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News, views and comments

The 2021 Congress of the International Society for Twin Studies: Twin Research at the Cutting Edge/Twin Research: Maternal Bonding with Twins; Twins with Peters Anomaly; Selective Termination in Dichorionic Twin Pairs; Neuropsychological Function in Twins with Neurofibromatosis/News Reports: World's Most Premature Survivor — A Twin; Twins Confuse Giants' Baseball Fans; Malaysian Twins Switched at Birth; 'Biracial' Twins

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Abstract

Selected highlights from the 2021 Congress of the International Society for Twin Studies are reviewed. The perspectives of a new graduate student member to the society are also included. Timely research covering issues related to maternal bonding with twins, twins with Peters anomaly, selective termination in dichorionic twin pairs and neuropsychological functioning in twins with neurofibromatosis is reviewed. The final part of this article includes interesting and informative media reports related to the world's most premature survivor who is a twin, identical male baseball players, Malaysian twins switched at birth and a pair of the so-called 'biracial' twins.

The 19th Congress of the International Society for Twin Studies (2021): Twin Research at the Cutting Edge

Professional congresses are remembered for the research that is presented and for the people behind the reports. The 19th Congress of the International Society for Twin Studies (ISTS), held between November 11–14, 2021, was extraordinary in these respects — attendees were treated to three and a half days of twin research that was truly at the cutting edge. The majority of presenters and listeners attended the event online, but several were on site in Budapest where monozygotic (MZ) twins, Ádám and Dávid Tárnoki, served as hosts for the second time. As a board member, I witnessed the planning and preparation for this meeting over the course of many months. Despite the challenges posed by COVID-19 restrictions, ISTS President Jeffrey Craig of Deakin University in Geelong, Australia put together a memorable event, assisted by dedicated board members and other conference personnel.

One of the society's future goals is promoting student membership. Therefore, I encouraged my graduate student, Elizabeth Pratt-Thompson, to attend the meeting and to share the perspectives of a young investigator interested in twin research; her comments about the meeting follow mine. We made it a point to attend different sessions to cover as many topics as possible, although we could not cover them all. Fortunately, the presentations are taped and available for viewing.

Opening Sessions

The conference hosts, identical twins Drs Ádám and Dávid Tárnoki, of Budapest, are shown in Figure 1. Professor Miklós Kellermayer DSc, Dr Med. Habil, Dean of the Faculty of Medicine at Semmelweis University, began by welcoming attendees on November 12. However, the meeting actually began the previous day with a series of specialized workshops. Workshop topics covered International Council of Multiple Birth Organisations (ICOMBO) activities, statistical analyses, technologies, twin registries, the microbiome, conducting systematic reviews and twin birth cohorts. These sessions continued on November 12, with some topics repeating, but with the new subjects included; for example, the future of ICOMBO, imaging and epigenetics.

ISTS President Jeffrey Craig formally opened the 19th Congress of the ISTS following the remarks made by Dr Kellermeyer. Contributions from the board members, local hosts and staff members were acknowledged. The over 200 attendees were encouraged

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Figure 1. International Society for Twin Studies Congress hosts, monozygotic twins Ádám (left) and Dávid Tárnoki. Courtesy of the twins.

to attend the business meeting on the final day to learn the names of the prize winners and the location of the 2023 congress. More specific statistics about attendees and society membership are summarized at the end of this article.

A special treat was a piano performance by Hungarian identical twins, Drs Zsombor and Gergely Toth-Vajna. Both twins are musicians and physicians.

In Memoriam

A regretful, but important responsibility on the president's part is recognizing late colleagues who left us a rich legacy of twin-related research. This year there were four such eminent individuals:

- Dr Isaac Blickstein (1953–2020). Dr Blickstein was a faculty member at the Sackler School of Medicine in Tel Aviv, Israel.

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He served as Chairman of ISTS's Working Group in Multiple Pregnancy, 1999–2010. He is also well known for his five books — I especially like his volume *Multiple Pregnancy*, co-edited with the late Dr Louis G. Keith, who was a lifelong ISTS member (Blickstein & Keith, 2005). A longer tribute to Isaac Blickstein appeared in an earlier issue of *Twin Research and Human Genetics* (Segal, 2021a).

- Dr Roger V. Short (1930–2021). Dr Short was Professor of Reproductive Biology in the Department of Physiology at Monash University in Melbourne, Australia. Prior to that, he was Director of the Medical Research Council Unit of Reproductive Biology, Edinburgh 1972–1982. Dr Short studied multiple birth in many mammalian species. A longer tribute to his life and work appears in *Twin Research and Human Genetics* (Craig et al., 2021).
- Dr Sandra Scarr (1926–2021). Dr Scarr was a leader in behavioral genetics, making lasting contributions to both twin and adoption research. She authored cutting-edge papers on twin methodology and the equal environments assumption, defending twin studies against alleged bias (Scarr, 1982; Scarr & Carter-Saltzman, 1979). She served on the National Research Council Committee on Epidemiology's subcomittee on twins (1976–1981). A subsequent issue of *Twin Research and Human Genetics* will feature a longer tribute to Dr Scarr's illustrious career.
- Dr Walter E. Nance (1933–2021). Dr Nance was a Professor of Human Genetics, Pediatrics and Medicine, and Chair of the Department of Human Genetics at the Medical College of Virginia/Virginia Commonwealth University (1975–2002). Prior to that he was a faculty member at Indiana University, Indianapolis. He edited the 1978 and 1981 *Twin Research* series that followed the ISTS Congresses, in Washington, D.C. and Jerusalem, Israel. A subsequent issue of *Twin Research and Human Genetics* will also feature a longer acknowledgement of Dr Nance and his career.

Keynote Address

I was privileged to introduce the keynote speaker, Dr L. Scott Forbes. Dr Forbes is a Professor of Biology at the University of Winnipeg, Canada. He is a biologist interested in the behavioral ecology of the family; for example, sibling rivalry, and conflicts and cooperation. I shared a personal anecdote in my congress introduction: In a 1990s publication, Dr Forbes proposed that dizygotic (DZ) twinning, which is more frequent among older women, could be conceptualized as an adaptive trade-off between the benefits of multiple reproduction and the costs of producing defective children — thus, he challenged the conventional wisdom that DZ twinning simply reflects an age-related increase in abnormal egg production (Forbes, 1997). Several years later, I conceived a similar idea and shared it excitedly with a colleague who informed me that the idea had already been proposed. I was disappointed, of course.

Several points from Dr Forbes' address, 'Evolutionary Biology of DZ Twinning in Humans', are outstanding: (1) Mothers of twins appear to be selected for, given their favorable phenotypes and favorable environments. (2) Twins have lower birth weights than nontwins, but twins are not necessarily disadvantaged as are nontwins, due to macronutrients anticipated by the mother. (3) Dr David Anderson suggested the 'insurance ova hypothesis', namely that DZ twinning may represent an insurance mechanism compensating for chromosomal defects (Anderson, 1990). (4) Recent empirical work has supported Anderson's insurance hypothesis in humans (Hazel et al., 2020). (5) MZ twinning is a puzzling phenomenon — but it is not just a developmental accident.

I would encourage everyone to listen to Dr Forbes's address, which is available among recorded conference talks. Space limitations of this journal article prevent complete coverage of his important remarks. In addition, given time constraints and my location, I can describe several sessions I attended, but I could not attend them all.

New Twins Book Discussion Panel

Five books were discussed at the congress's first book panel. Authors and editors and their books included Tania Kiehl Lucci, PhD: E. Otta and T. K. Lucci (Eds., 2021): *Twin Studies in Behavioral and Health Research: Current Status, Prospects and Applications*; William Viney, PhD (2021): *Twins — Superstitions and Marvels, Fantasies and Experiments*; Alexandra Matias, MD: A. Matias and I. Blickstein (Eds., 2020): *Developmental and Fetal Origins of Differences in Monozygotic Twins: From Genetics to Environmental Factors*; Dávid László Tárnoki, MD, PhD: A. D. Tárnoki, D.L. Tárnoki, J. R. Harris, & N. L. Segal, (Eds., in press): *Twin Research for Everyone: From Biology to Health, Epigenetics, and Psychology;* and Nancy L. Segal, PhD (2021b).: *Deliberately Divided: Inside the Controversial Study of Twins and Triplets Adopted Apart.* Participants described their books and responded to questions from one of the two co-chairs. Publication information for these books is provided in a separate reference list at the end of this article.

A new books session is planned for the 2023 congress. I would also like to see film showings presented in the future because they provide another informative way to appreciate the many fascinating features that uniquely define twins and twinship. In addition, the new ISTS website will post titles of recent, current and forthcoming books on twins. Please send suggestions to Dr. Nancy L. Segal, nsegal@fullerton.edu.

Psychological, Biological and Social Perspectives

I was delighted to co-chair a session on psychological, biological and social perspectives in twin research with Dr Maria Antoinetta Stazi of the Italian Twin Registry in Rome, Italy. Topics included what research reveals about young twins' social and emotional skills, how cognitive and noncognitive skills affect academic achievement, three different twin myths, testing criminological theory and factors affecting general intelligence. Interestingly, one of the participants, Margherita Malanchini, and I were both interviewed for an article on twins and motivation that was released during the conference (Rabesandratana, 2021).

These talks were followed by short presentations from new investigators. Topics included twin studies of savings and finance (Alesnadros Giannelis); memory (Zhizin Zhu); communication (Rita Hegedüs); sleep (Juan J. Madrid-Valero) and family dysfunction (Olakunle Oginni). It is important to engage the contributions of junior researchers who are positioned to shape the future goals and directions of ISTS. It is also vital to include a wide array of topics in a session on psychological, biological and social issues; suggestions for future congresses include attachment, bereavement, language abilities, behavioral problems and decision-making to name a few.

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Participant Engagement and User Involvement

Fostering investigator and twin–parent relationships was the focus of an informative session on participant engagement and user involvement. I encourage additional creative efforts along these lines. Co-chaired by ISTS board members Drs Carolyn Lister and Petra Zwijnenburg, this session stressed the importance of engaging twins and their families in the research process — not just as study participants, but as active members of a collaborative venture. Ways of providing feedback, support and other forms of assistance were discussed.

Disseminating findings in clear and useful ways is paramount. Beyond parents, twin research findings must be made available to educators, camp counselors and others who work directly with twins and their families. As an example, I have provided legal counsel to attorneys representing MZ twins who have been wrongly accused of cheating because the twins' answers and test scores were so similar. Of course, MZ co-twins *should* perform similarly given their common genetic background and demonstrated genetic influences on abilities and performance (Segal, 2017). The fact that teachers, college professors and school administrators have raised charges of cheating with minimal evidence demonstrates that they are unaware of MZ twins' matched skills and talents, hurting twins and their families in the process.

Controversial Topics

The debate style presentation of individuals representing opposing sides of selected twin-related issues was very effective. I heard the pros and cons of encouraging zygosity testing for all same-sex twins, and the benefits and drawbacks of different procedures for managing twin anemia polycythemia syndrome (TAPS). Polls taken before and after the debates showed that most attendees favored zygosity testing for same-sex twins, although the percentage dropped at the conclusion of the arguments (89% vs. 56%). The same pattern was observed for the debate on TAPS procedures (89% vs. 76%). Clearly these are topics that warrant further discussion.

Many controversial issues in twin research deserve serious dialogue and consideration. Examples include the classroom placement of twins at school, the surgical separation of conjoined twins, the selective termination of one or more embryos in multiple pregnancies, associations between zygotic division and placental arrangements and who takes credit for developing the classic twin method. Many of these topics can be informed by experts outside *ISTS* who should be encouraged to attend future meetings.

A Graduate Student's Perspective — Elizabeth Pratt-Thompson

As a current graduate student at California State University, Fullerton (CSUF) I was looking forward to the 2021 Twin Congress, especially because it was my first experience attending a twin conference. I am currently studying behavioral problems in twins for my master's thesis, under the direction of Dr Nancy L. Segal at the Twin Studies Center, CSUF.

It was intriguing to attend a virtual conference based in Budapest, somewhere so far from Southern California. It was inspiring to see so many researchers and parents interested in twins come together from different areas of the world to share ideas, present studies and interact with one other. Being able to attend the same session or workshop as other twin enthusiasts from across globe is also testament to the wonders of technology. I was particularly interested in Session 6: New Twin Registries and Studies. As a student who is new to this field, I was eager to learn more about the current work being done within this area of twin research.

Marton Piroska, MD delivered an update on the development and changes of the Hungarian Twin Registry website. I do not participate in large-scale twin recruitment; therefore, understanding the process of finding and registering twins from across Hungary and compiling them into such a vast registry was fascinating. Looking to the future, it is beneficial to know that registries are being compiled here and in other locations, furthering twin research.

Another interesting registry update came from Nizal Sarrafzadegan, MD. She explained the recruiting process being done in Isfahan, the second largest territory in Iran. She also explained the many different measures of data being collected and the goal to continue a longitudinal study based on their findings.

Both Professor Bas Heijmans and student Sophie Groene spoke about the work being done within the Twin Life Study located in the Netherlands. It was interesting to hear their different perspectives. Prof. Heijmans offered an overview of the study and gave the history of how it came to be, while doctoral student Grene presented research she is currently conducting while working on her PhD degree. Both individuals emphasized the importance of examining fetal discordance and the long-term effects of growth restrictions, although Groene additionally explained specific methods being used and was explicit about what her current research will entail. As someone who is currently working on a master's thesis and aspires to complete a doctoral program, this student gave great insight into what I should expect from such a program. I admired her observable passion, knowledge and commitment to research.

Several other speakers gave captivating presentations that I enjoyed during this session. It was also informative to see participants' and presenters' communications in the comments section of the chat. The chat brought forth ideas and conversations that I might not have been able to access, had this session been conducted in person. I look forward to the 2023 Twin Congress to learn how the research and registries described in this session have progressed.

Closing Session

The closing session of the ISTS congress took place on Sunday, November 14. President Jeffrey Craig introduced the event, followed by Dr Asma Khalil, who supervised dissemination of the awards. It was my pleasure to announce that the winner of the James Shields Award for Lifetime Contributions to Twin Research was Dr Jeffrey Craig. Dr Dávid Tárnoki recognized Sophie Groene and Christopher J. Yoon as winners of the Galton Prize for Best Student Presentation. Dr Tárnoki also acknowledged Nikki Hubers and Rebekka Koeck for winning the Gedda Prize for Best Student Poster. The ICOMBO prize was announced by Monica Rankin and given to Sophie Groene.

The next item on the agenda involved attendee participation in a game of Kahoot, an interactive quiz hosted by Ádám and Dávid Tárnoki. Attendees were presented with 20 twin-related questions that everyone in the audience should have been able to answer. Unfortunately, the technology during this game was poor on my end, causing me to choose the wrong answer to the question about the title of my new book! The winner of this game was medical student Peter Vincze of Hungary.

Following the game, President Craig offered final closing remarks. The 2021 ISTS Congress was one of the best attended with representatives from many countries around the world; see the

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summary statistics below. He also announced that the 2023 ISTS congress will be held in St Petersburg, Russia, an event to be anticipated with great excitement.

As I stated at the start of this article, congresses are remembered for the research that is presented and for the people who report it. The 2021 ISTS congress was exceptional in this respect.

Summary Statistics

Registrations: 235 (25 on site in Budapest)

Participating countries: 52
New membership: 53
Student representation: 35 undergraduate, 64 early career researcher, 39 post-graduate

Twin Research

Maternal Bonding with Twins

Maternal bonding with twins differs from bonding with singletons, but little is known about the specific aspects of mothers' experiences (Gowling et al., 2020). Dr Gowling, the senior investigator, is located at Coventry University in the UK. A qualitative methodological procedure, known as Interpretive Phenomenological Analysis, was applied to the responses of six mothers of twins. These participants were recruited from notices placed on a multiple birth social media platform. The two major themes that emerged from the analysis were (1) twin guilt and shame (e.g. concern by mothers over giving each twin individual time), and (2) mothers and twins missing out (e.g. worry that both mothers and twins did not gain as much from the developing relationship as would mothers and nontwin infants).

The researchers recognized that the small sample size of first-time mothers posed limitations as to the generalizability of the findings. I would also note that no mention was made of the method for zygosity determination — two pairs were fraternal due to their being opposite-sex, but three pairs were listed as nonidentical (one female set and two male sets) and one pair was listed as identical female. It is possible that twin type and/or sex composition affected maternal bonding; in some cases, favoring one twin was mentioned. I would refer readers to a chapter by Mann (1992) on maternal preference for one twin in premature pairs. Zygosity was not specified in Mann's study either, but the evolutionary framing of issues is illuminating.

Twins with Peters Anomaly

Peters anomaly is a rare condition involving the anterior part of the eye, leading to amblyopia (reduced vision in one eye) or congenital blindness. The condition was first described in 1906 by Dr Albert Peters. Its rarity and later organization into three classification types has made the search for its etiological factors challenging; however, evidence suggests a role for genetic factors (Almarzouki et al., 2016). Currently, Peters anomaly is considered part of the group of congenital corneal opacities, affecting 3–6 individuals per 100,000 (Medline Plus, 2020). Given the foregoing, a case of Saudi Arabian MZ male twins born at 36 weeks to consanguineous parents and showing symptoms of Peters anomaly at age 6 months is of interest. It was noted that the twins shared a mutation of the PROC gene (a mutation associated with protein C deficiency), evidence that the condition has a genetic basis. Prior to this case, protein C deficiency had not been linked to Peters anomaly.

Selective Termination in Dichorionic Twin Pairs

Outcomes after selective termination of an abnormal fetus in dichorionic twin pairs were evaluated by researchers in Barcelona, Spain (Bennasar et al., 2021). The 147 cases were first divided into 3 categories with reference to the timing of termination: early (before 18 weeks), intermediate (between 18 and 23 weeks) and late (after 23 weeks). The cases were then retrospectively reviewed for overall survival at 28 days post-delivery, pregnancy loss and preterm delivery before 32 weeks of gestation; percentages were 93.4%, 6.9% and 15.5%, respectively. Intermediate timing of termination yielded lower survival rates (86%) than early (96.9%) and late termination (100%). It was concluded that selective termination should be performed early, prior to 18 weeks, but can be performed late if diagnosis of the fetal anomaly is delayed.

Neuropsychological Function in Twins with Neurofibromatosis

The first case of MZ twins concordant for neurofibromatosis, type 1 was reported by Pendergrass and Peraza (2020) who conducted a comprehensive neuropsychological assessment of the pair. Neurofibromatosis, type 1 is also known as Recklinghausen's disease and affects approximately 1/3000–4000 individuals. Its inheritance pattern is autosomal dominant, but half of the affected cases lack a family history of the disorder, suggesting that it often arises through new mutations. Physical traits include short stature and scoliosis, but cognitive complications are more salient.

The 19-year-old twins in question were African American females, born at full term via Cesarean section. Both twins weighed 5 pounds, 5 ounces at birth. Both twins were also right-handed. They were referred for evaluation following academic difficulties during their first college year, although they had experienced previous academic challenges. The twins underwent extensive testing across multiple cognitive domains, displaying both similarities and differences. Both twins performed in the low average range, with cognitive problems noted in attention, working memory and processing speed. Co-twin differences mostly involved verbal skills in which 'Twin A' excelled, scoring at least one standard deviation above her twin sister. Many other interesting findings are reported. A difficulty with this report is failure to document the methods by which the monozygosity of the twins was established. It is also unclear whether the twins were evaluated independently by separate investigators.

News Reports

World's Most Premature Survivor — A Twin

A male twin born at 21 weeks gestation has been recognized by the Guinness World Records as the most prematurely born infant survivor (Cramer, 2021). The child, Curtis Means, is now 16 months old and healthy. Unfortunately, his twin sister C'Asya did not survive. The twins were born to 35-year-old Michelle Butler at the University of Alabama, Birmingham. Butler is also the mother of a 7-year-old daughter and 14-year-old son. The twins were conceived as their mother was entering the lower end of the age range in which DZ twinning rates rise (Segal, 2021b).

Twins Confuse Giants' Baseball Fans

Tyler and Taylor Rogers are identical twins who play for different baseball teams (Pavlovic, 2021). Tyler (a right-hander) pitches for the San Francisco Giants and Taylor (a left-hander) pitches for the Minnesota Twins. Fans unaware of the twinship were confused after Tyler delivered two pitches, while his identical twin was in

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the stands seated next to them. It turned out that Taylor had come to watch the game with the rest of his family.

Malaysian Twins Switched at Birth

I have documented the switching at birth of 10 pairs of twins from 6 countries and the USA territory of Puerto Rico (Segal, 2019). An 11th pair was identified in June 2021, involving identical twin sisters and an unrelated 'twin' sister from Malaysia (Zulkifli, 2021). The truth came to light when the three young women were 19 years of age. The story began when one of the twins, Adryani, attended a motivational camp and was told that she looked like someone at another school. She attributed the resemblance to chance until she personally met her twin sister, Noratirah, at a carnival in March 2019. Their birthdays matched, as did many of their other features, prompting them to undergo DNA testing. Blood samples were obtained from the twins, the unrelated sister Adryana, the twins' biological parents and Adryana's biological father. The results showed that Adryani and Noratirah are twin sisters with 99.9% certainty, and that Adryana is the biological child of the couple who had raised Noratirah.

The truth was kept hidden at first because the twins' mother did not wish to inflict psychological harm on Adryana. Adryana has since moved in with her biological family, but the family still regards Noratirah as part of their family. According to Islamic law, only biological children have rights to inheritance, so the switched women have been officially approved to take the name of their biological family. Hospital negligence was held responsible for the error.

Biracial Twins

So-called 'biracial twins' are becoming increasingly more common, most likely because the frequency of mixed couples has risen. A recent case occurred in Manchester, England with twins Cole and Klay, born to a Caucasian mother and half-Jamaican father (Abrahamson, 2021; Muzaffar, 2021). The twins were 15 months old as of July 2021. One twin has blonde hair and blue eyes, while his co-twin has dark hair and dark eyes. I find the term 'biracial twins' to be problematic because the twins, conceived by the same mother and father, are both of mixed race; they only appear to represent different population groups, due to the particular genes each inherited from their parents. I hope to study the life experiences of these unique twins at some time in the future.

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