

NEWS, VIEWS AND COMMENTS

Oliver Sacks: Our Correspondence About Twins/Twin Research: Vanishing Twins Syndrome; Discordant Sex in MZ Twins; Pregnancy Outcomes in IVF and ICSI Conceived Twins/Print and Media: Superfetated Twins; Twins Discordant for Smoking; Twins in Fashion; Yale University Twin Hockey Players; Conjoined Twin–Visiting Professor

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The late neurologist and author, Oliver Sacks, published an insightful 1986 review of Marjorie Wallace's book, *The Silent Twins*, in the *New York Times*. Taking exception to his assertion about Sir Francis Galton, I wrote a letter to the *Times*' editor. The letter was unpublished, but it brought a wonderful response from Sacks himself that is reproduced and examined. Next, brief reviews of twin research concerning the vanishing twin syndrome (VTS), discordant sex in a monozygotic (MZ) twin pair, and multiple pregnancy outcomes from assisted reproductive technology (ART) are presented. This section is followed by popular coverage of superfetated twins, smoking-discordant co-twins, twins in fashion, Yale University twin hockey players, and a visiting professor who was a conjoined twin.

Oliver Sacks: Our Correspondence about Twins

The British neurologist and author, Oliver Sacks, passed away in August 2016, leaving a rich professional and personal legacy. One of my favorite essays included in his well-known collection, *The Man Who Mistook His Wife for a Hat* (Sacks, 1985), is simply titled 'The Twins'. The identical twins in question, John and Michael, were 26-year-old autistic savants. They displayed extraordinary memory and abilities to manipulate numbers, skills that brought them a great deal of joy. Sadly, their physicians separated them in 1977 to prevent what they saw as the twins' injurious communication with one another, a decision that cost them their remarkable mental skills and their life's happiness.

Sacks was interested in many forms of atypical human behavior, among them selective mutism (SM). SM is a rare

social anxiety disorder characterized by failure to speak in some situations despite normal verbal behavior in others. It is usually diagnosed when children enter a novel or unfamiliar setting and affects approximately 0.1% of school children, mostly female. Studies suggest a familial component underlying a pre-disposition to this disorder (Segal, 2003).

In 1986, Sacks published a comprehensive review of Marjorie Wallace's book, *The Silent Twins* (Wallace, 1986), for

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the *New York Times* (Sacks, 1986). The twins, June, and Jennifer Gibbons, were born in 1963 and of West Indian descent. They were raised on an army base in Haverfordwest, Wales along with elder siblings (National Public Radio, 2015). The twins were inseparable and uncommunicative at age five, and were bullied and tormented by their classmates at age eight and a half. Together, they entered a life of fantasy and crime, followed by placement in Britain's Broadmoor psychiatric facility, in Berkshire, in 1982.

Sacks's (1986) review opens with a historical summary of twin research. A sentence in his second paragraph caught my eye and I immediately realized it was incorrect: 'Through the study of twins—especially identical twins, early separated and separately reared—it would be possible, Galton wrote, to 'weigh in just scales the relative effects of Nature and Nurture, and to ascertain their relative shares in framing the disposition and intellectual ability of men' (p. 3). Galton was *not* the first to recognize the research

significance of reared-apart twins. The first investigator to apply this approach was Popenoe (1922) in his detailed case study of identical twins, Bessie and Jessie (Segal, 2000). In fact, Professor Thomas J. Bouchard, Jr. and I determined that the first mention of reared-apart twins occurred in the play *Menaechmi* (*The Twin Brothers*) by the Roman comic dramatist Plautus, who lived from 254–184 BC. I explained this error in a letter to the *New York Times*, which was forwarded to Oliver Sacks. My letter and his personal reply are reproduced in Figures 1 and 2, respectively. Note that Sacks always wrote his letters by hand, using a fountain pen (Hayes, 2017).

Given the intense interest in Oliver Sacks's work during his lifetime and in the months since his death, I believe it is appropriate to publish these letters. They will be meaningful to individuals engaged in twin research, those concerned with the history of psychology, and anyone who is fascinated by the life and work of Oliver Sacks.

Twin Research

Vanishing Twins Syndrome

The VTS was first described by Levi (1976). It involves the disappearance of one sac or embryo of a twin pair during the first trimester of pregnancy. Reasons for this loss have been variously linked to chromosomal anomalies and unfavorable implantation sites.

The outcomes of singleton twins who have lost their co-twins from VTS have been of interest. Concern has been especially high among physicians involved in providing ART to women having difficulty conceiving. ART is largely responsible for the dramatic rise in fraternal twinning observed since the 1980s (Segal, 2017). In an attempt to address this issue, Sun et al. (2017) from Guangzhou in Guangdong, China conducted a meta-analysis comparing the delivery rate and birth weight of (1) babies born to women who conceived multiples following ART (either by in vitro fertilization-embryo transfer or freezing-thawing embryo transfer), but experienced the disappearance of one sac or embryo (VTS group), and (2) women who conceived via the same assisted methods, but carried a singleton at the start of their pregnancy (comparison group).

The investigators found that gestational age and pre-term delivery rate (<37 weeks) did not differ between babies born to VTS and non-VTS mothers. However, the extremely pre-term delivery rate (<34 weeks) was higher in the VTS group. In addition, the mean birth weight of the VTS group was lower than that of the non-VTS group, but the difference was slight. Sun et al. (2017) concluded that additional research is needed to understand the health outcomes of the two infant groups. It was also suggested that

the findings should allay the anxieties of prospective parents regarding the health of a twin whose co-twin was lost due to VTS.

Monozygotic Twins Discordant for Sex

The first case report of MZ sex-discordant twins with a XXY/XX karyotype is of interest with respect to phenotypic features and twinning mechanisms (Tachon et al., 2014). The twins, a phenotypic male and phenotypic female, were delivered via cesarean section at 32 weeks. Two amnions and two chorions were detected. The pregnancy was uneventful, except for a risk of pre-mature delivery at 28 weeks. The mother and father were 28 and 33 years of age, respectively, at the time of spontaneous conception.

The twins were medically referred at five years of age due to their behavioral disorders, cognitive problems, speech disorders, and enuresis. The male twin had external hydrocephalus (congenital disorder in which there is an unusual level of fluid in the cerebral ventricles) at six months and autoimmune bullous dermatitis (various skin disorders that can be debilitating, even fatal) at four years. The female twin had a cardiac conduction disorder (slowing of impulse conduction that controls heart rate) with a normal heart at age one year. Molecular and cytogenetic studies showed mosaicism for XXY/XX in both twins, but with differences. The male twin had 78% 47,XXY in a buccal smear, 23% 47,XXY in a blood sample, and 55% 47,XXY in urinary sediment. In contrast, the female twin showed 100% 46,XX in a

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October 26, 1986

To the Editor: In his insightful review of *THE SILENT TWINS*, by Marjorie Wallace (New York Times Book Review, October 19, 1986), Oliver Sacks highlights many curious aspects of twin relationships that have fascinated writers and scientists for years. He skillfully describes the powerful bonds that can evolve between identical twins, and briefly traces the history of scientific work on twins. Sacks does, however, assign credit to Sir Francis Galton where credit is *not* due. He states, "Through the study of twins - especially identical twins, early separated and separately reared - it would be possible, wrote Galton, 'to weigh in just scales the effects of Nature and Nurture..'" It is true that Galton, in 1875, was the first to formally recognize that comparing resemblance within identical and non-identical twinships could be informative as to hereditary and environmental influences on human development (twin study method). The idea of comparing reared apart twins is not, however, mentioned in any of his writings. Recent efforts by my colleague, Dr. Thomas J. Bouchard, Jr., Director of the Minnesota Study of Twins Reared Apart, at the University of Minnesota, to identify the origin of the twins reared apart design point to a play, "Menaechmi," by Plautus, described by Elizabeth M. Bryan in a book, *THE NATURE AND NURTURE OF TWINS* (1983). In this play, a twin named Menaechmus, was lost at seven years of age. The twin brother, also named Menaechmus, launches a search for his lost brother. Instances of mistaken identity convince him that the search will prove successful. The first published case study of reared apart twins, by Paul Popenoe, in 1922, also fails to explain the earliest suggested use of this method as a systematic research design. Bouchard offers this situation as an example in which "art has preceded science." Information identifying the originator of the simple, but elegant, method of studying twins raised separately from birth would be most welcome.

Sincerely,

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FIGURE 1

Letter from Nancy Segal to the *New York Times*, commenting on Oliver Sacks's review of *The Silent Twins*. Courtesy: Nancy Segal.

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12/29/66.

Dear Dr. Segal,

You are, of course, right. My original review (of about 6000 words) got paid down — and, in particular, a long section on Galton reduced to almost nothing (and distorted in the process). I have read the major (English-language, or translated) studies of separately-reared twins published in this century — I don't know if there were any studies before 1900 [one wd. think ~~there~~ should be — or ~~can~~ ~~be~~ anecdotal?]. Your own work looks fascinating and original, and I will look forward to reading it in depth when I have time. At present, alas!, I am distracted by a dozen deadlines —
with my ^{travels} ~~travels~~,
and her ^{yard} ~~yard~~,
mine ^{is} ~~is~~.

FIGURE 2

Oliver Sacks's letter to Nancy Segal. Courtesy: Nancy Segal.

buccal smear, 75% 46,XX in a blood sample, and 95% 26,XX in urinary sediment. A schematic representation of the hypothesized mechanisms believed to give rise to these twins shows meiotic non-disjunction eventuating in a 47,XXY/46,XX zygote. Division of the fertilized egg would then have produced two zygotes that were identical with the exception of the different degrees of mosaicism for the sex chromosomal lines.

The instability of chromosomes in human cleavage-stage embryos has been recognized. The authors' note that the

type of mosaicism that can result, and its phenotypic consequences, depend upon when and where the chromosomal non-disjunctions and mosaicism occur.

Pregnancy Outcomes in IVF and ICSI Conceived Twins

ART has allowed countless couples to raise sons and daughters they might not have otherwise, due to fertility problems. Understandably, the extent to which children

conceived via ART show greater adverse outcomes, relative to children conceived naturally is of importance to physicians and families. A 2017 meta-analysis of outcomes among infants conceived by in vitro fertilization (IVF) or by intracytoplasmic sperm injection (ICSI) was conducted by Qin et al. (2017) from Hunan, China.

The data, drawn from 64 studies worldwide, included information on 60,120 multiple births from IVF/ICSI and 146,737 multiple births from spontaneous conception. Adverse pregnancy outcomes of interest included pre-term birth, very pre-term birth, low birth weight, very low birth weight, small for gestational age, perinatal mortality, and congenital malformations. In contrast with some previous studies, the two multiple birth groups did not differ in the prevalence of unfavorable outcomes. However, some significant differences were observed among twin infants from

different continents, countries, and income groups. For example, Singapore showed the highest prevalence of pre-term and very pre-term birth, followed by Italy and Australia, while Finland showed the lowest prevalence of early birth. Multiples born to high income families showed a higher prevalence of congenital malformations and a lower prevalence of perinatal mortality, relative to multiples born to upper middle income families.

The investigators noted that some differences between this study and earlier studies could be explained by factors such as variation in defining adverse outcomes, heterogeneity in the estimates across studies and/or the retrospective nature of the analysis. They called for the construction of worldwide population-based registries that would enable more precise indications of adverse perinatal outcomes among multiples.

Print and Online Media

Superfeted Twins

I received a recent Q & A entry from a *New York Times* reader, with an attached note that read ‘Are these twins?’ The article, titled ‘Pregnancy Twice Over’, attempted to answer the following question: ‘Can a woman have two fetuses in different stages of development in the uterus at the same time?’ (Ray, 2017). The answer to that question is *yes*. The process giving rise to this unusual pregnancy is called superfetation and while it happens fairly often in mammals, it is not often observed in humans. That does not necessarily mean that superfetation is infrequent in humans—it may simply mean that it is not often detected. That is because superfetation involves the release and fertilization of a second egg several weeks after the fertilization of a first egg. The two babies that result would show developmental discrepancies, since one would have been gestated for a longer period. However, it is likely that such twins would be considered ordinary fraternal twins who generally differ in appearance and other physical manifestations.

My sense that superfetation in humans may be underreported is underlined by personal communication from a superfetated twin, born in 1959 (Personal communication, February 7, 2017). This individual’s mother had carried her for one month when a male was conceived. The twins’ birth weights were 5 pounds, 10 oz. and 3 pounds, respectively, and the male twin’s lungs had not fully matured. Doctors were concerned that the male twin would not survive, but he did and remained healthy. This case has never been reported in the medical literature—in fact, the doctor had told the twins’ mother that while superfetation can occur in mammals, it had never been demonstrated in humans; that was in 1959, approximately 60 years ago. However, the

idea that superfetation can occur in humans has been in existence for some time, as I explain below.

Aristotle (384–322 BC) discussed this topic in *De Generatione Animalium* (*Generation of Animals*, translation by Arthur Platt, 1910), identifying hares and humans as examples of species in which superfetation was believed to occur (Carter, 2002). The first indication that superfetation might occur in humans came from an 1856 case in which a woman aborted a 4- to 5-month-old fetus, followed by a healthy 4-week ovum one half hour later (Studdiford, 1936). Bulmer (1970) questioned the availability of conclusive evidence of human superfetation, but more recent medical studies have yielded some consistent findings (Rhine & Nance, 1976; Roellig et al., 2011; Soudre et al., 1992).

The answer to the reader’s question of whether superfetated infants would be considered twins is also *yes*. Elsewhere, I have considered the various criteria that uniquely define twins—one criterion is simultaneous fertilization, but I noted that there are natural exceptions to this rule, superfetation being one of them (Segal, 2002).

Twins Discordant for Smoking

The detrimental physical effects linked to smoking, such as cancer and heart disease, are well known. The best, and perhaps most persuasive case along these lines can be made by comparing the characteristics of smoking-discordant identical co-twins whose matched genes control for extraneous factors affecting appearance (Froelich, 2015). The photographs of seven such pairs dramatically highlight the effects of smoking on facial features, including wrinkled skin, creases around the lips, and bags under the eyes. Readers of *Twin Research*

and *Human Genetics* are invited to examine these compelling pairs of portraits at: <http://www.trueactivist.com/this-is-what-7-smoker-vs-non-smoker-identical-twins-look-like-after-years-of-lighting-up/>.

Twins in Fashion

Twins continue to attract attention in the fashion world (*New York Times*, 2017). Dynasty and Soull Ogun from Brooklyn, New York have developed a personal line called *L'Enchanteur*. Their collection includes t-shirts and jeans with sculptural rings, as well as gold and silver jewelry. Clayton and Christopher Griggs from New York's SoHo neighborhood have created custom-made embroidered accessories that are available in the men's section of the retail space *Opening Ceremony*. Their unique pieces are described as coming from a 'sewing class at NASA'.

Both sets were described as identical, although photographs of the twins who were very differently positioned make it difficult to assess the degree to which their appearance is matched. Both twin sets also come from fashion-conscious families, suggesting that their rearing environments are largely responsible for their interests and occupations. Of course, genes and environments are confounded in intact biological families, so it is likely that a blend of both sets of factors underlie their interests and talents in fashion.

Yale University Twin Hockey Players

Evan and Mitchell Smith are one of two identical twin pairs currently playing National Collegiate Athletic Association (NCAA) hockey (Bracken & Galay, 2017). The other twin pair, Jonah and Nathan Renouf, play for the University of Alaska, Anchorage, while a third pair, Tylor and Tyson Spink, graduated last season from Colgate University in Hamilton, NY. According to one of their teammates, '... it's like having two of the same player ... typically whatever one can do, the other can do just the same ... Not to mention they're great penalty killers together'. These comments partly reflect the genetic factors shown to underlie sports abilities and interests (Segal, 2000, 2015). The twins are also extremely close to one another and it appears advantageous for them to play on the same team. Mitchell commented that, '... whenever we change teams we don't have that awkward stage of finding out who you play well with'.

Despite their similarities, differences between the Smith twins have been observed. One twin is known to be quieter, while the other has been described as better looking. It has been important for their coach and teammates to distinguish between them, no doubt explaining their detection of these minor variations.

Visiting Professor—Conjoined Twin

Thirty-six-year-old Nguyen Duc, born as a conjoined twin in Vietnam in 1981, was surgically separated from his co-

twin Nguyen Viet at age seven years, with assistance from Japan (*Japan Times*, 2007, 2017; Rafu Shimpo, 2017). The twins had been joined in the lower halves of their bodies and had shared some organs. It is believed that the conjoining was linked to the release of huge doses of Agent Orange during the Vietnam War. Nguyen Viet passed away ten years ago, at age 26, due to pneumonia and abdominal bleeding.

Nguyen Duc will serve as a visiting professor in the Faculty of Health and Welfare at Hiroshima International University, beginning in April 2017. He plans to visit Japan several times each year and deliver lectures about his experience as a conjoined twin. He is extremely saddened by the loss of his identical twin, who will be unable to join him in celebrating the 20th anniversary of their October 4th separation. Interesting and informative life histories of ten other surgically separated conjoined twin pairs have been summarized by Votteler and Lipsky (2005).

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