THE UNSPEAKABLE HISTORY OF THORACOPAGUS TWINS' SEPARATION

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Surgery is the art, craft and science of miracles.
– Joan Cassell: Expected miracles, Surgeons at work (1991)

The incidence of conjoined twins is estimated at 1 in 50,000 births. Thoracopagus is the most common form of conjoined twins, with fusion from the anterior thorax to the umbilicus. They often present a common pericardial sac and sometimes, conjoined hearts. Approximately half are stillborn and a smaller fraction of pairs born alive have abnormalities incompatible with life. The condition is more frequently found among females, with a ratio of 3:1. Living thoracopagus twins rarely share a vital organ, except for the liver. In xiphopagus, the two bodies are fused mostly by the xiphoid cartilage.

HISTORY

The earliest known documented case of conjoined twin separation dates from the year 942, when a pair of conjoined twin brothers from Armenia was brought to Constantinople for medical evaluation. Leon Diakonos (950-992 AC) recalls that they had the same trunk from the armpits to the hips. Their members were proportionate and had no anomaly. When, at the age of thirty, they came back to Constantinople from where they had been chased away previously because their presence was considered a bad omen, one of the twins died suddenly. The surgeons decided to try to detach the body of the dead one. The scene is represented in a miniature of a Madrid Manuscript at the end of the 12th century, the Byzantine Chronicle of John Skylitzes (Figure 1). Apparently the initial result of the operation was successful; however, the surviving twin died three days after.

Figure 1: A Byzantine separation of a dead conjoined twin. (Codex Skylitzes Matritensis, fol. 131 (12th c.) Madrid National Library)

Since antiquity, and even up to recent times, these deformities were considered as monstrous and often displayed in fairs and circuses. They are described and pictured in a number of chronicles during the Middle Ages and belong to the bestiary of monsters of the famous surgeon of the Renaissance Ambroise Paré (Figure 2). He attributed the conjoined twins to an excess of semen, but he never advised to operate on them. For him, the

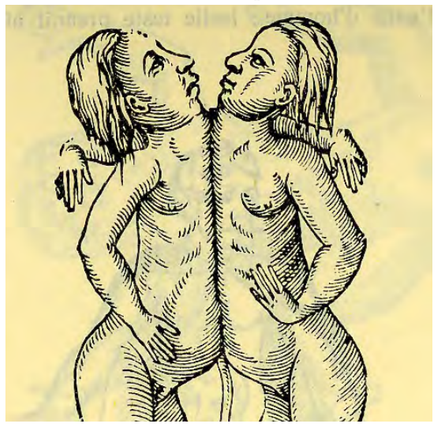

Figure 2: Thoracopagus represented by Ambroise Paré in the 16th century. (Paré A: Les oeuvres de chirurgie, Paris, Buon 1598)

Monsters differ from the Prodigious and the Mutilated in that they are creatures against nature and are often signs of some misfortune to come. His contemporary surgeon, Pierre Franco, however, refused to call them "monsters." They are God's creatures, and if possible they should be operated.

The French writer Montaigne living also in the same period, gives a detailed description of thoracopagus twins: "Two days ago I saw a child that two men and a nurse carried about to get money by showing it by reason it was so strange a creature. Under the breast it was joined to another child. . . ." Montaigne concludes: "Those that we call monsters are not so to God, who sees in the immensity of His work the infinite forms that He has comprehended therein. From His all wisdom nothing but good, common, and regular proceeds, but we do not discern the disposition and relation. Whatever falls out contrary to custom we say is contrary to nature, but nothing, whatever it be, is contrary to her."

Following the Byzantine operation, the first attempt to separate conjoined twins was recorded in 1689, on the omphalopagus girls Catherine Elizabeth by a German surgeon "with a sharp blade." The girls apparently survived. In 1700, the French naturalist Buffon recalls the story of the pygopagus Hélène-Julie, separated with a cautery by the surgeon Treyling, at the age of four. The two girls died immediately.

Nineteenth Century

During the 19th century, the most famous pair of conjoined twins was Chang and Eng Bunker (1811–1874). Thai-American brothers, born in Siam, Chang and Eng were joined at the torso by a band of flesh and cartilage at their sternum, with apparently fused livers. In 1829, the British merchant Robert Hunter "discovered" them and paid their family to let them be exhibited as a curiosity during a world tour. They travelled with the PT Barnum circus for many years and were labeled the Siamese twins. In 1935, the two brothers were examined by a number of scientists at the Academy of Science in Paris. Debates were mainly concerned with the nature of the junction, its origin and the particular psychology twins had developed, which fascinated the observers. It was the starting point for a think tank on teratological malformations and the capacities of surgery to correct them.

Upon termination of their contract with Hunter, the brothers successfully went into business for themselves and settled in a farm in Traphill, North Carolina. They bought slaves and adopted the name of Bunker. On April 13, 1843, they married two sisters: Chang to Adelaide Yates and Eng to Sarah Anne Yates. Their Traphill home is where they shared a bed built for four. Chang and his wife had eleven children; Eng and his wife had ten. In 1870, Chang suffered a stroke and his health declined over the next four years. On January 17, 1874, Chang died while the brothers were asleep. A doctor was summoned to perform an emergency separation, but he was too late. Eng died approximately three hours later.

When in Paris, an embryologist, Jean Victor Coste, had been in favor of the possibility of separating the Siamese twins, because, he said, "their viscera are probably free of any adhesion and an operation to divide them presents the better chances of success."

The famous French naturalist Isidore Geoffroi Saint-Hilaire had examined not only the Siamese twins, but also later on the twins of Prunay, Hortense-Henriette and Marie Louise, who were attached by their whole lower body as well as the monsters publicly exhibited like Millie-Christine or Rosa-Josepha united by their lower back with a single anus and vulva, in any case impossible to separate. Geoffroi Saint-Hilaire was however a fan of surgical operations for congenital malformations, in opposition to ineffectual medicine:

> 'For surgery, contrariwise, its benefit towards abnormal individuals is almost unlimited. Conducting useful unions, repairing unfortunate displacements, removing accessory and harmful parts, one can see that surgical operations sometimes give life to an individual, sometimes deliver him from organic flaws.' He agreed however that operable cases of conjoined twins 'must be and are in fact extremely rare.'

Toward the end of the 19th century, a number of living cases of conjoined twins had been recorded all over the world. A few surgeons had considered performing bold operations, but either the patients died prematurely, or their parents or they themselves, refused for fear of the complications or because they could count on their malformation to make their living by presenting themselves in circuses. However, between 1870 and 1881, three operations of separation took place. A German surgeon, Bochum, on his own daughters performed it right after birth in his private clinic. One of the twins apparently survived. In 1874, Lardier, a practitioner in Moselle (France), separated shortly after birth an incomplete parasitic child inserted in the epigastrium. The Medical Society of Nancy considered it as a premiere, but in fact, it was more like removing a tumor. In 1881, two Swiss surgeons, Biaudet and Bugnon, separated two three-month-old twins Marie and Adèle. One died immediately and the other a few days later. The doctors declared: "And now, what can we conclude from this unsuccessful procedure: that the operation of xiphopagus is impossible, that it is not justified; that in front of such a great and moving misery, nothing else can be done than crossing our arms? We don't think so."

Before their attempt, Baudet and Bugnon had in fact required the opinion of a famous teratologist, Camille Dareste, who had made a classification of congenital double monsters: the ones where the organs are not inversed and less interdependent, who are due to late fusion of the fetal bodies, would be more prone to an operation; the cases presenting a *situs inversus* (sign of early fusion, according to Dareste) should not be separated. He made also a distinction between the thoracopagus twins (intimate early fusion), where the operation should be "absolutely rejected," and the xiphopagus, for whom he encourages the surgeons to attempt a separation after a careful examination: "The progress of surgery and particularly the use of antiseptic methods allow today to attempt operations in cases in which we would have renounced before."

Twentieth Century

On the 30th of May 1900, 36-year-old Eduardo Chapot-Prevost operated the separation of Maria and Rosalina (Figure 3) in Rio de Janeiro. He had made before an exploratory laparotomy and tests with a radio-opaque bismuth compound to be certain that their digestive

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Figure 3: Maria and Rosalina in 1899

tracts were separate. It revealed, however, joined livers, and no conjoined twin operation thus far had successfully divided a shared liver. Although he was aware of Dareste's warnings he determined that they could be separated successfully, thanks to his experiments with dogs, which had shown that the liver healed rapidly as long as bleeding was controlled. The surgery lasted an hour and a quarter and was initially successful, but Maria developed an infection and died six days later. Rosalina, on the other hand, recovered quickly. She lived for many years after the operation, although she suffered from some paralysis in the left side of her face and body. In a 1964 interview, she recalls, "My earliest memory is that my sister and I were always squabbling, in spite of our affection for each other. We slept badly; one of us always wanted to turn over when the other didn't. It was the same when we walked; we always wanted to go in different directions. We ate off the same plate and wore a single dress specially designed by Mother. Our house was like a prison. We both longed to be separated, but in different ways. I longed for a successful operation, but Maria always feared she would never survive one."

Thanks to his spectacular operation, Chapot-Prevost became a national scientific hero and the Brazilian parliament allocated him credits to tour Europe and present this sensational "first case of living thoraco-xiphopagus operated at the age of seven." On the 9th of October 1900, he exhibited Rosalina at the Salpétrière in Paris, with pictures and x-rays, showing the inversion of the heart on the operated child, underlying its importance, considering Dareste's declaration ten years earlier. Chapot-Prévost published then a book, *Chirurgie des tératopages,* where he claimed that he himself could have been able to cure several of these historical conjoined twins, like Chang-Eng, Marie-Adèle or Rosa-Joséfha, if he had been asked. He went to Berlin to examine carefully a new phenomenon, the "Chinese brothers" considering himself to be the indisputable and inescapable authority on these matters: "All these cases are absolutely operable; and it is really regrettable that modern civilization cannot prevent this odious slavery imposed on these creatures who have all the

Figure 4: Radica and Doodica in 1896

rights to freedom and independent life." Back in Paris, he became interested in the case that got the most attention at that time: Radica and Doodica. Born in India in 1889, Radica and Doodica (Figure 4) were sold in 1893 to London showman Captain Colman, who exploited them commercially. In 1900, they came with the PT Barnum circus to Paris and were admired by a great number of onlookers. Chapot-Prévost tried to negotiate with the Barnum the right to operate them, but either the offer was insufficient or the health of the girls was not alarming and the project failed.

However in February 1902, it was the French surgeon Eugène-Louis Doyen who performed the separation of Radica and Doodica in his private Parisian clinic (Figure 5). A month before, one of the sisters had become sick with bronchitis which was most probably tuberculosis, and they had been hospitalized in Hôpital Trousseau. A few days later, they had been literally kidnapped, to be brought to Doyen's Clinic. The operation took place in the presence of selected personalities and filmed by a camera installed by the operator himself. A few journalists were wondering about this transportation from a public hospital and insinuated that the surgeon had paid Colman for the exclusivity

Figure 5: Dr. Doyen separating Hindoo twins (The Library of Congress)

of the operation. But Doyen justified this choice for calm and safety measures.

One day after the procedure, newspapers like *Le Figaro, Le Petit Parisien, Le Matin, L'Echo de Paris* announced on their first page, with engravings, pictures and accounts, the spectacular achievement of Doyen, who declared: "the separation of well conformed and viable monsters linked together by a large bridge of tissue at the level of the sternum, and scientifically labeled xiphopagus, was for a long time considered impracticable." Radica died one week later and Doodica, who had also contracted tuberculosis, died one year later. The film of the operation was often shown in sideshows specialized in exploitation of 'freak' films. It was last shown in the UK documentary series The Last Machine in 1995.

At 43, Eugène-Louis Doyen was reputed for his daring, difficult, spectacular and lucrative operations. In pursuit of modernity, he became interested since 1898 to the newborn cinematograph, for "teaching purposes," as he said, and started filming autopsies and operations in his private clinic. Most of his colleagues considered however that he did it to flatter his ego, for publicity or to resell the movies. He was accused, as several of his contemporary surgeons, to be mainly interested in money and to harm the idealized disinterested and philanthropic medicine. Concerning the case of Radica and Doodica, he claimed that his operation was far superior to the one performed by Chapot-Prévost in that it was quicker (20 minutes) and more difficult, and that the section of the liver could be achieved only thanks to his original method of compression of the hepatic pedicle with a special double lever instrument of his invention.

This was the start of an incredible quarrel between the two surgeons in defense of their prestige. In a number of professional journals and newspapers, they tried do discredit each other about the difficulty of the procedure, its duration and its achievement: Chapot-Prévost would have retouched the x-rays to show the heart inversion in Rosalina. . . . The death of Radica was due to poor hemostasis and so on.

COMMENTS

Although these operations performed by Chapot-Prévost and Doyen seem very benign by today's standards, this incredible struggle between two surgeons at the beginning of the last century raises several questions that are worth discussing for today's practice, the first being the ethical considerations concerning the decision whether or not to do a life-threatening operation on twins who could live up to an advanced age like Chang and Eng. In her book, *One of Us: Conjoined Twins and the Future of Normal,* bioethicist and writer Alice Dreger succeeds in questioning such an accepted concept as normal and the practices that enforce it, particularly in the presence of living conjoined twins who share an important or vital organ. A whole chapter is concerned with the "split decision" and by whom the decision to operate is made. Most often, the parents and the doctors think it should be done for a better reassignment in society, without questioning the true feeling of the children who might be perfectly happy as they are. This questioning becomes even more acute, when the only solution is to sacrifice one twin, to preserve a vital organ for the other. This type of euthanasia has been the subject of great debate in recent cases. Although Dreger's focus is on conjoined twins, she also explores intersex, and cranio-facial malformations, where the question arises: who should make the decision to operate at an early age: the doctors, the parents? Nowadays, with the security of modern anesthesiology, the separation of xiphopagus or the cure of cleft lip and palate are widely recognized procedures and encounter few opponents; but what about intersex reassignment – a subject of high controversy today – what about craniofacial operations for pure cosmetic reasons?*

The second issue raised by these conjoined twin separations is related to the concept of innovation and performance in surgery. Although we agree with Riskin et al., that it is clear that surgical innovation is fundamental to surgical progress and has significant health policy implications, we have to be very cautious about the motivations leading to innovation. For a few surgeons, innovation signifies a performance whose main purpose is to enhance its own fame and ego, and prove his superiority to colleagues and the general public.

The so-called "world premiere operations" have often led to unspeakable rivalries between self-centered surgeons, as was the case between Doyen and Chapot-Prévost, or recently concerning the first facial transplantations. These shameful and indecent disputes certainly discredit our profession.

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* As an example, a patient of mine was born with severe craniofacial malformation including cleft lip, plagiocephaly with asymmetrical height of the orbits and hypertelorism. the cleft was operated at 6 months, the plagiocephaly at age 2, with the plan to correct the hypertelorism at age 3, but the parents refused, saying that the girl would decide later for herself. she came back at age 18, asking only for a rhinoplasty, being perfectly happy with the wide distance between her (now symmetrical) orbits.