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Successful Separation of Pygopagus Twins at a Tertiary Center in a Third World Country: A Literature Review with Case Presentation

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Abstract

Introduction: *Conjoined twins, a rare phenomenon, have long fascinated medical practitioners, with their origins and development still not fully understood. This article delves into the intricacies of conjoined twins, focusing on those sharing parts of the brain and spinal cord, and the surgical techniques used for their separation.*

Case presentation: *we report the case of a pregnant woman with pre-eclampsia who delivered parapagus twins at 37 weeks gestation. The twins, connected from the pelvis, were successfully separated at 4 months of age, with one twin requiring resuscitation at birth.*

Discussion: *A literature review was done on various types of conjoined twins, including craniopagus, parapagus, ischiopagus and parasitic rachiopagus, each presenting unique challenges in surgical separation. Surgical approaches differ based on the extent of fusion and shared structures. Challenges include preserving neurological function and managing potential complications.*

Conclusion: *Advancements in surgical techniques and understanding have improved outcomes, but separating such twins remains a difficult task requiring multidisciplinary collaboration and planning among surgical teams.*

Keywords: Pygopagus Twins, Conjoined Twins, Sacral Fusion, Separation Surgery, Magnetic Resonance Imaging

Abbreviations and Acronyms
CE: Cauda equina
CM: Conus medullaris
CSF: Cerebrospinal fluid
CVS: Cerebral Venous Sinus
DS: Dural sac
FT: Filum Terminale
NC: No complications
OP: Operation
SC: Spinal cord
SCV: Superior Cerebral Vein
SSS: Superior sagittal sinus
VC: Vertebral column

Introduction

For many centuries, conjoined twins have garnered a lot of attention and interest. They were first mentioned when there was little information available about them. Conjoined twins are

physically merged during pregnancy and their formation has two possible explanations. 13 weeks or longer after fertilization, the zygote either incompletely or entirely split with a partial fusion at a specific site depending on how the twins are positioned in relation to one another; on top of many other factors, we do not fully understand [1]. These justifications, however, do not apply to every case [2]. The crowding hypothesis has also been suggested as a third explanation that demonstrates the origins of conjoined twins in addition to the fission and fusion theories. According to this view, the onset of ventrally, caudally, and laterally conjoined twin development is caused by the existence of an initial multiplication of horizontally positioned morphogenetic potent primates within a single core cell mass [3].Conjoined twins are rare. Their incidence is estimated to be 1 in 30,000 to 1 in 250,000 live births; more than 60% of these twins die before birth, and the prognosis of survival depends on the site of fusion along with the extent of shared body parts [4,5]. In this article, we discuss different types of conjoined twins who share part of their brain and spinal cord, as well as the surgical approaches used in their separation. We also present a case report detailing the successful separation of conjoined twins by the neurosurgical team at our institution.

Case presentation

A 30-year-old pregnant female patient was referred to our center for urgent c-section delivery in the setting of pre-eclampsia and conjoined twins at 37 weeks of gestation. The patient was noted to have conjoined twins at 25 weeks of gestation, suspected on prenatal ultrasound, then confirmed on prenatal magnetic resonance imaging (MRI) which showed fusion at the level of the pelvis down to the sacrum (pygopagus). At birth, the first member of the conjoined twins weighed 3100 g, was vigorous and required no resuscitation, while the second member weighed 2000 g, was limp, cyanotic, apneic, and required resuscitation by the neonatal intensive care team.

On physical assessment, the twins were moving all extremities with good muscle tone, and both had open, soft, and depressed fontanelles. The first member had a head circumference of 34 cm, while the second member had a smaller head circumference of 29.5 cm. The second member of the twins had a 10 cm posteriorly located encephalocele (figure 1). The twins were found to be connected at the level of the sacrum (figure 2), including the genitalia, with a single anal opening, and the skin connecting the patients was covered with a small tuft of hair (figure 3). No obvious deformities of the skin or back in both twins were noted.

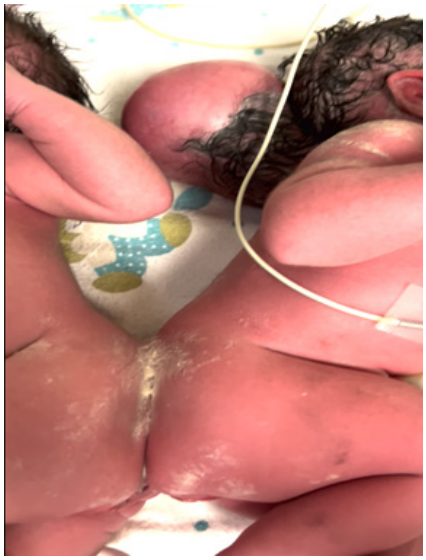


Figure 1: Photograph showing the pygopagus twins and the encephalocele of Twin B.

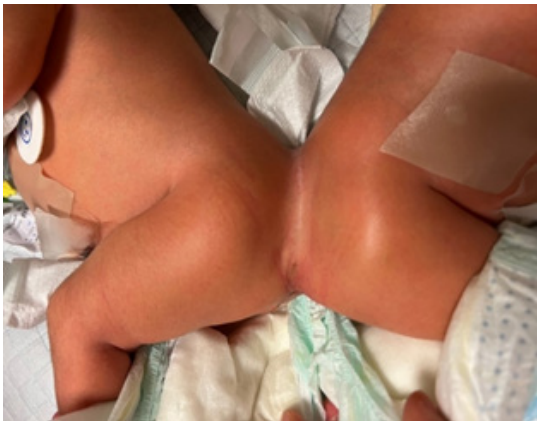


Figure 2. Photograph of the pygopagus twins at day 0 of life connected at the level of the sacrum

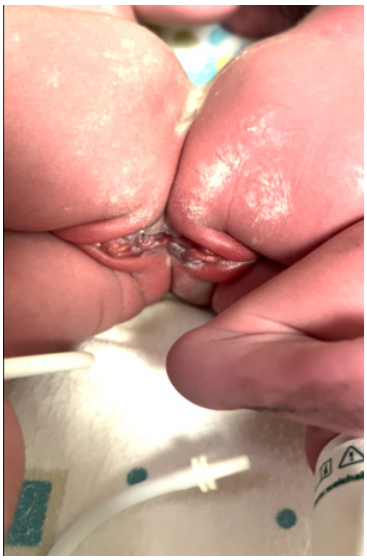


Figure 3: Photograph showing the fusion at the level of the genitalia with a single anal opening.

On the day of the operation, at 4 months of age, the main challenge in the surgery was to put the patients in the appropriate position for us to be able to work properly and have good access (figure 4). Keeping in mind that the position should be appropriate for repair of the encephalocele of twin B as well. It was also a challenge for the anesthesia team where several anesthesiologists worked together to ensure intubation and sedation were properly performed since the twins also share common blood flow which impacts the process of putting them under anesthetics. We first started with repair of the encephalocele where a bony defect was identified with an opening in the dura that was closed ensuring no CSF was leaking after the tight closure. After discussing with the pediatric surgeons, as well as with the plastic surgeons, we planned the skin incision at the lateral aspect of the back and above the connection of the two babies (figures 5 & 6). This was performed in a way that allows us to take as much as we can of the skin and subcutaneous tissue to do adequate flaps for proper closure. Two attending neurosurgeons each working on one of the babies did the skin incision. Then, going deeper towards the bony coccygeal connection that was cut using Rongeurs and kerrisons. The bony disconnection was done circumferentially around the communicating dural sac. At this point, we opened the dura identifying the connection between the two filum terminale that was cut, then each surgeon started working on one of the babies. Then, we released the arachnoidal adhesions tethering each cord to the dura. Finally, we did a watertight closure of the dura on each side. Then, the pediatric and plastic team started working.

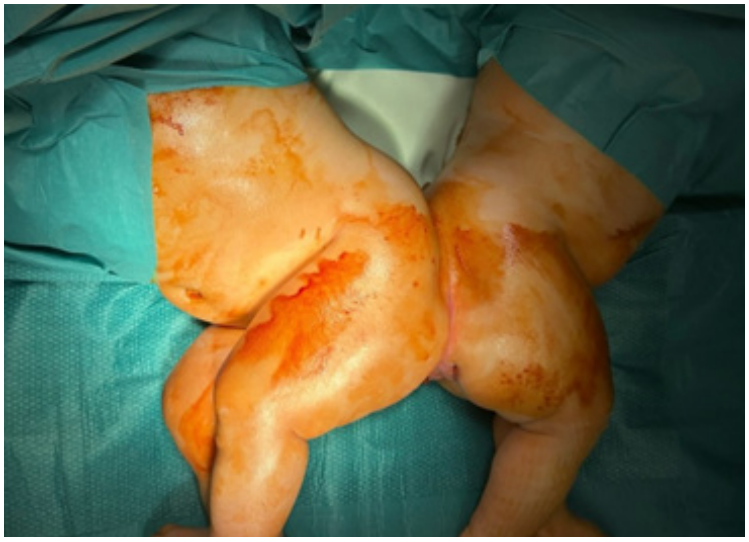
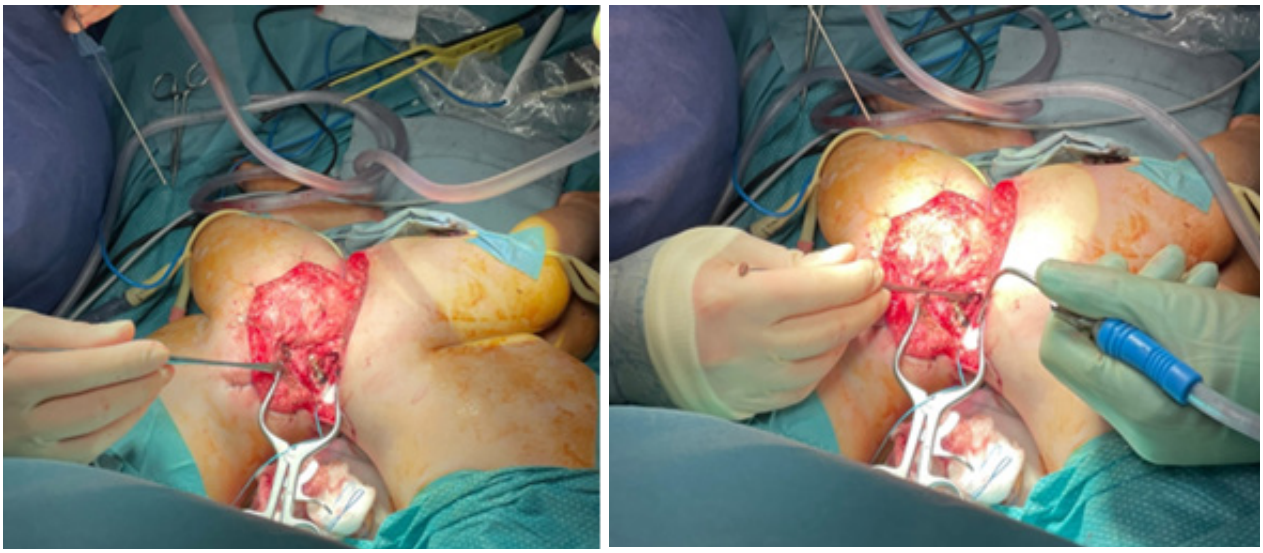


Figure 4: Intraoperative positioning of the twins



Figures 5 & 6: Intraoperative photographs of the pygopagus conjoined twins

Background & Discussion

Craniopagus twins

Multiple types of conjoined twins share parts of the brain or spinal cord. 2% to 6% are conjoined at the cranium and are known as craniopagus twins. They account for 1 in 2.5 million births [6,7]. CPT has been classified using multiple categorizations.

In 1976, O'Connell classified CPT as total or partial. Total CPT shares a large surface area with extensively linked neurocranial parts, whereas partial CPT only affects a small, superficial area. In addition, he separated partial CPT into vertical or angular based on the extent to which one head rotated in proportion to the other, each group having a distinctive array of brain abnormalities and aberrant blood flow [8].

In 2006, and based on the evaluation of 64 cases, Stone and Goodrich identified partial CPT as the absence of significant shared venous sinuses; compared to CTP that shares a significant amount of DVS and manifests with severe brain compression, which causes deformations inside the skull [9].

In 1987, Winston opted for a more thorough classification. He divided CPT into 4 groups according to the degree of shared parts between the two twins [10]. Group A twins shared only part of the cranium with separated meninges and superior sagittal sinuses. Part of the periosteal dura is shared in group B. It is the part that separates the two superior sagittal sinuses of the two twins, and because there is only one layer left, there probably isn't enough epidural space for the two lumens to develop [11]. Group C and D have two shared circumferential sinuses and fused leptomeninges at the site of attachment with group D having merged brain parenchyma in addition.

CPT separation surgery presents significant challenges. Literature of cases that we collected for review (table 1) reveals that a critical decision for neurosurgeons is the selection of the twin with priority for the complete shared structure based on preoperative neurological assessment. A study by Dunaway et al. elucidated the separation process, highlighting that as Twin 1 uses the shared superior sagittal sinus to drain its deep venous system while Twin 2`s deep venous system is independent of it, neurosurgeons decided that the shared superior sagittal sinus must go to twin 1 [12]. However, in cases where twins exhibit distinct brain activities, as reported by Wolfowitz et al. neurosurgeons must equally divide and preserve shared structures [13]. A major challenge arises from potential intraoperative hemorrhage due to interconnected cortical veins from one twin to the other's venous sinus, complicating the process. Addressing this, Wolfowitz et al. [20] demonstrated that preoperative venography, revealing a connection between Twin 1's superior sagittal sinus and Twin 2's superior cortical veins, facilitated hemorrhage control during separation with Gelfoam. Another complexity arises when both twins share and depend on the same sinus. In a case documented by Drummond et al. where the posterior third of the superior sagittal sinus, torcula herophili, and a common lateral sinus were shared, the initial approach involved gradual occlusion of the posterior superior sagittal sinus over several days, enabling drainage to be taken over by collateral veins [14]. Nonetheless, even after such meticulous dissection, the risk of post-dissection bleeding remains high. Despite these challenges, several reports [20-23] noted minimal postoperative complications, with one study by Drummond et al. [21] mentioning a mild cerebrospinal fluid (CSF) buildup in the flaps, subsequently resolving with normal drainage, and overall positive post-surgical outcomes for the twins.

Pagopagus twins

20% of all CT are pygopagus, 6 to 19% share part of the spinal cord, genitourinary system, and GI tract to varying degrees [15, 16]. They stand for a set of conjoint where there was insufficient caudal separation of the embryonic axis [17].

In our pygopagus twins' case, proper patient positioning presented the primary surgical challenge. Consequently, neurosurgeons opted for a lateral incision above the area of connection. Subsequently, the fused coccygeal bone was excised without complication, the dura mater was opened, and the shared filum terminale was sectioned. Upon completing dural closure for each infant, the neurosurgical team concluded their part, allowing the pediatric surgery team to take over. According to the pediatric surgeons, the most effective method for identifying the anal

sphincter muscles was through direct muscle stimulation. Subsequently, the muscles were identified and dissected [18].

As our case proceeded smoothly and both twins were healthy, we contrasted our experience with a case documented in the literature where twin 1 was healthy but twin 2 was deemed unlikely to survive [19]. Consequently, the neurosurgery team focused on maximizing twin 1's chances of a long and healthy life, given twin 2's prognosis (see table 2).

In pygopagus twins, identifying which nerve root belongs to each baby is highly challenging for neurosurgeons aiming to separate them fiber by fiber while preserving their functionality. Hence, in many papers reviewed (see table 2), intraoperative root stimulation via tEMG was employed. In the aforementioned case, neurosurgeons successfully identified the most dorsal extent of Twin 2's spinal cord supplying only twin 2, using root stimulation [19]. Following the identification of the precise point of innervation sharing, the spinal cord was coagulated and separated just posterior to that point, thus retaining the innervation of twin 1. A study by Yokota et al. also underscored the utility of tEMG for guidance [20]. This step is particularly challenging as neurosurgeons must be exceptionally careful not to sever any fibers to preserve maximal function for each twin. Additionally, recognizing that the filum terminale lacks neurons to fire action potentials, Yokota et al. emphasized in a detailed separation process the necessity of stimulating the shared, fused filum terminale before its incision to ensure no tiny nerves or fibers are attached to it [20].

In a case reported by Feiggen et al. the twins shared a fused conus medullaris, and since their neurological functions were found to be equivalent, neurosurgeons worked to preserve both twins' functions [21]. Consequently, the conus was meticulously dissected in the midline, preserving the spinal cord of each twin. Case reports by Chou et al., Awasthi et al. and Yokota et al. showed no complications post-separation process of pygopagus twins [20, 22, 23]. In the sole case by Feiggen et al. where CSF leakage occurred on day 9 postoperatively, both babies exhibited the same level of weakness as before surgery two weeks later [21]. This retention of symptoms could be attributed to several explanations, including intraoperative damage to nerve roots during the fiber-by-fiber separation process, or preexisting damage to the nerve roots due to the merging and fusion process, preventing the infants from regaining full neurological function.

Ischiopagus twins

Ischiopagus twins are connected at the pelvis and often have three or four legs. They account for 6% of all CT. Usually, the terminal ileum, which drains into a single colon, serves as a junction point for the digestive tract. Ischiopagus twins have two bladders and four kidneys, and frequently the bladder of one twin crosses halfway and empties into the other twin's bladder [24]. In addition, the spinal cords of ischiopagi are shown to be fused from end to end. Conjoined twins from the ischiopagus and pygopagus twins have a unique spectrum of spinal deformities that pose difficulties during separation as well as postoperative management [25].

MRI conducted on ischiopagus twins can reveal a fused terminal thecal sac. However, visualizing any merging cauda equina nerve roots in such cases, as reported by Cywes et al. in (table 3), is challenging [26]. This difficulty likely stems from the perpendicular alignment of fusion between the twins' vertebral columns and spinal cords.

Parasitic rachipagus twins

Parasitic rachipagus twins, which are two distinct twins, one of which is a full twin and the other merely a body part, usually affect the cervical and thoracic regions of the spinal cord with the partial twin originating from the craniocervical junction or thoracic region. The complete twin usually shows diplomyelia, which is usually left untreated since it was believed to be asymptomatic [27].

In a separation surgery reported by Khavanin et al. in (table 4), the partial twin required removal along with its entire neuro-vasculature structure to mitigate potential harm to the other infant [27]. Regarding diplomyelia, since the complete twin remained asymptomatic, the duplicated spinal cord was left untreated. While this condition may not manifest negative effects on the infant, the spinal cord's tethering during growth necessitates ongoing neurological monitoring.

Table 1: Literature review of cases of craniopagus twins' separation.

Author	Age at separation	Neurosurgical separation process	Neurological status, shared neural structures (+)/ (-) associated deficits and follow up	Postop- neurological complications
Wolfowitz et al. [20]	3 M	1. Incision was made at the junction between the 2 heads. 2. A curved raspatory was inserted in Twin 2`s anterior fontanelle under the frontal bone to separate the dura along the bony bridge. 3. A complete separation of the dura was made 4. Administration of Gelfoam controlled bleeding during the process of separation.	**Preop:** - No obvious impairments. - No cortical connections given that the twins have independent behavior along with separate brain activity. - Twin 2: possible Dural defect. **Postop (1 month):** Twin 1: Mild internal strabismus of the left eye and hypoplastic SCM.	Twin 1 had a mild meningismus with fever on the 5th day. -Same episode on the 18th day.

Drummond et al. [21]	OP 1 & 2: 9M OP 3: 17 M	1. Slowly occluding the posterior SSS over several days. 2. A screw clamp was placed at the junction of the middle third of the SSS of each twin to occlude the junction. 3. After ensuring that the common posterior third of the SSS was completely occluded, an incision was made across it. 4. Dissecting venous pools of the tentorium cerebelli caused rapid bleeding and twin 2 became hypotensive. This incident delayed the final step to allow for adequate neurological recovery. 5. At the final operation, the twins were finally separated and the dura of twin 1 was left intact, whereas the brain of twin 2 was wrapped with collagen material.	**Preop:** - The posterior third of the superior sagittal sinus and torcula herophili were shared between the two twins along with a common lateral sinus. **Postop:** The twins regained consciousness quickly after the anesthesia was reversed.	Both twins had a mild build-up of CSF behind the flaps followed by normal drainage.
Dunaway et al. [22]	6 M	1. A midline plane was created between the two brains along with a phased method to divert Twin 1`s superficial venous contribution to the shared SSS. 2. At the first operation: The dura was opened, and it was easy to view the SCV emptying into the CVS. Stage 1 separation of twin 1`s superficial veins was done as planned with no venous congestion. 3. Second operation (4 weeks after): Dissection of the remaining veins in Twin 2. The 2 arteries that were supplied by twin 2 to twin 1 were also cut. The 2 brains were totally separated.	**Preop:** - The twins are TV type III - Absence of neurological abnormalities. -Twin 2 was hypertensive due to the asymmetrical venous drainage resulting in shunting of blood from twin 1 to twin 2 - Twin 1 uses the shared SSS to drain its deep venous system. Twin 2 is independent of it - One common dura separating the 2 brains. This shared dura has a defect which allows a small brain area connection. - The exact location of the artery crossing from twin 2 to twin 1 is marked by the small region of adjacent brain tissue suggesting a minimal arterial supply of twin 1 from twin 2. **Postop:** - Absence of any neurological deficits. - Proper development of the brain.	NC

Kheir et al. [23]	11 M	_	**Preop:** - The twins shared one cranium vertically with abnormal parietal bones. -Communica tion between the ventricular systems of both twins. -Common posterior part of SSS. **Postop:** Absence of any neurological defects	NC

Table 2: Literature review of cases of pygopagus twins' separation.

Author	Age at separation	Neurosurgical separation process	Neurological status, shared neural structures (+)/ (-) associated deficits and follow up	Postop- neurological complications
Fowler et al. [24]	10 W	1. Laminectomy was performed on each baby to expose the conjoined SC and DS. 2. U shaped SC was revealed after opening the dura. 3. To track down muscle responses to motor root excitation, muscle relaxants were not used. 4. Sacral roots were shared since their stimulation caused the muscles of both twins to contract at the sacral region. 5. The most dorsal extent of Twin 2`s SC was identified using roots stimuli, and it supplied twin 2 only. 6. SC was coagulated and separated 7. Separation of twin 2`s sacrum using the sacroiliac joints retained a complete sacrum for twin A. This procedure also helped to avoid the severe bleeding seen in previous pygopagus twin separations.	**Preop:** -The twins shared the terminal end of the spineand Dural sac. - Twin 1 had tethered S1 innervation defects to the left extremity at the shared end of SC. In addition, twin 1 had positional scoliosis as well as vertebral structural abnormalities: Butterfly vertebra at T4 and hemivertebra at T1,2. -Twin 2 was found to have the "smooth brain" AKA Lissencephaly, microcephaly and associated hydrocephalus. Twin 2 also had scoliosis and diastematomyelia with S1 innervation abnormalities of the right extremity. **Postop follow up:** At the age of 5, twin 1 was diagnosed with neurogenic bladder probably due to abnormalities in the pelvic parasympathetic innervation.	-Twin 1 had an extensive pelvic floor hernia that was concealed by a healthy peritoneum. The closure was achieved by attaching the gluteal and residual levator muscles.
Feiggen et al. [11]	9 M	1. Neurological functions were the same in both twins. The aim was to keep both twins` functions intact. 2. Laminectomy was done. 3. The dural sac was opened. The fused end of both SC was exposed as a single CM. 4. FT was cut and the conus was separated in the middle. Right away, the SC of each twin retracted upwards probably because before separation, they were stretched distally by the short nerve roots.	**Preop:** - Twin 1 had a mild atrophy of the left calf muscles along with reduced ankle eversion and plantar flexion. Twin 2 on the right side. - Both twins had scoliosis and flat head syndrome AKA plagiocephaly. - The twins shared the terminal end of the SC at the sacral end and the DS. - Twin 1had a T9 hemivertebra on the left side. **Postop follow up:** 2 years later, both twins were doing fine.	-CSF leakage in one baby on the 9th day. - 2 weeks later, both twins showed the same level of weakness as it was before the operation.

Chou et al. [13]	7 M	1. The aim of surgery was to preserve the SC function in both twins as there was no discernible neurologic differences between the twins. 2. The epidermal cyst was removed from the united CM, and the combined dural sac was incised in a "Y" shape. Finally, the dural sacs were closed without using any grafts.	**Preop:** -Two separate sacral bones with a single thecal sac at the site of SC fusion. - Y- shaped type of terminal SC convergence. - Epidermal cyst at the site of fusion. **Postop follow up:** No CSF leaking No other complications	NC
Awasthi et al. [25]	1.5 M	1. A vertical cut was made along the site of fusion with little extension above and below the site starting with normal structures all the way down to the fused ones. 2. The dura was opened, and the shared FT was removed. 3. The end of the two separate SC of each twin were in the single thecal sac. They were separated and put back in position. 4. Duraplasty was done, and the cords were sealed.	**Preop:** -The twins are conjoined at the lumbosacral region with separate SC and sacrum but fused thecal sac and FT at the level of S3 where the sac normally ends. - No hydrocephalus or any motor or sensory impairments. - Both twins had spina bifida at the level of L4 and down. **Postop follow up:** Full recovery	NC
Yokota et al. [26]	6 M	1. Dorsal incision was made to expose the conjoined sacrum and get access to the thecal pouch containing the spinal canal. 2. Sacrum was separated using an anatomical 3D representation and dural sac was assessed. 3. Adhered nerve roots were separated from the meninges (dura and arachnoid) and FT. 4. tEMG was used to distinguish whether the nerve roots belong to twin 1, twin 2 or both. 5. After dividing nerve roots into two groups (shared and not shared), common roots were now subdivided according to which twin each nerve fiber belongs and finally separated fiber by fiber. 6. FT was cut at the midline after being stimulated. 7. Dura matter was divided equally and closed tightly	**Preop:** - The twins were connected at the sacrum (beginning with S1 vertebra) and had a continuous SC tube enclosed inside a single terminal dural pouch. A fused FT was found at the level of S1 - No signs of muscular atrophy, foot deformity, or any motor or sensory deficits in both twins. **Postop:** No neurological impairments at the lower extremities or pelvic organs were noted.	No CSF leakage or SC tethering.

Table 3: Literature review of a case of ischiopagus twins' separation.

Author	Age at separation	Neurosurgical separation process	Neurological status, shared neural structures (+)/ (-) associated deficits and follow up	Postop- neurological complications

Cywes et al. [27]	9 M	1. The continuous VC was exposed following a dorsal incision. 2. The abnormal vertebra at the site of junction was removed piece by piece to expose the common thecal pouch. 3. Massive epidural bleeding was reported when the dura was opened posteriorly. 4. Two connected CE nerves were witnessed intraop in twin 1. 5. Two more vertebras had to be removed to have a good closure of the twins. 6. After the additional vertebrectomies, dura was sealed tightly	**Preop:** - Twin 1 had atrophy of the left calf and foot. -No neurological deficits were noted. -The twins had only one continuous vertebral column with some structural abnormalities: Common right hemivertebrae at T10 and left hemivertebra at T5 in twin 2. Although the MRI showed a fused terminal thecal sac, there was no evidence of a merging SC or CE. **Postop:** -No new neurological deficits were developed. **Postop follow up:** -Twin 2 was diagnosed with a mild asymptomatic syringomyelia at the age of 2 - An extensive enlargement of the syrinx resulted in an abnormal gait and muscle weakness at the level of knee and ankle of the same twin at the age of 3. - Both twins suffered from scoliosis	NC

Table 4: Literature review of a case of parasitic rachipagus twins' separation.

Author	Age at separation	Neurosurgical separation process	Neurological status, shared neural structures (+)/ (-) associated deficits and follow up	Postop- neurological complications
Khavanin et al. [19]	9 M	1. The partial twin was removed and the neuro-vasculature supplying it was completely ligated. 2. The doubled SC and intradural lipoma were left untreated because they were totally asymptomatic as the neurological examination showed no impairments.	**Preop:** -The complete twin had right plagiocephaly but a normal neurological examination -The partial twin is a pair of lower extremities connected at the craniocervical junction of the complete twin. Twin 2 showed a movement in the toes along with noticeable sensation. - MRI showed that twin 1 had two separate spinal canals and SC (AKA: diplomyelia) with patent intervertebral foramina for each canal. In addition, arches of the cervical vertebra were found to be absent with an intact nuchal ligament. - The complete twin had also lipoma inside the dural sac which extended to the extradural space at the cervicothoracic junction. **Postop follow up:** The development of motor skills has accelerated significantly.	NC

Conclusion

Conjoined twins exhibit a range of neurological symptoms and pose surgical risks after separation due to the extent of neural structural connections. Although this area of medicine is always progressing, it is still challenging to separate such twins. Difficulties are posed not only by craniopagi but also by pygopagi and ischiopagi, which are linked at the sacrum and the pelvic outlet, respectively. They have earned a lot of attention in neurosurgical literature on conjoined twins. Excellent results have been highlighted in the literature as necessitating neurophysiologic intraoperative monitoring, a highly trained neurosurgery team, along with profound harmony and concise communication with all other surgical teams.

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