# SUCCESSFUL SEPARATION OF CRANIOPAGUS PARASITICUS

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**OBJECTIVE:** Craniopagus parasiticus is an extremely rare condition. The first attempt to separate such twins was performed in the Dominican Republic in 2004. The infant died 7 hours after surgery. The aim of this report is to present a case in which surgical separation was successfully performed on February 18, 2005. In February 2006, the child was still alive and in relatively good health.

**METHODS:** The authors operated on a patient with craniopagus parasiticus at Benha Pediatric Hospital in Egypt, 45 km north of Cairo. The child was 10 months old when the surgery was performed. By minimizing the time of surgery and adequate control of intraoperative bleeding, a successful surgical separation was achieved. Computed tomography, magnetic resonance imaging, magnetic resonance angiography, and computed tomographic angiography provided the information necessary to perform surgery.

**RESULTS:** The child underwent operation at the age of 10 months; the duration of surgery was 9 hours. Bleeding was the most serious problem, with the child receiving four liters of blood. The main arterial supply to the parasite was via the middle cerebral artery and was ligated in the Sylvian fissure. Bleeding, however, was mostly venous and was mainly controlled by diathermy and thrombin soaked packs of Surgicel, as well as clipping. After separation of the parasitic head, the dura was repaired using artificial dural grafts. Free bone flaps from the parasite were used to cover the osseous defect in the autosite. Skin flaps from the parasite were also used to cover the cranium.

**CONCLUSION:** This is the second case of craniopagus parasiticus in which separation was attempted. The first patient, operated on in the Dominican Republic, died 7 hours after surgery. In the present case, the child is still alive and without neurological deficit.

**KEY WORDS:** Conjoined twins, Craniopagus, Craniopagus parasiticus, Joined heads

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The word "craniopagus" refers to twins joined only at the head and each child is a component of a craniopagus. The term was derived from Greek (*kranio* meaning "head" and *pagus* meaning "together"). The plural is craniopagi (7). Craniopagus represents 2 to 6% of conjoined twins and is the rarest type of this disorder (4). Conjoined twins are always genetically identical and share the same sex. Females are more commonly affected with a male to female ratio of 1:4.

Craniopagus parasiticus is an anatomic malformation in which conjoined twins are united at the crowns of their crania, with one twin being underdeveloped and forming a parasite (2). The parasitic group has an evident male preponderance.

Craniopagus parasiticus is an extremely rare anomaly. In 1989, Bondeson and Allen (2) reviewed the literature and found six cases of craniopagus parasiticus. The first case on record was reported in 1790 by Everard Home in Bengal. Quoting Bondeson, the second case was reported by Vottem in 1828 in France. The third and fourth cases were reported in Germany by Donitz (1866) and Berndt (1935), and the fifth case was reported in France by Brocard (1939). The sixth case was reported in Brazil by Camara (1940).

Aquino et al. (1) published a report of the seventh case in North America; the eighth was operated on in the Dominican Republic by Lazareff (6), but is still unpublished. Seven of the eight reported patients were males. Attempts at surgical separation have not been commonly performed because the majority of these twins have traditionally been stillborn.

## PATIENT AND METHODS

### Case Report

The diagnosis of this case of craniopagus parasiticus was not made during pregnancy. The mother lives in Benha, a town 45 km north of Cairo, Egypt; she is 29-years-old and has one normal child. After 3.5 years of infertility, she was given

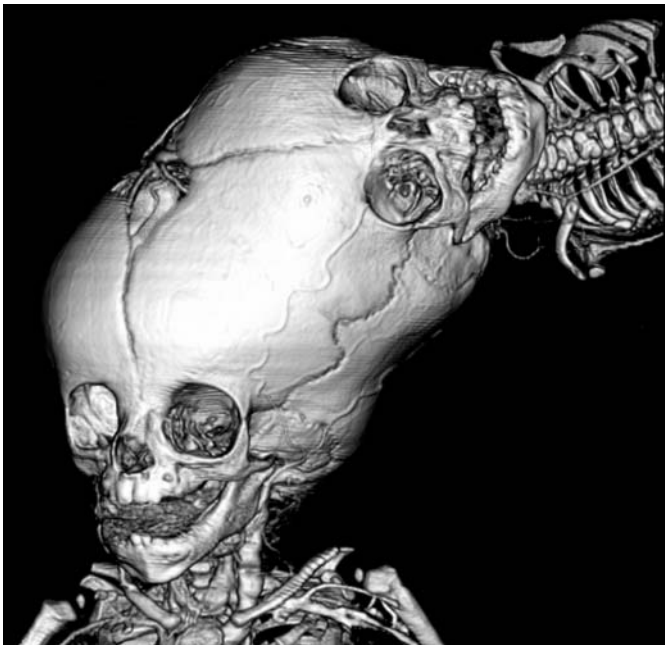

**FIGURE 1.** *CT scan of the cranium with 3-D reconstruction. The left frontal, parietal, and temporal bones of the autosite join the right frontal, parietal, and temporal bones of the parasite.*

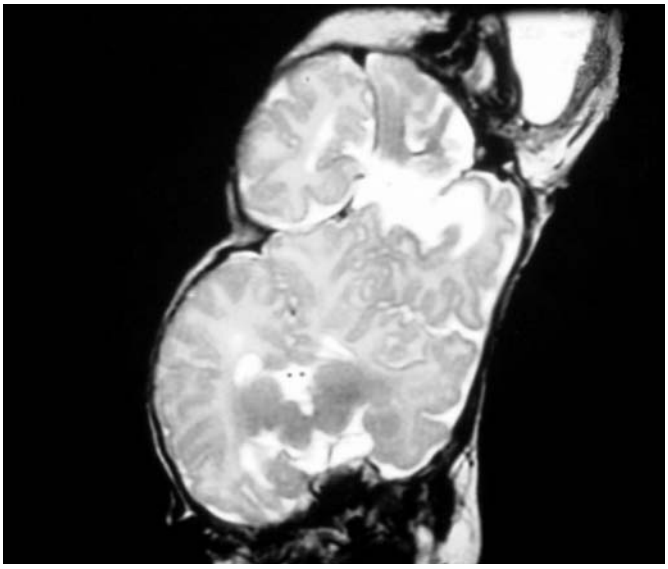

**FIGURE 2.** *Coronal T2-weighted magnetic resonance imaging sequence showing that the left cerebral hemisphere of the autosite is confluent with the right cerebral hemisphere of the parasite.*

clomiphene citrate to induce ovulation. Her subsequent pregnancy was uneventful. On March 30, 2004, the mother gave birth to a normally developed female baby. The delivery of a second baby was obstructed. A Caesarian section succeeded in delivering a second girl with an undeveloped parasitic child attached at the head. The parasitic twin had a normal head attached to a 15-cm trunk without limbs.

The size of the parasitic head was almost equal to that of the normal child. The left side of the cranium was joined to the right side of the cranium of the parasitic twin. The conjoined calvaria had a circumferential base of 42 cm and was connected in the frontal, parietal, and temporal regions (*Fig. 1*) of the twins. The interaxis angle between the two heads (7) was 110 degrees with a small rotational angle. The two eyes of the parasite were normal, although the eyelids were closed most of the time. The nostrils were normal. The mouth of the parasite was normal down to the pharynx and salivation was evident. The parasitic child, although sleepy most of the time, had separate emotional response. X-ray studies demonstrated the presence of 19 vertebrae, of which the last three were fused into a mass. Seven pairs of ribs were also seen, as well as one clavicle and a left humerus. The lungs and heart were not evident on x-rays.

## Postnatal Condition of the Twins

At the age of 3 months, the child developed heart failure because her heart was used to pump blood to both her body and the parasitic twin, which was approximately one-third of her weight. She was diagnosed with cardiac myopathy and her liver began to enlarge.

A computed tomographic (CT) scan showed that the bones of the crania were joined. There was no apparent separation between the two brains. Magnetic resonance imaging scans showed that the right cerebral hemisphere of the autosite baby was normal. The left cerebral hemisphere of the complete girl was attached to the right cerebral hemisphere of the parasite (*Fig. 2*). Magnetic resonance angiography showed that the arteries of the parasitic twin anatomose with those of the complete child. CT, magnetic resonance imaging, magnetic resonance angiography, and CT angiography findings were reviewed by the neurosurgical team. After consulting the neuroradiologist for the possibility of conventional four vessels angiography, another CT angiogram with better quality was considered as an appropriate alternative. The CT angiography revealed significant arterial connection and sharing of the venous sinuses. The superficial temporal artery, as well as the middle cerebral artery and some of its branches, carried blood from the complete child to the parasitic one. The anterior thirds of the superior sagittal sinuses of each twin were separated. The middle and the posterior thirds of the superior sagittal sinuses were united into one sinus (*Fig. 3*). The fused sinus ran around the perimeter of the connection between the two heads, referred to as the circumferential sinus (4, 7).The circumferential sinus was closer to the parasitic brain and was draining both brains. A complex and primitive deep venous system existed with eventual drainage into the shared circumferential sinus. Conventional angiography was not used in this case because of the increased risk this procedure poses in babies.

The neurosurgical team met several times to discuss possible surgical procedures. The team reviewed the literature on the surgical separation of craniopagus in general. The most impor-

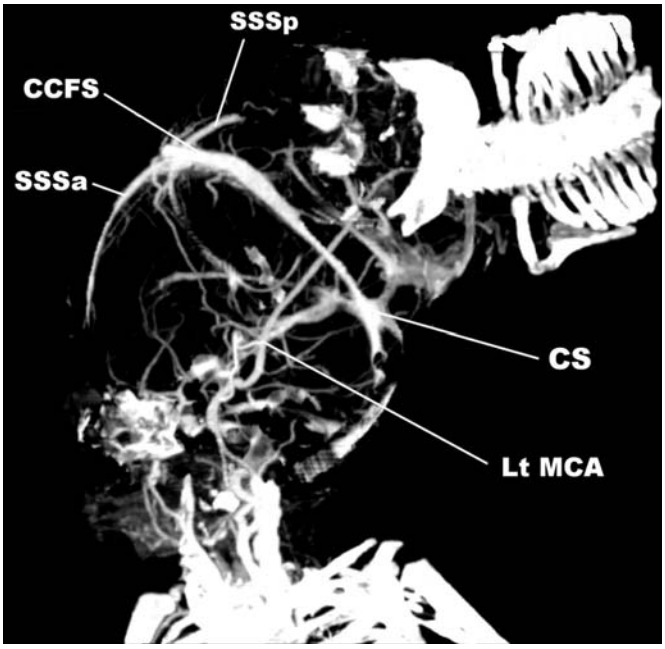


**FIGURE 3.** *CT angiogram of the craniopagus parasitic twins. The left middle cerebral artery (*Lt MCA*) of the autosite is enlarged and anastomoses with the right middle cerebral artery of the parasite. It carries blood from the autosite to the circle of Willis of the parasite and then to the rest of its body. The anterior third of the superior sagittal sinus of the parasite (*SSSp*) joins the anterior third of the superior sagittal sinus of the autosite (*SSSa*) to form the circumferential sinus (*CCFS*). The circumferential sinus ends in the confluence of sinuses (*CS*).*

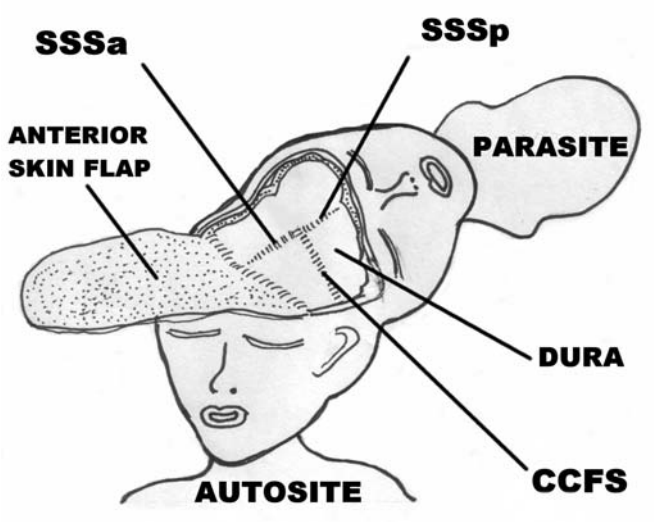


**FIGURE 4.** *Illustration showing the early steps of craniopagus parasiticus surgical separation with the twins in the supine position. A large skin flap, taken mostly from the parasite, has its broad base at the forehead of the autosite. Free bone flaps are removed. The superior sagittal sinus of the autosite (*SSSa*) and that of the parasite (*SSSp*) are seen joining the circumferential sinus (*CCFS*).*

tant cause of failure in published cases was death of the patient during surgery. This occurred most commonly because of bleeding from the sinuses and major veins at the end of the operation (7). By that time, the members of the previous neurosurgical teams were exhausted and improper decisions may have been made.

A meeting was held with the medical team and the parents to discuss the condition of the twins. The parents, after considering the options, asked for surgical intervention. Because the operation would lead to death of the parasitic twin, religious advice was sought and an approval was obtained from Al Azhar, the highest Islamic organization in Egypt, to perform surgery to save the complete twin's life.

## Surgical Procedure

On the morning of February 18, 2005, the twins were taken from the intensive care unit at Benha Pediatric Hospital to the operating room for surgical separation. An artificial dural graft was prepared for the subsequent closure of the dural defect. The twins were initially put in the supine position. The first step was to outline a large skin flap, prepared mostly from the scalp of the parasitic twin and based on the scalp of the normal twin. The flap was made by making an incision just above the eyebrow of the parasitic baby (*Fig. 4*). The flap included the skin and the periosteum to ensure good vascularity and was made as wide as the position of the patient could permit. This, together with two additional flaps, provided total skin coverage of the defect of the autosite. It also obviated the need for preoperative tissue expansion.

Multiple bone flaps were removed from the parasitic side up to the junction of the twins. These flaps were planned to cover the forthcoming defect in the autosite after separation. An incision was made in the left frontal dura of the parasite just above the level of the eyebrows. A small incision was made in the dura of the right frontal lobe of the parasite just lateral to the beginning of the superior sagittal sinus. The superior sagittal sinus was then under-run by two ligation sutures and was cut between them; the falx was then cut. The left hemisphere of the parasite was separated from the dura. At that point, massive venous bleeding occurred, which was stopped by cottonoid packing. The dural incision was carried laterally to the right temporal area of the parasite and the dura was reflected towards the autosite. The Sylvian fissure was exposed and dissected. The right middle cerebral artery of the parasite was followed to its anastomosis with the left middle cerebral artery of the autosite, and the middle cerebral artery of the parasite was clipped by four silver clips and cut. The anterior part of the right hemisphere of the parasite was separated from the anterior part of left cerebral hemisphere of the autosite. Cutting was done by bipolar coagulation encroaching on the parasitic side for approximately one centimeter. Finally, the cottonoid used to pack the bleeding from the left hemisphere of the parasite was removed and venous bleeding was stopped by bipolar coagulation. The field was packed with cottonoid, and

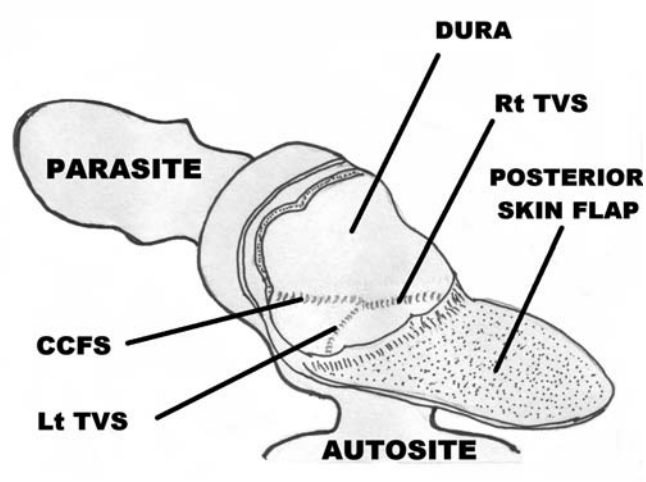


**FIGURE 5.** *Illustration showing the second stage of surgical separation with the twins in the prone position. A large posterior skin flap, taken mostly from the parasite, has its base at the occipital region of the autosite. Free bone flaps are removed. A bridge of bone with overlying skin is left connecting the two heads until the end of surgery. This helped to avoid the twisting of soft structures when the position of the twins is changed. The circumferential sinus (*CCFS*), right transverse sinus (*Rt TVS,*) and left transverse sinus (*Lt TVS*) are seen through the dura.*

the skin flap was closed with continuous silk suture. The incision was covered by sterile drape.

The babies were then put in the prone position, and a new operative field was prepared and draped. A skin flap was made with its free end above the nucheal line of the parasite (*Fig. 5*); multiple free bone flaps were also made. A small part of skin and bone connecting the left parietal area of the autosite to the right parietal area of the parasite was left continuous to act as a splint between the two heads. This avoided twisting when the position of twins was changed. The dura over the occipital lobe of the left hemisphere of the parasite was incised. The posterior part of the left hemisphere was separated from the dura. The dura of the posterior fossa of the parasite was then opened. The left transverse sinus of the parasite was closed using sutures. Separation of the joined cerebral hemispheres was achieved through an incision of the dura on the right hemisphere of the parasite. A trial to extend the dural incision towards the circumferential sinus resulted in profuse bleeding. Progress was slow, with moments of difficulty controlling bleeding points. Titanium clips were frequently applied over pieces of Surgicel (Ethicon, Inc., Somerville, NJ) to prevent laceration of the wall of blood lacunae. Most of the sinuses and blood lacunae in the parasitic twin were ligated, with the exception of the major draining sinus of the parasite. A left occipital lobectomy of the parasite was performed in hopes of providing better access to the area of the major draining sinus; this attempt was unsuccessful. The incision was packed with cottonoid and the skin was closed with a continuous silk suture. Sterile drape was used to cover the sutured incision.

The babies were rolled back into the supine position, and the incision was cleaned and redraped. The anterior skin flap was reopened. A third small skin flap was made from the skin overlying the bone bridge between the two crania. The last piece of bone joining the two crania was then cut, and the parasite was retracted slowly away from the autosite. The separation of the conjoined cerebral hemispheres was completed. A clamp was placed across the remaining area of the falx, including the major draining sinus. The parasitic child was then separated from the complete child and the clamped area was oversewn.

An artificial dural graft was sutured to the edges of the dural defect. To fix the flap by sutures, holes were drilled near the edges of the bone flaps, which were then used to cover the bone defect. Extradural and subgaleal drains were inserted to divert any cerebrospinal fluid while the wound was healing, as advised by Bucholz et al. (3). The redundant skin was excised and the wound was closed.

The duration of the surgery was 9 hours, the last 2 hours of which were spent mostly repairing the cranial defect. Hemostasis consumed a considerable portion of the operative time. The child received four liters of blood to replace the inevitable blood loss.

## Postoperative Condition

The child was weaned from the ventilator and extubated. Forty-eight hours after surgery, she was alert and paid attention to light and sounds. The cranial nerves showed no deficit. Her eyes were spontaneously opened, moved freely, and could follow light and sound. The child had no facial affection. The glabellar, grasping, and pupillary reflexes were normal. There were very slight hypertonia and hyper-reflexia in both lower limbs compared with her upper limbs. Nevertheless, the child could move her four limbs freely. There were no tremors, fasciculations, or abnormal movements. No sensory deficit was detected, and the child reacted to painful stimuli on all parts of her body.

An important postoperative complication we witnessed in our patient was early seizure activity; this was controlled by phenobarbitone and phenytoin. The second problem was an elevation of the liver enzymes, which was probably a result of prolonged anesthesia, blood transfusion, and antiepileptic drugs. This was controlled adequately with liver supports. Hydrocephalus was another postoperative complication for which a ventriculoperitoneal shunt was inserted on postoperative Day 10 (*Fig. 6*). The shunt, however, became obstructed and was replaced by a new one on July 18, 2005, 5 months after surgery. At the age of 18 months, the child developed pneumonia and was hospitalized for 20 days.

One year after operation, when the child was 22 months old, her neurological condition was the same. Her developmental milestones were lagging behind normal. She was able to cry and speak in monosyllables, could sit with her head erect, had a social smile, and recognized her mother. She was still on two antiepileptic drugs.

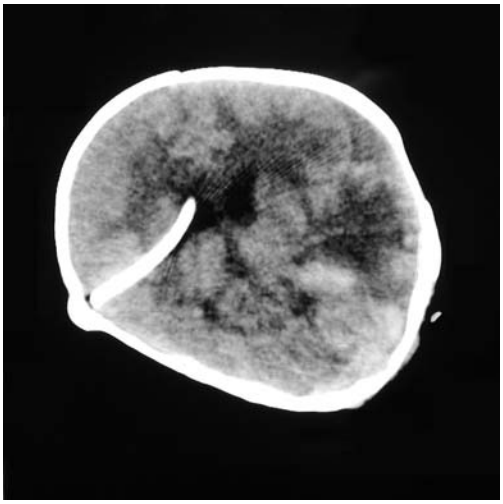

**FIGURE 6.** *CT scan of the brain obtained 2 weeks after surgery.* A *ventriculperitoneal shunt is seen with its tip in the frontal horn of the right lateral ventricle. The left hemisphere of the brain shows postoperative edema from surgical manipulation.*

## DISCUSSION

### Present Case Report

Craniopagus parasiticus is an extremely rare anomaly and the present case is the ninth reported. All cases have the feature of two conjoined heads, but with one of the twins, the parasite, being underdeveloped (1). The crucial feature of this anomaly is that the parasite has a grossly anomalous heart that is incapable of providing a separate circulation. Accordingly, the parasite receives all of its blood supply through the area at which it is connected with the normal twin. The arterial blood supply to the parasite may come from the internal carotid artery (2) or the posterior cerebral (1) artery of the autosite. In the present case, the main supply to the parasite was through the middle cerebral artery of the autosite (*Fig. 3*), and the venous return was via the dural sinuses.

### Surgical Intervention

The literature on the surgical separation of craniopagus parasiticus is very poor. This is owing to the fact that this anomaly is extremely rare (only eight cases have been recorded) and because most of these twins have traditionally been stillborn. The first attempt to separate such twins was performed in 2004 by a team led by Lazareff (6) in the Dominican Republic. Their patient, a young girl, underwent operation at the age of 4 weeks. The operation lasted 11 hours and the child was given 15 L of blood to replace the blood lost during the operation. The child, however, died approximately 7 hours after surgery. It is unfortunate that the information about this case has been obtained from the internet. To our knowledge, that case has not been published in a journal.

The present patient underwent operation on February 18, 2005 at the age of 10 months. Reviewing the literature for the ideal time for craniopagus surgery, we found that successful separation was commonly achieved when the twins were between 2 and 15 months old (8). We decided to use this range as a guide in the present case, but there were two other factors also taken into consideration. The twins had heart failure and were given treatment until the time of surgery. The twins' parents were also hesitant in giving their approval for surgical separation.

Surgery for craniopagus parasiticus should differ from that for cases in which both twins are equally developed. First, the aim of surgery in the parasitic cases is to preserve the developed twin by sacrificing the parasitic. The parasitic twin is naturally a good donor of skin and bone to the autosite; accordingly, the separation could potentially be accomplished with a one-stage surgery. In the present case, the fact that the parasitic twin was much smaller in size and weight compared with the autosite made it possible to use a normal surgical table rather than a special one. Because the parasitic twin is sacrificed, surgical division of the brain tissue should always encroach 1 cm in the parasitic twin. We think that this policy made it possible to perform separation without postoperative neurological deficit.

Craniopagus parasitic twins have only one functioning heart, which carries the burden of supplying blood to both twins. During surgical separation of the parasitic twins, bleeding is expected to be extensive because all the blood that goes to and returns from the parasite has to traverse the connecting zone between the two heads.

During surgery, bleeding occurred mostly from the dural sinuses and venous lacunae. The arteries were clipped first; then, the venous sinuses were adequately controlled by bipolar coagulation, surgical foam, and clipping. On many occasions, when clips were applied, the dura was torn and subsequent return of venous ooze resulted. We found that a better method to secure bleeding is to apply the clips over pieces of Surgicel to provide support to the friable wall of the sinuses and lacunae. The time interval between ligation of the arterial circulation and separation of the two heads should not be very long to avoid a possible accumulation of toxic material which might affect the autosite. In the present case, the interval was 4 hours

## CONCLUSION

Craniopagus parasticus is an extremely rare anomaly. The present case is the ninth to be reported in the literature. Many such twins have traditionally been stillborn. Surgical separation is best attempted when the twins are between 2 and 15 months. The operation should not be delayed to allow for normal physical and psychological development of the autosite. Accurate preoperative planning is very important, and bleeding from the dural sinuses is the most dangerous cause for surgical failure. The arterial supply to the parasite must be ligated first and then the dural sinuses can be dealt with. Adequate skin and bone flaps taken from the parasitic twin help in closing the surgical defect in a one-stage operation. In the present case, no postoperative neurological deficit was observed after the oper-

ation. One year after surgery, the girl was still alive and in reasonably good health.

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## COMMENTS

The more common occurrence that is classically described and the subject of much media attention is craniopagus twins, a condition in which both children are complete individuals, viable but joined at the head. Although potentially sharing cerebrovascular supply or venous drainage, the rest of the body and brain are separate and functional. In this rare syndrome, craniophagus parasiticus, described by the authors, one of the conjoined twins is underdeveloped and depends on the cardiovascular and physiolgical support from the normally developed twin. In this report, the authors describe the first case of a successful operative detachment of the parasitic "twin" allowing the normal child to survive and be neurologically intact. A previous attempt at separation resulted in non-survival after surgical intervention. The lessons learned by the surgical team from those earlier procedures assisted them in successfully separating the two in this case, leaving a viable and neurologically functional child. The lessons learned include surgical separation at an older age to allow for greater tolerance of the stress of surgery and blood loss, the development of adequate vascularized skin and bone flaps from the parasitic twin for adequate closure of the surgical defect in one stage, and the early ligation of the arterial supply to the parasite to reduce the intraoperative blood loss. Although successful for the first time for survival and "normal" neurological function, this surgery did result in "neurological" complications and impairment in that there was postoperative hydrocephalus and a seizure disorder requiring ventriculoperitoneal shunting and anti-seizure medication.

**P. David Adelson**
*Pittsburgh, Pennsylvania*

As the authors indicate, this is a rare anomaly for which they could find only one previous reference to attempted operative treatment. Vascular considerations were of paramount concern, particularly as related to the venous system. In this case, computed tomographic angiography was a useful alternative to catheter angiography and its attendant risks in the very young patient with a complicated medical condition. The arterial relationships were straightforward and did not present a particular problem because the parasitic infant obtained its effective arterial supply via conjoined middle cerebral artery branches. The venous circulation, on the other hand, was rather more complex and had to be properly appreciated preoperatively. Avoidance of venous insufficiency or infarction was, of course, one concern. Of greater importance was the potential for catastrophic venous bleeding at every turn in the operation. The authors are to be commended for their skill and perseverance in successfully dealing with this problem, which was essential for a successful outcome.

**Paul H. Chapman**
*Boston, Massachusetts*

This case report is a fascinating account of the separation of a cranial "parasite" from its more normal "twin." The authors' inclusion of their preoperative ethical conferences and their seeking of approval from the highest Islamic authorities in their country are another item of interest in this report, and a striking contrast to how such decision making is carried out in the United States in such cases. The operative procedure, described in detail is, by nature of the case, difficult to follow, but the authors were aided in their dissection by the knowledge that the parasitic twin would be sacrificed. As in every discussion of such surgical exploits, the long-term outcome of the patient is given short shrift. However, the authors are more frank than most similar reports, indicating that their patient is developmentally delayed, on two seizure medications, has already experienced pneumonia requiring hospitalization, and has shunted hydrocephalus. I also suspect that, in Egypt, there was much less of a circus atmosphere than attends similar cases in the Western hemisphere.

**R. Michael Scott**
*Boston, Massachusetts*

The authors have correctly concluded that, because of the rarity of craniopagus parasiticus, each case should be of significant interest to the medical community. They have done an admirable job of presenting a description of the surgical technique used for this successful separation. Among the many fascinating aspects of this case is the fact that seven of the eight previous cases of craniopagus parasiticus they referenced involved males. Generally, there is about a 2:1 incidence of female over male predominance in craniopagus twins.

It would be easy for readers to conclude that this case should have been done more easily because there was no need to try to salvage the parasitic twin. As was pointed out by the authors quite accurately, the venous sinuses can behave in a very abnormal way and tend to be quite friable. They wisely decided to cut through the parasitic brain leaving a margin of about 1 cm to avoid as much contact with the abnormal sinuses as possible. Unfortunately, they still encountered them, along with the expected significant hemorrhage. Having been involved in five cases of craniopagus and recognizing how complex and abnormal not only the vasculature, but also the dural reflexions, can be in these cases, I think this case would bolster the position of removing even more of the brain of the parasite circumferentially to skeletonize the abnormal vasculature and dural reflexions, while leaving a significant cuff of the parasite's tissues against the shared vasculature and the brain contact points. The techniques used to do this would not vary significantly from techniques used in the decortication and disconnection techniques used in cerebral hemispherectomy. The idea of pre- or intraoperative embolization of the arterial supply first and then the venous supply, might allow future operations of this type to be done even more quickly and safely. In summary, I think this is a valuable contribution to the literature and will provide more information to others as these challenging cases are tackled in the future.

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