

NEWS, VIEWS AND COMMENTS

Twin Classics: Research That Always Inspires/Twin Studies: Elder Twin Relationships; Superfecundated Twinning in Chimpanzees; Conjoined Twinning and Embryo Transfer; Reduced Frequency of In Vitro Multiples/Professional and Human Interest: First Identical Twin Renal Transplant; Identical Triplet Wedding; Spanakos Twins: Boxers; Twins in Space; Political Twins

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Ten classic works in twin research are described. These volumes are rich in data, interpretation, and impact, and contain life history material that adds depth and dimension to the quantitative findings. Next, research on social relationships in older twins, superfecundated twinning in chimpanzees; effects of embryo transfer on conjoined twinning and the reduced frequency of in vitro multiples is reviewed. Finally, there has been considerable public interest surrounding the first identical twin renal transplant, an identical triplet wedding; identical twin boxers, a twin living in space, and a politically active twin pair.

Twin Classics: Research That Always Inspires

There are books on twins that grow old on our shelves, but never age. We turn to them often because the material continues to excite and inspire, and because useful information in these sources is often unavailable elsewhere. And, not surprisingly, we occasionally find that some ideas and interpretations in the current twin literature have been pre-empted by the pioneers in our field.

In 2009–2011, when I was writing *Born Together — Reared Apart (BT-RA)* (Segal, 2012), Tom Bouchard and Len Heston were clearing out their libraries. Tom bequeathed to me some of his volumes from the Third International Twin Congress held in Jerusalem, Israel, June 16–20, 1980, as well as the texts *Human Genetics: Problems and Approaches*, by Vogel and Motulsky (1979) and *Human Genetics*, by Novit-

ski (1982); the latter includes the wonderfully informative photograph of MZ twin children holding their hands the same way. Len gave me his personal copies of the reared-apart twin studies by Newman, Freeman, and Holzinger (1937), Shields (1962), and Juel-Nielsen (1965/1980). Len's signature was only inscribed on the inside cover of Shields's work, but I placed a sticker inside the other two that read 'Gift from Len Heston' and added the date. Reading through these sources while writing *BT-RA* was kind

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of thrilling because of whose collections they came from. I felt part of a great legacy, especially because I was writing a book that would add to the history of reared-apart twin studies.

Several months ago, Irv Gottesman went through a similar pruning of his library, sending me a dazzling assortment of books and papers, among them copies of Burlingham (1952), Bulmer (1970), and Mittler (1971). Gottesman signed them all (I asked him to), but I was deeply touched by what he had written inside Bulmer. The book originally belonged to James Shields and Shields had apparently given

it to Gottesman. Gottesman drew a line from Shields's name to his, and from his name to mine.

These books are prominently displayed in my Twin Studies Center Library and have inspired me to think about 'twin research classics'. In an effort to formally acknowledge such works, I have narrowed the list to 10 and summarized them below in chronological order. The list is not exhaustive and other researchers may have different ideas about what could be included — or omitted. I would be happy to hear these views and will consider publishing comments in a future *TRHG* article.

Twin Research Classics: Segal's Top Ten

1. Newman et al. (1937). *Twins: A Study of Heredity and Environment*. Chicago: University of Chicago Press.

This book presents the first comprehensive psychological and physical study of 19 sets of MZA twin pairs. Some people may not realize that this study also includes comparative data on 50 MZT and 50 DZT twin pairs. The work, which took place at the University of Chicago, was a collaboration among a biologist (Newman), educational psychologist (Freeman), and statistician (Holzinger). The chapters include a literature review, critique of methods, causes of identical twin differences, and statistical analyses of twins reared apart and together. One of the best features of this book is its compendium of life history events, behavioral results, physical findings, and photographs for each of the 19 reared-apart pairs. The coupling of quantitative and qualitative data is rare today given participant confidentiality issues, thus increasing the value of this material.

A follow-up paper to the 1937 study by Gardner and Newman (1940) is well-worth reading. The investigators report findings for a newly discovered MZA female pair, while regretting that one member of a new MZA male pair refused research participation. As I noted in *BT-RA* (Segal, 2012), Newman regretted losing this pair because of their marked personality differences.

2. Burlingham (1952). *Twins: A Study of Three Pairs of Identical Twins With 30 Charts*. London: Imago Publishing.

Burlingham's *Twins* is an extraordinary record of the early behaviors of three young MZ twin pairs, Bessie and Jessie, Bert and Bill, and Mary and Madge. The children were variously observed from birth to age 5 years at the Hampstead Nurseries, a residential facility financed by the Foster Parents' Plan for War Children, Inc., in New York. The study was intended to identify the 'environmental and innate conditions which account for the differences between their development and that of ordinary children' (p. x).

Other intriguing topics include the fantasy of having a twin, twins' reactions to separation and parent-twin relations. My favorite chapter is 'The Beginning of the Twin Relationship', an informative look at co-twins' first awareness of one other — a topic rarely considered. Detailed charts tracking each child's behavioral and physical development are provided.

3. Shields (1962). *Monozygotic Twins: Brought Up Apart and Together*. London: Oxford University Press.

Shield's study presents findings from 44 MZA twin pairs and some additional data on 11 DZA twin pairs. Like the Newman et al. (1937) study, Shields provided a terrific overview of the field, statistical findings on physical variables, intelligence and personality, and appended informative case material for each twin pair with names altered to protect the twins' identity. Shields was a great thinker and a great observer of behavior. One of my favorite passages is his take on the social attraction between MZA co-twins: 'The youngest pair, S f 1, who became attracted to one another without realizing they were twins, gave rise to the reflection that there might be a biological basis for the frequently observed close attachment of monozygotic twins. Cattle twins, when reunited after segregation, can pick out one another from others in the herd ...' (p. 51). This reasoning anticipated issues currently being addressed by evolutionary-based researchers.

4. Rosenthal (1963). *The Genain Quadruplets*. New York: Basic Books.

I read this book in 1974 when I learned I would be spending the summer at NIMH in Bethesda, Maryland, coding follow-up data on the Genain Quadruplets. The four MZ sisters (whose falsified first names follow the letters of the institution where they were studied: Nora, Iris, Myra, and Hester), were concordant for schizophrenia, albeit with different symptoms that varied in severity. The book was published at a time when the roots of schizophrenia were linked

to parental upbringing and dysfunctional family patterns. However, Rosenthal and his colleagues underlined contributions from both genetic and environmental sources. They explained the disorder with reference to diathesis–stress theory, asserting that behavioral disorders are outcomes of both genetic and environmental risk factors working in concert.

5. Juel-Nielsen (1965/1980). *Individual and Environment: Monozygotic twins Reared Apart*. New York: International Universities Press.

Juel-Nielsen conducted the only reared-apart twin study that was population based. His interest in reared-apart twins began when an outpatient at the State Hospital in Risskov, Denmark mentioned that she had been separated from her twin at 3 weeks of age. Several weeks later, Juel-Nielsen was approached by a journalist researching a similar case. Given the availability of population registries in Denmark, he located 10 additional MZA twin pairs born between 1870 and 1910. This study, which compared the twins' rearing homes, general intelligence, personality traits, health-related measures, and psychiatric symptoms, was first published in 1965. A 1980 edition, with an introduction by Irving Gottesman, included 20-year follow-up data. The twins' life histories (with altered names) complement the quantitative results. A 1981 interview with Juel-Nielsen, conducted at the University of Minnesota by Tom Bouchard, provides a rare inside view of his findings, his thinking about the findings and their impact on the field; see Segal (2012).

6. Koch (1966). *Twins and Twin Relations*. Chicago: University of Chicago Press.

Koch's meticulously done study compares similarities and differences in cognitive measures, personality variables, and physical characteristics among 5- and 6- year-old twin pairs, organized by zygosity and sex. The social relationship qualities of each type of twin are also examined. In addition to its quantitative findings, the work includes qualitative accounts of many aspects of twinship. Koch's comparative descriptions of the psychological situations of the five types of twins (MZm, MZf, DZSSm, DZSSf, DZOS) remain among the best in the field.

7. Scheinfeld (1967). *Twins and Supertwins*. New York: J.B. Lippincott, Williams & Wilkins.

I believe this is the first twin research overview I read and I now own five copies of this book — the one I bought, the one I received from Gottesman, the two I received from Kay Cassill (former director of the Twin's Foundation in Providence, Rhode Island) — and the signed copy I received from Amram Scheinfeld in June 1977 when I met him and his wife Dorothy in New York City. I still love this book with its attention to psychological differences among twin types, significant twin-related issues (e.g., school separation, individuality), informative illustrations, twinning statistics, and list of 'trailblazers' in the twin research field. I

consult this wonderful volume from time to time because it contains material that is not included in other sources. For example, it was here that I learned about the first known case of switched-at-birth twins in Fribourg, Switzerland, and the names of the physicians who documented the genetic relatedness of the separated twin boys. A book written by the twins' mother is also worth reading; see below. Scheinfeld also discusses psychological and biological aspects of higher order multiples, and twins in myth, folklore, and literature.

8. Bulmer (1970). *The Biology of Twinning in Man*. London: Oxford University Press.

Bulmer's book on the biology of twinning has stood the test of time. Of course, more is known today about the genetics of twinning, biological variations among the different twin types, and changing population frequencies (e.g., Benin now delivers more twins than Nigeria; see Smits & Monden, 2011) than when the book appeared in 1970. Specifically, Benin's 2006 twinning rate was 27.9/1,000 births, with 53,595 live births in 10 years before the survey (1999–2008); Nigeria's 2008 twinning rate was 19/1,000 births with 83,003 live births (1996–2006). In addition, Bulmer's work was completed prior to the advent of assisted reproductive technologies that have dramatically altered twinning rates in Western countries. Still, Bulmer's book qualifies as a classic because of the material it presented at the time. It also presents one of the clearest and most complete discussions of polar body twinning in the literature.

9. Gottesman & Shields, (1972). *Schizophrenia and Genetics: A Twin Study Vantage Point*. New York: Academic Press.

'Gottesman & Shields' is a household term among twin researchers, the joining of two great names in a stunning collaboration that put forward a multifactorial polygenic threshold theory. This comprehensive twin study of schizophrenia, published in 1972, identified 75 twin pairs (24 MZ and 33 DZ) from the Maudsley Twin Register maintained by the Psychiatric Genetics Research Unit of the Medical Research Council, in London. The 57 pairs in the study included 62 probands who had been consecutively admitted into the hospital. The project broke new methodological ground in its quest to determine the effects of diagnosis on concordance rates. Patient summaries (that did not include zygosity or diagnosis) were distributed to six senior judges from the UK, USA, and Japan. Following categorization of the judges' classifications and elimination of two MZ twin pairs that did not include a schizophrenic proband, the proband-wise concordance rates were: 15/26 or 58% (MZ) and 4/34 or 12% (DZ), consistent with strong genetic effects. Other valuable features of this volume include overviews of genetic family studies, case studies of the participating pairs (personally interviewed) and a comparative evaluation of twin studies of schizophrenia. A foreword

by Eliot Slater and an in-depth 'critical afterword' by Paul Meehl are enlightening.

- Loehlin & Nichols, (1976). *Heredity, Environment, and Personality: A Study of 850 Sets of Twins*. Austin, TX: University of Texas Press.

Loehlin and Nichols's work is monumental, but not only because of its sample size ($N = 850$ twin pairs) and its wide range of topics (intelligence, personality, social relatedness, rearing factors). This study addressed (and I believe generally resolved) issues that are still being debated today. For example, the equal environments assumption (the concept that MZ and DZ twins are treated alike with reference to a given measure) was examined in analyses of similarity/dissimilarity of parental treatment and child behaviors. The investigators showed that whether or not parents treated MZ twins alike was unrelated to their developmental outcomes. Researchers will continue to find new bits of information and analyses upon subsequent readings. Whenever I research a new subject, I usually consult

Loehlin and Nichols's study to see if they had considered it previously. They often had.

Other works deserving of honorable mention are those by Newman (1917) on the biology of twinning, Newman (1940) on human twins, triplets and more, Lindeman (1969) on a pair of MZ co-twins who met at age 25, and Mittler (1971) on the findings and implications of twin research. The series of studies conducted on the identical Dionne quintuplets is well known (Blatz et al., 1937), but conducted under circumstances insensitive to the family's well-being. A little known book highly deserving of mention is *He Was Not My Son*, by Madeleine Joye (1954), the mother of switched-at-birth MZ male twins from Switzerland. Joye provided a compelling and heart-breaking account of her love for her 'fraternal twins', the realization that the boy she favored was not really hers, and the sad events following the return of each child to his biological family.

A paper on the top 10 classic papers in twin research is being planned. Suggestions are welcome; a copy of the manuscript should accompany all suggestions.

Twin Studies

Elder Twin Relationships

A study of 35 individual twins, ranging in age from 70 to 91 years, underlines the diversity and complexity of their social relationships with each other (Pietilä et al., 2012). The authors approached this subject from a life-history perspective, applying narrative analysis to the data they gathered during approximately one-hour-long interviews. They noted that unstructured sessions may be preferred to structured inventories and questionnaires among the elderly, given possible difficulties in form completion.

The twins were drawn from the Swedish Adoption Twin Study of Aging (SATSA) and the Gender Study. Twenty twins (from both intact and non-intact pairs) had been reared together while 15 twins had been reared apart. Among the reared-apart group, eight twins had been separated early in life and raised by unrelated families living far from one other; first contacts occurred several years later. In contrast, seven reared-apart twins had been raised by relatives and lived within close proximity to one another. Sample questions were: 'What was it like to be a twin when you were a teenager?' and 'What is it like now as you get older?'

The nature and quality of the twin relationships were organized into three categories: Nurturing (emotionally close; supportive; loyal), Draining (emotionally close, but dependent; conflicting communication; competitive), and Superficial (emotional distance; lack of familiarity; little in common). Twenty-four twins (10 MZ, 14 DZ) were in the Nurturing group, while eight twins (only MZ) were in

the Draining group and three twins were in the Superficial group (2 MZ, 1 DZ). All twins in the Draining and Superficial groups had been reared apart. The Nurturing group included both reared-apart and reared-together twins.

This paper is recommended for researchers interested in twin relationship dynamics, especially among older twins. The small sample urges caution in the interpretation, especially because 11 twins (approximately 25% of the sample) withdrew. The difficulties experienced by the mostly reared-apart twins are worth noting because they are at odds with results from the Minnesota Study of Twins Reared Apart (MISTRA). Specifically, more MZA than DZA twins felt extremely close to one another and felt that the co-twin was very familiar, both at the time of meeting and at the time of the study; only the first meeting ratings differed significantly. In addition, the reared-apart twins expressed higher levels of current closeness and familiarity toward their newly found co-twin than to the adoptive siblings with whom they had been raised. The mean age of the twins (45.28 years), sample size (MZA: 89, DZA: 65), and methodology (structured inventory) of the MISTRA may partly explain the discrepant findings (Segal et al., 2003).

Superfecundated Twinning in Chimpanzees

An article on twinning and heteropaternality in chimpanzees is a fascinating read for anyone interested in parallels

between human and non-human twinning. Ely et al. (2006), from the Alamogordo Primate Facility at the Holloman Air Force Base in New Mexico, found that DZ twin chimpanzees can have different fathers, as can humans; see Hansen and Simonsen (2008). A female chimpanzee, housed with two different males, delivered a pair of same-sex chimps (T4749 and T4750). DNA testing of 22 short tandem repeat markers indicated superfecundated twins.

The researchers also cited relevant data gathered between 1926 and 2002 across five different chimpanzee colonies. Among the 1,865 deliveries were 52 twins and two triplets. The mean MZ twinning rate was estimated to be 0.43% (usually given as 1/240), similar to the natural human MZ twinning rate of 0.48%. In contrast, the DZ twinning rate was 2.35%, considerably higher than the natural human DZ twinning rate in Caucasian populations of about 0.83% (usually given as 1/120, approximately double the MZ twinning rate). Female chimpanzees that had delivered twins were five times as likely to deliver a second set, relative to females that had delivered only singletons. It was concluded that both twinning, and possibly heteropaternality, are more significant aspects of chimpanzee reproduction than had been considered. Only two previous reports of twinning in chimpanzees had been reported at that time.

Embryo Transfer and Conjoined Twinning

Finnish investigators recently reported the first known case of conjoined twins conceived after transfer of a multinuclear embryo (Mankonen et al., 2015). The mother, 33 years of age and healthy, was referred to the clinic because of her partner's infertility. An eight-cell embryo with three multinuclear, even-size blastomeres was transferred at 2 days post

ovulation. Confirmation of pregnancy with detection of a single heartbeat was noted at 7 weeks, but thoracopagus conjoined twins (MZ twins joined from the upper chest to the lower belly) were confirmed at 12 weeks. An induced abortion took place.

Mankonen et al. noted that multinuclear embryos are observed in 15–30% of embryos in nearly 80% of IVF or ICSI cycles, so are not uncommon in pregnancies associated with assisted reproductive technologies. Such embryos are considered abnormal and are usually not transferred; however, the birth of a normal child conceived with a multinuclear embryo has been reported. The investigators also recognized the need for additional studies on embryo morphology and its relationship to twinning and health. The need to determine if there is a common origin to the presence of multiple nuclei in blastomeres and incomplete zygotic division was underlined.

Reduced Frequency of In Vitro Multiples

A recent report from the Society for Assisted Reproductive Technology found a reduction in the number of twin and triplet births conceived via assisted reproductive technologies (Kaplan, 2015). A total of 12,436 twin and 411 triplet births were reported in 2012, in contrast with a total of 12,085 twin and 376 triplet births in 2013. These figures were based on deliveries across all maternal age groups. The explanation appears to be a reduction in the number of embryos transferred. Given the infant and maternal risks of higher order multiple births following transfer of many embryos, physicians have encouraged women to transfer fewer embryos at a time. This practice has not reduced the chance of a successful pregnancy.

Professional and Human Interest

First Identical Twin Renal Transplant

The British Journal of Perioperative Nursing publishes a history of medicine series, typically the last article in each issue. The most recent such paper by Professor Harold Ellis from the Department of Anatomy at the University of London will interest readers of TR&HG (Ellis, 2015). The article documents the first identical twin renal transplant, performed in 1954, by Dr John Merrill and Dr Hartwell Harrison at the Peter Bent Brigham Hospital in Boston, Massachusetts. The twins were 24-year-old Richard (recipient) and Ronald Herrick, whose monozygosity was confirmed by concordance across the eight blood group systems known at the time. The twins had also shared a placenta, displayed the same relatively rare Darwin's tubercle of the ear (a slight projection sometimes present on the edge of the external human ear; some scientists believe it represents the pointed

part of the ear of quadrupeds; (*Encyclopedia Britannica*, 2015), had the same eye color (that differed from that of their two non-twin siblings) and had accepted a skin graft from their co-twin.

According to Ellis, the success of the renal transplant was largely the result of work by Alexis Carrel, who had developed techniques for creating anastomoses of the blood vessels. Carrel was able to transplant organs from one part of an animal to another (autoplastic transplantation), but was unable to do so between animals. This challenge occupied medical science for the next 50 years. Successful skin grafts between identical twins suggested that organ transplantation between co-twins would work — it did because the twins shared their immune system. Just as Carrel had emphasized, the key was finding ways of inducing immunosuppression in a genetically non-identical recipient.

Today, a range of drug treatments has enabled hundreds of individuals to successfully receive transplanted hearts, livers, lungs, and other organs from unrelated donors.

Identical Triplet Wedding

Twenty-nine-year-old identical Brazilian triplets, Rafaela, Rochele, and Tagiane, were married in the same ceremony at the Nossa Senhora Aparecida Cathedral, in Passo Fundo, Brazil. They have intentionally confused their fiancés on occasion.

There is also a bit of science here. The triplets wore the same dress and hairstyle at their wedding, but this was unplanned — they did not intend to match, but they liked the same gown, admitting that despite attempts to differentiate, they typically make similar choices. Genes for gowns do not exist, but body type combined with cost, feel of the fabric, personality (extraverted or withdrawn), and other relevant individual differences may be influenced by genetic factors at some level, explaining their choice of gowns.

Spanakos Twins: Boxers

The 77-year-old identical Spanakos twins, Petros and Nikos, have been described as ‘the most educated and bemedaled athletes in the history of amateur boxing’. In March 2015, I received an autobiographical statement (with that quotation) from Petros Spanakos, detailing the twins’ stunning boxing careers and the extraordinary achievements of his entire family (Spanakos, 2015, personal communication). I knew immediately who he was, having included information about these twins in *Entwined Lives* (Segal, 2000). Highlights from Spanakos’s letter are summarized below.

The twins were born at the New York Polyclinic Hospital, just across the street from Madison Square Garden. The twins eventually spent many boxing hours in that arena and others. They were inducted into six Halls of Fame, among them the New York Golden Gloves and Hall of Fame of the Greek–American International Fraternal Order. Each twin’s career included 200 boxing bouts from 1952–1964. The twins won 22 boxing titles, mostly at weights of 118- and 126-pounds. Petro won a bronze medal at the 1959 Pan-American games and Nikos was a member of the 1960 Rome Olympic Team.

The twins’ accomplishments do not stop there. Petros earned a JD degree from the New York Law School in 1965 and has since been involved with the New York City Board of Education. Nikos has a PhD in Business Administration and has taught in the State University of New York educational system. The twins’ five older brothers all combined athletics with military service. The twins’ parents, Michael and Stella Spanakos, arrived in the USA as children and neither had progressed further in school than the sixth grade. Regardless, they set high goals for themselves and for their children. In 1962, Stella was honored by New Governor Nelson Rockefeller and was runner-up for the title of Na-

tional Mother of the Year. An award-winning documentary about the Spanakos family, ‘American Story’, was produced by B’nai Brith/PBS in 1981.

Twins in Space

History is being made at this moment, both in space science and in co-twin control studies. On March 27, 2015 identical twin Scott Kelly headed for the International Space Station (ISS) where he will live for one year while his twin brother Mark will remain on earth (Hamilton, 2015). The launch took place at the Baikonur Cosmodrome in Kazakhstan. This genetically controlled experiment allows for the study of how space affects a range of psychological and medical variables, including eyesight, gut bacteria, and gene activity. Moreover, it exemplifies Einstein’s famous twins paradox, in which one identical twin takes a long space flight traveling close to the speed of light, while the co-twin remains on earth. Einstein reasoned that time would pass more slowly for the twin in space, making him or her younger since aging is a function of time (see Segal, 2000). However, Kelly will not be traveling as fast.

Another approach to the twins paradox was made by astronaut Charlie Duke in 1972 when he became part of the Apollo 16 mission to the moon. During his 11 days in space, Duke’s identical twin brother, Bill, a gastroenterologist, remained on earth. Bill’s decision to become a physician was largely a function of his own medical history, having been born with a heart defect that prevented him from choosing a physically strenuous occupation, as did his twin (Segal, 2000).

Political Twins

Andrew and Nick Diiorio are identical in many ways, but not in their politics (Schwab, 2014). The twins both began as Democrats, but Nick eventually embraced the Republican party. In November 2014, Nick ran against Democratic Congress woman Carolyn Moloney, the candidate for whom his twin brother Andrew once campaigned. Nick explained that he attended Providence College with the intention of becoming a priest, but that plan was abandoned when he decided he wanted to have a family. Nick also began questioning his allegiance to the Democratic party, thinking that Democratic plans did not always work. His twin, Andrew, completed law school and is more interested in politics as a staffer than a candidate. Nick eventually voted for George Bush while Andrew voted for John Kerry in the 2004 presidential election. However, Nick still embraces some Democratic policies, such as maintaining support for social programs.

Moloney was favored to win the congressional seat and did. She received over 80,000 votes compared with Diiorio’s 20,000. She continues to represent New York’s 12th congressional district (Wikipedia, 2014).

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