

Two early case reports on conjoined twins

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Case reports of thoracopagus/xiphopagus conjoined twins by Godofredus Christell (1751) and Francesco Luigi Fanzago (1803) have been recovered and can be added to the history of conjoined twins.

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Introduction

Conjoined twins are believed to result from incomplete division of the inner cell mass at around two weeks after ovulation. The incidence of conjoined twins is not well established but probably lies between 1 in 60000–100000 deliveries.^{1–3} Maternal age appears to have no effect on the incidence of conjoined twinning.^{3,4} They are named according to the attached regions, for example, 'thoracopagus' indicates that there is ventral union of the thoracic regions (pégnunai = attach). Most conjoined twins are females and of the thoracopagus variety.^{1,3} The purpose of this paper is to add two previously unnoted historical case reports of thoracopagus/xiphopagus twins to the literature.

The Strasbourg conjoined twins of 1750

In 1751, Godofredus Christell presented his doctoral thesis at the University of Strasbourg.⁵ It contained, inter alia, a case report of a 29-year-old woman who, on 3 June 1750, in the ninth month of her first pregnancy, gave birth to conjoined twins (Figure 1). She was delivered by a certain Dr Iosias Weigen, who later – from 1769 until his death in 1773 – became clinical teacher in Obstetrics at Strasbourg.^{6,7} Three feet presented in the vulva, and Dr Weigen performed a breech extraction of the first twin. During this procedure, he encountered the union between the twins. After the delivery of the first child, Weigen performed a (somewhat modified) breech extraction of the second twin. The two girls were born without the use of instruments. They were ventrally connected from xiphoid to umbilicus (xiphopagus type

of conjoined twins). The communal umbilical cord contained two arteries and one vein (Figure 2). The twins died during or shortly after delivery; the mother survived, however. In paragraph XV, Christell correctly postulated that conjoined twinning was caused by an incomplete separation of the twins *in utero*. The European literature as it existed to that time was discussed, and on the basis of this literature Christell presented in paragraph XVI of his

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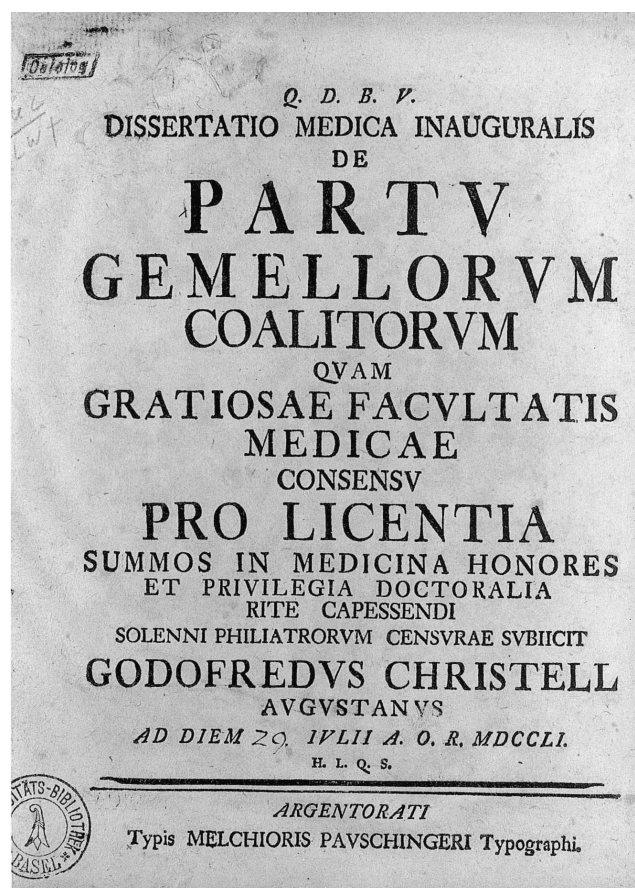


Figure 1 Title page of Christell's thesis (1751)

thesis the following classification of conjoined twins: I *Capite* (craniopagus), II *In pectore* (xiphopagus/thoracopagus), III *Lateribus*, IV *Abdomine* (xiphopagus/thoracopagus; the case by Iosias Weigen), V *Hypogastris*, VI *Inguinibus, perinaeo et natibus*, VII *Coalitis genitalibus*, VIII *Dorso* (pygopagus).

The Paduan conjoined twins of 1802

The treatise of Francesco Luigi Fanzago (1764–1836)⁸ describes two female xiphopagus twins born on 6 November 1802 in Brescia, Italy, to a woman in the seventh month of her second pregnancy. The spontaneous breech delivery was attended by a midwife and took place without the use of instruments [*sole forze della natura*]. There was one placenta, which had to be removed manually, and one umbilical cord. One of the twins was somewhat smaller than the other (Figure 3). They were displayed in several Italian cities and finally, on 24 May 1803, transported alive [*quasi setti mesi*

di vita] from Venice to Padua. The twins died shortly thereafter, and an autopsy was performed in Padua on 1 June 1803. It appeared that both livers were connected in the midline and the fused part was situated in the junction. The treatise is subsequently dated: Padova, 30 Guigno 1803 (Padua, 30 June). Fanzago's treatise predominantly consists of an anatomical description of the twins without reflections on the etiology or the pathogenesis of conjoined twinning. Except for one 'European' case report of conjoined twins by Rueff (1554), he mentioned only seven Italian case reports since 1748.

Discussion

In common parlance, conjoined twins are often called 'Siamese twins', after Chang and Eng Bunker of Siam (Thailand), born in 1811 of a Chinese father and a Siamese mother. These male twins were ventrally connected from xiphoid to umbilicus (xiphopagus). Until their 17th birthday they lived in Thailand; thereafter they were displayed in Europe and America by PT Barnum. Chang and Eng married two sisters and each of them had nine children.

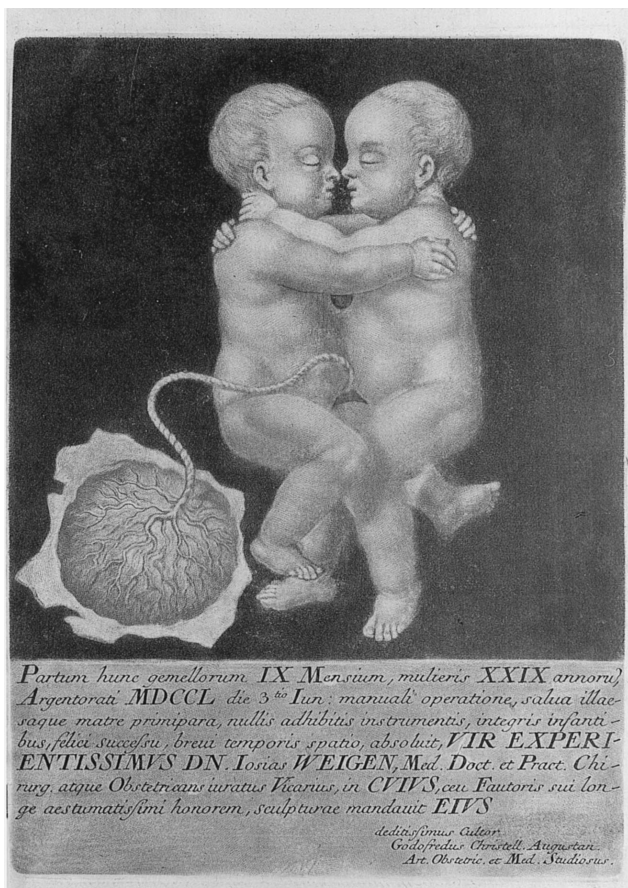


Figure 2 The Strasbourg conjoined twins of 1750 with placenta and part of the foetal membranes

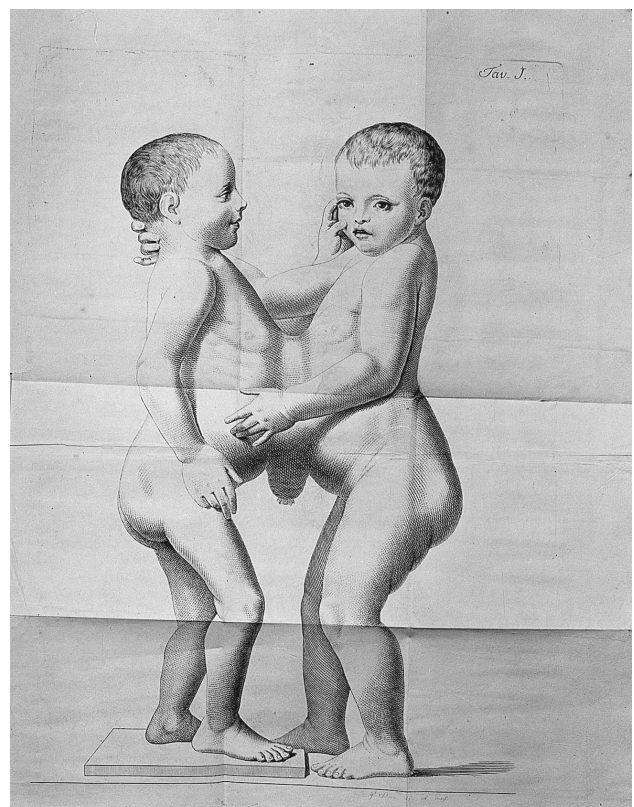


Figure 3 The Paduan conjoined twins of 1802. The left twin is somewhat smaller and positioned on a shelf

None of their children were twins and none had congenital abnormalities. Each twin built a house for his family and they alternately stayed in one house or another. After extensive investigations many eminent physicians and surgeons, among whom Sir William Ferguson and Rudolf Virchow, advised against an attempt at operative separation. In 1874, at the age of 63 years, Chang contracted pneumonia and died; Eng survived him for about 2 hours. At autopsy, it was seen that the liver and several major vessels were shared. An attempt at operation with 19th century techniques would probably have been fatal.^{9–12} As of 1971, more than 1000 descendants of Chang and Eng Bunker had been traced.¹ In 1959, four generations after the original Bunker brothers, a monozygotic twin pregnancy was reported.^{1,13} Interest in conjoined twins increased after the Bunker twins had toured Europe and demonstrated that such a malformation was certainly not incompatible with life for a considerable period of time.

In the preceding centuries, the rare births of conjoined twins were reported in popular treatises^{9,10} and as medical case reports.^{5,8} As early as 1689, the German Dr Koenig operated on a pair of xiphopagus twins by tightening ligatures around the connecting band of tissues, with complete success.¹⁰ This achievement was known to Christell⁵ who made mention of Koenig in his thesis ('Gemellas in pago Hüttingen prope Basileam A. 1689 a cartilagine ensiformi usque ad umbilicas, funiculus insuper contortis, coalitas et ligatura separatas profert KOENIG'). The spontaneous birth of the Strasbourg and Paduan xiphopagus twins demonstrated that vaginal delivery of conjoined twins may occur, although

dystocia is common. Until now, the majority of conjoined twins have been reported as case reports. In this respect but few changes can be observed during the past 250 years.

References

- 1 Tan KL, Goon SM, Salmon Y, Wee JH. Conjoined twins. *Acta Obstet Gynecol Scand* 1971; **50**: 373–380.
- 2 Filler RM. Conjoined twins and their separation. *Semin Perinatol* 1986; **10**: 82–91.
- 3 Edmonds LD, Layde PM. Conjoined twins in the United States. *Teratology* 1982; **25**: 301–308.
- 4 Barth RA, Filly RA, Goldberg JD, Moore P, Silverman NH. Conjoined twins: prenatal diagnosis and assessment of associated malformations. *Radiology* 1990; **177**: 201–207.
- 5 Christell G. De Partu Gemellorum Coalitorum. Thesis, University of Strasbourg (Argentorati), 1751, pp 34–86.
- 6 Fasbender H. *Geschichte der Geburtshilfe*. Gustav Fischer: Jena, 1906 (reprint: Georg Olms: Hildesheim, 1964), p 249.
- 7 Von Siebold ECJ. *Versuch einer Geschichte der Geburtshilfe*, vol. 2. Theod. Chr. Friedr. Enslin: Berlin, 1845, p 416.
- 8 Fanzago FL. *Storia del Mostro di due Corpi che nacque sul Bresciano in Novembre 1802*. Guiseppe e Fratelli Penada: Padua, 1803.
- 9 Bondeson J. The Biddenden Maids: a curious chapter in the history of conjoined twins. *J Roy Soc Med* 1992; **85**: 217–221.
- 10 Bondeson J. The Isle-Brewers conjoined twins of 1680. *J Roy Soc Med* 1993; **86**: 106–109.
- 11 Lotze R. *Zwillinge. Einführung in die Zwillingsforschung*. Verlag Hohenlohesche Buchhandlung Ferd. Rau: Oehringen, 1937, pp 25–36.
- 12 Pritchard JA, MacDonald PC, Grant NF. *Williams Obstetrics*, 17th edn. Appleton-Century-Crofts: Norwalk, Connecticut, 1985, p 508.
- 13 Aird I. Conjoined twins – further observations. *BMJ* 1959; **1**: 1313–1315.