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 monsters differed from the Prodigious and the Multilated 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 cecytomy by the surgeon Treluying, 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 1892, 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 physiology 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 Traphil, 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 Traphil 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 adhesions 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 Frunay, Horta-Lister 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.

Towards 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, Rochum, on his own account performed it right after birth in his private clinic. One of the twins apparently survived. In 1874, Lardiet, a practitioner in Moselle (France), separated shortly after birth an incomplete parasitic child inserted in the epigastrum. The Medical Society of Nancy considered it as a première, but in fact, it was more like removing a tumor. In 1881, two Swiss surgeons, Baudet 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 read the opinion of a famous teratologist, Camille Dareste, who had made a classification of congenital double monsters: the ones where the organs are not inverted 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 encouraged 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, 16-year-old Eduardo Chupot-Prieto 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 system dimensions: 1224.0x792.0

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.

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.

Monsters differ from the Prodigious and the Multilated 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.”

HISTORY

HISTORY

THE UNSPEAKABLE HISTORY OF THORACOPAGUS TWINS’ SEPARATION

Denys Montandon, MD – Geneva, Switzerland

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

Found among females, with a ratio of 3:1.
All these cases are absolutely operable; and Josépha, if he had been asked. He went to Berlin to examine that he himself could have been able to cure several of these his-
in Paris, with pictures and x-rays, showing the inversion of the survive one.

Both longed to be separated, but in different ways. I longed for a shared liver. Although they could 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-Prévost became a national scientific hero and the Brazilian parliament allocated him credits to tour Europe and present this sensational “first case of living thoraco-xipho-pagus operated at the age of seven.” On the 9th of October 1901, he exhibited Rosalina at the Salpêtrière 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 trétaurapages, 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-

Joseph, 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 rights to freedom and inde-

As an example, a patient of mine was born with severe craniofacial mal-
formations, including cleft lip, plagiocephaly with asymmetrical height of the orbits and hypertelorism, the cleft lip operated at 6 months, the pl-
giocephaly at age 3, with the plans to correct the hypertelorism at age 5, but the parents refused, saying that the girl would decide later if she herself would come back at age 18, asking only for a rhinoplasty, being perfectly happy with the wide distance between her (now symmetrical) orbits.

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 con-
nerning 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 Uv 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 reassessment in society, without questioning the true feeling of the children who might be perfectly happy as they are. This questioning becomes even more acute, as 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 Doyen’s focus is on conjoined twins, she also explores inter-
s, 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 xipho-pagus or the cleft of cheek lip and palate are widely recognized procedures and encounter few opponents; but what about intersex reassignment—a sub-
ject of high controversy today—what about craniofacial opera-
tions for pure cosmetic reasons?9

The second issue raised by these conjoined twin separations is related to the concept of innovation and performance in sur-
gery. 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 cau-
tious about the motivations leading to innovation. For a few sur-
geons, 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 unseizable rivalries between self-centered surgeons, as was the case between Doyen and Chapot-Prévost, or recently con-
cerning the first facial transplantsations. These shameful and indecent disputes certainly discredit our profession.

REFERENCES


Figure 4: Radica and Doodica in 1896

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