion with their parasites [3]. The most common malformations include congenital heart defects, especially ventricular septal defects [1]. Furthermore, they can suffer from congestive heart failure due to the additional volume load on their cardiovascular system that has to supply the parasitic twin as well. There is also a high incidence of associated omphaloceles, especially in omphalopagus twins [2,12]. Spinal cord defects are less commonly seen [1]. In our case report we could confirm these observations. One patient had a mild heart defect in the form of an atrial aneurysm, one patient had a spinal column and cord defect in the form of a partially split/double sacrum with intraspinal lipoma and tethered cord and two patients had an omphalocele.

Vascular communications seem to be less marked than in symmetrical conjoined twins, making surgical separation less complicated [1,2]. The vascular communications in our patients were also less complex and vascular separation could be performed easily. Shared vital organs that are often observed in symmetrical conjoined twins are usually not seen in heteropagus twins [14]. Visceral connections between the heteropagus twins are also observed rarely. More frequently, residues of parasitic organs within the body of the autosite have been described [1,2,10]. However, there are a few case reports of shared organs and organic connections between heteropagus twins. Shared pelvic organs are especially common among ischiopagus twins and shared visceral organs among omphalopagus twins [3,15]. In our patients we found some shared organs and organic connections as well as parasitic organs within the autosites involving visceral and pelvic organs.

Parasites sometimes contain viscera, especially intestine [1,3], as it was observed in our cases. Even the first parasite that was only rudimentarily developed showed differentiated intestinal structures. Shared visceral organs or visceral connections are common in symmetrical conjoined twins with the hepatobiliary system, the duodenum and the jejunum being most affected. Separation procedures are often very complex and the outcome is unfavourable in twins with marked fusions, especially of the liver and the bile ducts [16]. In contrast, intestinal and hepatobiliary connections seem to be very rare in heteropagus twins and there are no reports about complicated intestinal separation procedures [3,11]. Only one of our patients showed connections to the parasitic intestine by a colonic duplication and separation procedure was uneventful confirming that complex intestinal connections are not to be expected. Additional visceral malformations, such as omphaloceles, that are not associated with the twins' fusion may occur in the autosites [1,3,4]. Two of our patients showed an omphalocele and we also found a high incidence of imperforate anus (two out of three patients). To our knowledge, this has not yet been described in the literature. These additional malformations can have a negative impact on the patient's outcome and quality of life in different ways. For instance, both our patients with anal atresia suffered from faecal incontinence. The third patient had permanent defecations through the two perineal fistulas before corrective surgery and the second patient suffered from persistent faecal incontinence after anal atresia repair. The final outcome was favourable. The third patient was continent after anal atresia repair. The second patient achieved social faecal continence by the installation of a bowel management using a caecostomy catheter for regularly applied anterograde enemas.

In symmetrical conjoined twins certain similarities are seen in upper urinary tract anomalies while the lower urinary tract and genitalia mostly show unique characteristics in each twin pair [7,15,17]. These observations have not yet been described in asymmetrical conjoined twins and our patients also showed unique upper and lower urinary tract anomalies. Some organs of the urinary system were supernumerary or duplicated. We also found connections between the twins' urinary tract systems. The second patient showed a third kidney and ureter belonging to the parasite, one common urinary bladder and two urethrae. In the third patient we found tree kidneys, one of them was supposed to belong to the parasite, three ureters, two urinary bladders, the right one with normal function, the left one with neurogenic dysfunction and additionally two urethrae. Urological problems including urinary incontinence, vesicoureteral reflux and recurrent urinary tract infections often accompany urinary tract malformations. Beyond the challenges of surgical separation, further complex reconstructions may be necessary to achieve urinary continence and in order to avoid severe complications such as renal function impairment close follow-up is recommended. Additionally, heteropagus twins often have concomitant spinal cord anomalies that may lead to neuropathic bladder dysfunctions and that require ongoing monitoring as well as potential treatments [1,7,15]. Despite complex urinary tract malformations in two of our patients, urinary continence was observed. It remains to be seen whether the spinal cord malformation in the third patient will have an impact on the bladder function in the future. A case of left sided vesicoureteral reflux with a subsequent episode of pyelonephritis in the same patient was no longer detectable after ureteral reimplantation. So far, there was no renal function impairment in any of our patients.

Shared duplicate genitalia seem to be very common in ischiopagus twins [3] and these features could also be observed in two of our patients. Both showed completely developed duplicated internal and external genitalia that were symmetrically located between the twins' legs. Genital reconstruction was very complex including the challenge of

ventral approximation and fusion of the external genitalia and in the second patient several surgical steps were necessary. However, satisfying cosmetic results could be achieved in both patients. It is unclear if these two patients will be able to father children or to conceive in the future. We were not able to find any information concerning fertility and childbearing in the literature, possibly because most of the successfully operated children are still too young.

Additionally to the supernumerary limbs that mostly contain bone tissue, the presence of other bony structures like pelvic or shoulder bones in the parasitic twins and complex fusions with the autosites' skeletal structures have previously been reported. Orthopaedic reconstruction in the first years of life and close orthopaedic follow-up seem to be very important as the parasitic twin may interfere with the autosite's locomotion development and may also lead to further skeletal deformities such as scoliosis [18,19]. In our patients we found parasitic pelvic structures and in two of our patients there were complex fusions with the autosites' pelvises. These patients had severe impairments of their locomotion development and were not, or only with support, able to stand and walk. Complex orthopaedic reconstruction and, in the case of the second patient, a second intervention were necessary to improve pelvic anatomy and stability. Good functional results could be achieved and both children are now able to stand upright and to walk independently.

Prognosis regarding survival of the autosite is generally favourable. The main reasons for a negative outcome include congenital malformations, especially heart defects, of the autosite himself [20]. Other possible non-lethal long-term complications include incisional hernia and teratoma formation at the site of retained parasitic tissue [6].

6. Conclusion

In summary, only very few case reports describe the outcome of conjoined twins regarding gastrointestinal, genitourinary and motor functions. Most of these reports include symmetrical conjoined twins that usually show other anatomical features [7,15] and are rarely about asymmetrical conjoined twins as we describe in our cases [15,21]. Furthermore, these case reports often originate from low-income countries with less developed health-care systems. Therefore, they are not always comparable to reports from high-income countries. In this case series we could demonstrate that despite complex anatomic features and severe malformations, good functional and aesthetic results concerning gastrointestinal, genitourinary and motor functions can be achieved in asymmetrical conjoined twins. A detailed study of the anatomic features by clinical examination and with the use of different imaging modalities was mandatory for the surgical planning and several surgical reconstructive steps had to be performed by a multidisciplinary team over the first years of life. Ultimately, however, we believe that quality of life improved considerably and it is likely that these patients will be able to live an independent life in the future.

Patient consent

Consent to publish the case report was obtained.

Funding

This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

Authorship

All authors attest that they meet the current ICMJE criteria for Authorship.

Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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