James T. Goodrich, MD, PhD, 1946–2020: a historical perspective and his contributions to craniopagus separation

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Dr. James Tait Goodrich was an internationally renowned pediatric neurosurgeon who pioneered the neurosurgical procedures for the multistage separation of craniopagus twins. As of March 2020, 59 craniopagus separations had been performed in the world, with Goodrich having performed 7 of these operations. He was the single most experienced surgeon in the field on this complex craniofacial disorder. Goodrich was a humble individual who rapidly rose through the ranks of academic neurosurgery, eventually serving as Director of the Division of Pediatric Neurosurgery at the Children's Hospital at Montefiore Medical Center in the Bronx, New York. In this historical vignette, the authors provide context into the history of and sociocultural attitudes toward conjoined twins; the epidemiology and classification of craniopagus twins; the beginnings of surgery in craniopagus twins; Goodrich's neurosurgical contributions toward advancing treatment for this complex craniofacial anomaly; and vignettes of Goodrich's unique clinical cases that made mainstream news coverage.

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INTERNATIONALLY renowned pediatric neurosurgeon, Dr. James T. Goodrich, was a caring, empathetic individual and a pioneer in the field. Prior to his untimely and unfortunate passing on March 30, 2020, due to the novel coronavirus disease, 2019 (COVID-19), he led a highly accomplished and dedicated academic neurosurgical career over the years, advancing many techniques for complex craniofacial anomalies, brain reconstruction, and neuronal regeneration. Most notably, his most significant contribution to the field was the multistaged surgical protocol for the separation of craniopagus twins, which is one of the most complex neurosurgical procedures. As a result of Goodrich's passion for and contribution to the field of craniofacial anomalies, he soon became a well-known lecturer in the field and published numerous peer-reviewed journal publications and book chapters regarding craniofacial anomalies. As of March 2020, 59 craniopagus separations had been performed in the world; he performed 7 of these

operations and became the single most experienced surgeon on this disorder. This historical perspective provides context into the scientific, medical, and clinical advancements made in craniopagus twin surgical separation prior to Dr. Goodrich, along with his key clinical contributions and surgical outcomes.

History of Craniopagus Twins

It is no accident or surprise that Dr. Goodrich took a fascination in both the medical history of the Old World and surgical management of conjoined twins. In his article "Medical and surgical practice as represented in cultural figures from the pre-conquest Mesoamerican territories," he reviewed terra-cotta, stone, and stela figures from Olmec and Mayan civilizations in order to illustrate various depictions of diseases and information related to cranial deformation and early neurosurgical practices from pre-conquest Mesoamerica.¹ Ironically, a finding was that

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Mayan terra-cotta figures commonly depicted conjoined twins. Dr. Goodrich had a unique gift for unearthing and relating unique treasures of the past, and was one of the most prolific medical historians the neurosurgical profession has ever witnessed. As we detail in this review, Goodrich was inspired by the complex and intricate history of craniopagus twins, along with the original and current leaders in his time, to make progress toward providing these “oddities of nature”² with a seemingly normal life.

Conjoined twins are a medical anomaly in the truest sense. The history and origins of this medical rarity or peculiarity make for fascinating conversation because humans by their very nature are separate living entities with their own physical and emotional well-being. There are 4 main classifications of twins conjoined at the head based on their anatomical connectivity: 1) cephalopagus (i.e., connected ventrally); 2) parapagus diprosopus (i.e., connected laterally); 3) rachipagus (i.e., connected dorsally); and 4) craniopagus (i.e., connected along the calvaria solely, or any place along the cranial vault, which is unique to this classification of conjoined twins, and is the topic of this review). Craniopagus twins can be connected in an unlimited number of ways. Additionally, they have separate thoracic and abdominal cavities, but may asymmetrically share portions of their cortex, venous sinuses, and meninges.

The first documented case of craniopagus twins originated in Bavaria, Germany, and were illustrated as “Ein monstrum” (or a monster), depicting a cautionary or warning signal from God.² These twins, born in 1491 close to Worms, Germany, were frontal craniopagus twins and were documented to have been alive for 10 years.² Other later historical works have documented craniopagus twins in the 16th century by Ambroise Paré,³ in the 18th century by Sir Everard Home,⁴ and in the 19th century by August Förster.⁵ Additionally, in the 19th and 20th centuries, traveling “sideshows” popularized the notion of Siamese twins, such as Chang and Eng, who profited from being in the public eye.² Moreover, exhibitions of these “freaks of nature” were often used to pay for their medical bills, as was the case for Yvonne and Yvette McCarther in Los Angeles during the 1950s–1980s.²

Epidemiology and Classification of Craniopagus Twins

Craniopagus twins are an exceedingly unusual phenomenon (0.6 in 1,000,000 live births⁶) that takes place in approximately 2%–6% of all conjoined twin cases, with a 4:1 female/male ratio. On a review of the literature, only 22 cases were reported through 1987.⁷ They present at a few different junction sites along the calvaria. O’Connell originally proposed classifying craniopagus twins as “partial” or “total” based on the quantity of surface area shared between the twins.⁸ Bucholz and colleagues proposed further classifying O’Connell’s “total” craniopagus twin definition based on combinations of connectivity at the frontal, temporal, parietal, and occipital regions.^{8,9} Parietal (or vertical) craniopagus twins can be further subclassified by their angle of rotation, with type I (i.e., facing identical directions) (Fig. 1), type II (i.e., facing contrast-

FIG. 1. Example of O’Connell type I vertical craniopagus twins. These female craniopagus twins were 5 months old and were born in September 1995, and they were treated at Primary Children’s Medical Center at the University of Utah by Dr. Marion L. Walker and colleagues. Modified from Browd et al: *J Neurosurg Pediatr.* 2008;1(1):1–20. Copyright AANS.

ing directions), and type III (i.e., intermediate degree of rotation) variants. One group proposed that postoperative survival is associated with axial orientation of the junction,¹⁰ and another group proposed that the anatomy of the dural venous sinuses predicts postoperative survival—with “total” (greater sharing) having worse survival (at approximately 48%¹¹).⁷ In addition to asymmetrical cranial and brain abnormalities, cardiac, craniofacial, pulmonary, renal, genitourinary, and colorectal abnormalities may present.⁸ If the twins survive past the perinatal period (40% stillborn rate¹¹ and 33% death rate during the perinatal timeframe¹¹), surgical separation is possible for approximately 25% of cases.^{12,13} Moreover, surgical separation is absolutely necessary for a possibility of long-term survival because 80% of craniopagus twins may succumb to underlying medical complications by 2 years of age, and more than 90% will not live past 10 years of age.² Surgical separation must take into account gravitational forces, air embolism, increases in venous pressure, bleeding, cerebral edema, and adrenal dominance.¹⁴

Beginnings of Surgery in Craniopagus Twins

In the modern era, prior to Dr. Goodrich, separation of craniopagus twins was done in a single lengthy operation, which previously caused severe issues in forming collateral circulation, along with high rates of morbidity and mortality. Stone and Goodrich performed a literature review and found that the pooled mortality rate across 41 cases of craniopagus twin separation was significantly lower for staged surgery at 23% compared to a single surgery at 63%.¹¹ Looking back in history, the first reported craniopagus separation occurred in 1928 and was published in *The Lancet* by H. C. Cameron, who unsuccessfully separated 12-day-old twins with parietal connection.¹⁵ Other reports subsequently followed that were also largely unsuccessful following experimental craniopagus twin separation.^{16–18} Nearly 3 decades passed following this original report until the first successful separation of

craniopagus twins was reported in *JAMA* in 1953 by H. J. Grossman and colleagues; they separated 14-month-old twin boys with parietal connection in 1952.¹⁹ Additionally, the same group published another report in *Transactions of the American Neurological Association* in 1953.²⁰ The boys, Roger and Rodney Brodie, were treated by Dr. Oscar Sugar in 5 surgical stages, with the final 10-hour operation necessitating 6000 ml of whole blood transfusion and occurring on December 17, 1952. At the time of that operation, this was the longest-duration pediatric surgery, and required the greatest amount of blood transfused.¹¹ However, neither of these twin boys survived long term postoperatively, with Roger dying 34 days and Rodney dying 11.5 years postoperatively due to hemorrhage and hydrocephalus, respectively. Achieving successful outcomes for these patients was largely limited by a lack of both anatomical understanding (i.e., poor imaging modalities) and robust surgical strategies and techniques.

The long-term survival of craniopagus twins was not described until 1957 in the *Journal of Neurosurgery* by H. C. Voris and colleagues; they reported on the separation of 7-month-old twin girls with limited parietal connection.²¹ The girls both survived long-term (> 28 years) even though they endured complications such as hemorrhage and neurological injury.⁷ From the 1960s to the 1970s, surgical staging was modeled after the efforts of Dr. Oscar Sugar, in which he used stepwise venous vasculature clipping and ligation, and placed polyethylene sheets to delineate his separation plane; additionally, in 1974, the operating microscope was first used in craniopagus surgery.²² However, after these operations, complications in separation limited the possibility and success of long-term survival for these patients.^{8,10,23,24} Dr. Ted Roberts from Salt Lake City, Utah, was acknowledged by Dr. Goodrich² as a pioneer in the field during the 1980s for successful staged separation of type I and II total vertical craniopagus twins with long-term survival.^{25–27}

Moreover, staged separation of craniopagus twins was described by G. Drummond and colleagues in 1991 in the *British Journal of Plastic Surgery*, where they reported on the separation of craniopagus twins in stages by using a screw clamp to slowly close the connecting vein; however, long-term survival was unknown.²⁸ Following this report, other studies gradually demonstrated improvement and successful separation of craniopagus twins through use of improvements in neuroanesthesia, intensive care, neuroradiology, and surgical instrumentation/tools (e.g., tissue expanders and rotating operating tables).²⁹ During the 1980s to early 2000s, a single lengthy marathon operation of 22–100 hours became popular.^{30–37} However, Goodrich noted¹¹ that these lengthy single-stage operations may be no more efficacious than the prior multistage resections performed by Drs. Sugar, O'Connell, and Roberts.^{19,20,23–26,38} Therefore, Goodrich focused his efforts on developing a robust classification tool (Table 1)² and pioneering a modern multistage surgical separation protocol.

These initial reports of craniopagus separations inspired Dr. Goodrich's techniques, in which he focused on a multistage separation that included appropriate timing between operations (i.e., months and not days) for complete recovery. Moreover, Goodrich was an advocate of proper

TABLE 1. Proposed craniopagus classification scheme*

Characteristic	Score
Scalp	
Minor surface area shared (<10 cm ²)	1
Major surface area shared (>10 cm ²)	2
Calvaria	
Minor surface area shared (<10 cm ²)	1
Major surface area shared (>10 cm ²)	2
Dura mater	
Independent dural envelope surrounds cortex	1
Dura is shared along 1 or more planes	2
Neural tissue	
Completely separate	1
Interdigitated, but not fused	2
Minor areas of fusion (<5 cm ² total surface area)	3
Major areas of fusion or involvement of eloquent cortex (>5 cm ² total surface area)	4
Arterial connections	
None	1
Minor feeding branches (M ₄ , A ₄ , P ₄)	2
Distal branches (M ₃ , A ₃ , P ₃)	3
Proximal branches (M ₂ , A ₂ , P ₂)	4
Major vascular trunks (M ₁ , A ₁ , P ₁ , ICA)	5
Venous connections	
None	1
Separation of major sinuses w/ minor shared draining veins	2
Shared along anterior 1/3 SSS	3
Shared distal (transverse/sigmoid sinus)	
Shared along posterior 2/3 of SSS w/o involvement of torcular herophili	4
Shared along posterior 2/3 of SSS w/ involvement of torcular herophili	5
Deep venous drainage	
Present	1
Absent	2
CSF (ventricular anatomy)	
Separate ventricular systems	1
Shared ventricular system	2
Venous outflow	
Ipsilateral	1
Contralateral (crossed)	2
Arterial flow	
Ipsilateral	1
Contralateral (crossed)	2

ICA = internal carotid artery; SSS = superior sagittal sinus.

* Higher score equates with more difficult separation. Minimum score = 10; maximum score = 28. Goodrich used this tool to stratify patients based on surgical risk and potential for separation success. Reproduced from Browd et al: *J Neurosurg Pediatr*. 2008;1(1):1–20.

physical and occupational therapy prior to operation and in between staged procedures to allow for vascular supply equilibrium, ameliorating venous drainage, facilitating improved scalp reconstruction, and fewer medical complications and neurological deficits associated with surgery.²⁹ He aimed that through these approaches he could avoid drastic brain damage to one of the twins (i.e., typically the one not sharing the majority of the vascular supply).

Goodrich's Scientific Contributions to Craniopagus Surgery

Dr. Goodrich was a true neurosurgeon-scientist and contributed many scientific advancements toward novel surgical approaches for separating craniopagus twins. Additionally, he was a visionary in terms of spearheading a multistage surgical approach for the separation of craniopagus twins. He authored many peer-reviewed articles and was an international lecturer in this highly specific field. His first publication in 2004 in *Child's Nervous System* titled "Craniopagus twins: clinical and surgical management"³⁹ and subsequent publication in 2005 in *Clinics in Plastic Surgery* titled "Separation of craniopagus conjoined twins: an evolution in thought"²⁹ described the case of 2-year-old total vertical (15° rotation) craniopagus twins born on April 21, 2002, to a 29-year-old mother via cesarean section. An abortion was offered to the mother when the anomaly was found via a level 2 ultrasound; however, religious reasons deterred her from pursuing that option. Therefore, the twins underwent a 4-stage operation over 9.5 months for separation.^{29,39}

In these publications he detailed the remarkable surgical ability to separate craniopagus twins, along with the preoperative and operative management strategies for such separation. The workup done at the twins' local facilities (outside the United States) described them as healthy with no medical conditions; however, photographs suggested otherwise. Upon examination in the United States, the twins were found to have chronic medical conditions due to positioning on their backs for nearly their entire 20-month life: they were underweight; had abnormal blood pressures (twin A had a blood pressure of 210/125 mm Hg and twin B had a blood pressure of 60/40 mm Hg); aspiration pneumonia (with fever); tachycardia; minor urological abnormalities; and neurological deficits (i.e., intellectual, speech, and motor problems [hemiparesis]) that were due in part also to social isolation. To ameliorate these medical conditions, which was necessary prior to staged separation, the twins were administered a series of blood pressure medications and put on feeding tubes with repositioning to allow for proper nutrition and resolution of aspiration pneumonia, respectively. Additionally, per Goodrich's recommendation and advocacy, physical and occupational therapy provided at Blythedale Children's Hospital in Valhalla, New York, improved their medical conditions prior to the surgical operations.

Radiology workup allowed for 3D depictions and holograms (via craniofacial CT and MRI/MR venography) of the arterial and venous circulations, which permitted visualization of abnormal venous circulation patterns, with one twin dominating the venous vascular supply over the

other. This determined the need for a staged separation to allow for rerouting of venous vasculature from one twin to the other, thus establishing proper venous collaterals, and avoiding serious complications such as venous stasis and stroke. These 3D depictions inform the physicians on the extent of bone fusion and allow for creation of an acrylic 3D model. With this model, it allows the surgeons preoperatively to prepare visualization of the surgical corridor and separation location, and organize the anticipated calvaria reconstruction.² Goodrich typically used the company Medical Modeling, LLC, to construct these models. Overall, Goodrich strongly favored using advanced high-resolution CT, MRI, MR venography, angiography, and tractography to ascertain information regarding bone architecture, brain formation/structure, vascular architecture, and anatomy of white matter pathways. Intraoperatively, stereotaxic sequences of multiple imaging modalities can be co-registered to map surgical trajectories. However, he acknowledged their limitations in the potential inability of MRI to discern a conjoined brain, specifically the cortex.

Moreover, this landmark paper discussed the anesthesia issues (i.e., each twin has different circulations and thus responses to medications); operating room issues (i.e., need for a large operating room, positioning of the twins, and surgical personnel needed); and staged-surgery separation issues (i.e., need to form collateral circulation and avoid neurological deficits and bleeding).³⁹ Last, Dr. Goodrich was mindful of the final reconstruction needed and the collaborative efforts between plastic surgery and neurosurgery to allow for proper scalp soft-tissue quantity to avoid CSF leakage, hydrocephalus, and/or meningitis.

From Dr. Goodrich's perspective, the surgical staging for craniopagus twin separation was necessary to separate conjoined fused brains, which need time to redevelop proper brain vasculature and circumvent serious neurological complications that could occur during a single marathon operation. The timing of the stages would be unplanned (open-ended) and would be up to the discretion of the surgeon when proper recovery was achieved. The unique aspect noted by Goodrich that is distinct from a single-stage approach is that the multistage approach requires tissue expansion to be appropriately planned, timed, and designed.²⁹ Goodrich advocated for tissue expanders to be placed during an independent operation performed 4–6 weeks prior to the penultimate separation stage to circumvent infection risk.² To potentially avoid infections over the multiple stages, Goodrich was keen on proper draping, antibiotics (intra- and postoperatively), and limited use of drains or indwelling catheters; however, infections were experienced and he treated them with antibiotics.² The surgical staging for the separation of the boys from the Philippines took place in 4 stages, with silicone dissection sheets (Fig. 2) used in between stages to avoid scarring and fusion of the dissection plane. Two separate anesthesia teams (Fig. 3) were used to observe and treat each twin distinctly, especially in the fourth (and final) stage, in which the operations become independent. Goodrich recommended that at the initial stages 1 team of surgeons may be used; however, during the final stages 2 neurosurgeons and 2 plastic surgeons were advised.

FIG. 2. Illustration of the use of silicone (Silastic) dissection sheets between surgical stages in order to avoid scarring and fusion of the dissection plane. Reprinted from Browd et al: *J Neurosurg Pediatr.* 2008;1(1):1–20. Copyright AANS.

The first stage was performed on October 20, 2003, with removal of a large frontal midline sagittal sinus via craniotomy in one twin. Approximately 1 month later, on November 24, 2003, the second stage was performed, which involved removal of a large posterior sagittal vein via craniotomy in the same twin. Approximately 3 months postoperatively from the second stage, the third stage was performed on February 20, 2004, and involved further ligation and division of venous bridging veins. Approximately 6 months postoperatively from the third stage (9.5 months postoperatively from the first stage), the fourth (and final) stage was performed on August 4, 2004, and separation of the twins was accomplished with division of the residual dura mater and veins. Of important note,

FIG. 3. Photograph demonstrating 2 separate anesthesia teams used during craniopagus twin separation surgeries. These female craniopagus twins were 5 months old and were born in September 1995, and they were treated at Primary Children's Medical Center at the University of Utah by Dr. Marion L. Walker and colleagues. Modified from Browd et al: *J Neurosurg Pediatr.* 2008;1(1):1–20. Copyright AANS.

the physiological issues noted previously (i.e., blood pressure and urological abnormalities) were ameliorated as the staging procedures progressed. For the first through third stages, the twins had inpatient hospital stays of between 3 and 4 days. For the fourth stage, the twins were discharged 21 days (3 weeks) postoperatively and underwent further feeding, physical, and occupational therapy to improve speech, motor, and neurological conditions. Further cranial vault reconstruction was required to allow for full scalp coverage.

In his publications, Goodrich was appreciative of his collaborations with his nurses, physical medicine and rehabilitation, pediatrics, anesthesiology, radiology, neurology, and plastic surgery colleagues in aiding proper care management for these craniopagus patients pre- and post-operatively. His collaboration on these procedures extended interinstitutionally as he worked with Dr. Marion L. Walker at Primary Children's Hospital in Salt Lake City, Utah, whose team had successfully operated on 3 sets of

craniopagus twins. Goodrich's other notable publications in this field described the history, epidemiology, novel comprehensive classification scheme, neuroimaging techniques, surgical indications, risk and complications assessment, advanced surgical approaches and operating room tactics, and other notable subsequent cases.^{2,11,40,41}

Goodrich's Clinical Outcomes Following Craniopagus Surgery

The case report published in *Child's Nervous System* in 2004 described the efforts Goodrich began in October 2003.³⁹ To summarize, Dr. Goodrich spearheaded a 10-month effort to separate craniopagus twins from the Philippines, Clarence and Carl Aguirre. These twins were conjoined vertically and shared a sizeable quantity of cranial neurovasculature. Goodrich implemented surgical staging over several months to fully separate the twins. Although the final operation proved difficult, Goodrich's preoperative surgical planning and imaging had made him keenly aware of the potential obstacles ahead, such as an unsuspected 2-inch fusion of posterior parietal lobe brain matter, through which he developed an "elegant" solution for division along the vascular pattern.⁴² On August 4, 2004, the separation of the twins was finalized following a 17-hour operation with a crew of 16 physicians. They reported no CSF leaks or meningitis. In one of Goodrich's articles he reviewed the case 3 years postoperatively and noted that Clarence developed seizures 1 year postoperatively, which resulted in delayed motor coordination.² Carl was the first twin to talk or walk, exhibited more aggressive behavior, and had a determination to learn. By 5 years of age, the twins were engaged in preschool activities within their school district. In a recent news media update, despite both twins surviving until the present day (they are 17 years old as of this writing), Clarence endures more neurological deficits than Carl. The mother of the Aguirre twins conveyed her gratitude and appreciation for Dr. Goodrich:

"I'm grateful and thankful for everything you've done to us especially for the boys Carl and Clarence. You gave me a greatest gift of seeing my boys separated and giving them a whole new lives. We will never forget you and you will always be part of our lives."

Despite Dr. Goodrich's rise to international fame, he remained extremely humble and cared deeply about the well-being of his patients. He continued to serve as a leader and innovator in his field, and in October 2016, famously directed a team of 40 physicians through a 27-hour operation to separate 13-month-old craniopagus twins, Jadon and Anias McDonald, who split a 5 × 7-cm piece of brain matter.⁴³ The operation was also significant due to the integration of 3D printing technology for fabricating skull caps and performing presurgery virtual planning.⁴⁴ The series of operations cost more than \$2 million and was documented by CNN and reported in a documentary titled "Separated: Saving the Twins."⁴⁵ Incredibly, despite an array of medical complications postsurgery, approximately 3 years since their final operative separation both twins are still relatively healthy, with Anias being neurodevelopmentally delayed compared to Jadon.

Conclusions

The recorded history of craniopagus twins dates back to the late 15th century, when it was seen as a warning from God representing an act of misfortune based on a variety of hypothesized reasons. Their surgical separation in the modern era began with the use of a single lengthy operation. However, Dr. Goodrich made significant key innovations in the surgical separation of craniopagus twins as he drew upon inspiration from the former pioneers and current leaders to become the field's foremost expert. His focus on a multistage separation with adequate planning and timing between stages, use of physical and occupational therapy to improve the health statuses of the twins, along with the use of 3D models, significantly improved these pediatric patients' outcomes.

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Disclosures

The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

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