The New Face of Research
Noémi Dahan-Oliel, PhD OT

12th Annual AMCSI Conference
Las Vegas, Nevada
June 29th, 2017
Disclaimer

Prior to 2014, I knew very little, actually almost nothing about arthrogryposis
SPARC Research Program

**Stakeholders Partnering for Arthrogryposis Research: An Initiative for Client-Centered Care**

- SPARC-Network
- SPARC-Collaboration
- SPARC-Rehab
- AMC Registry
SPARC Research Program

Value, Engagement, Group Work, Across Disciplines, Support Change, Share Knowledge
Summary of The 2nd International Symposium on Arthrogryposis, St. Petersburg, Russia, September 17-19, 2014

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2Arthrogryposis Clinic, Turner Scientific and Research Institute for Children’s Orthopedics, Saint-Petersburg, Russia
3Department of Pediatric Orthopedic Surgery, Karolinska University Hospital, Stockholm, Sweden
4Department of Orthopedic Surgery, Shriner’s Hospital for Children, Philadelphia, Pennsylvania

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Enormous progress has been made in understanding the etiology and therapies for arthrogryposis (multiple congenital contractures). A 2nd International Symposium on Arthrogryposis was sponsored by the Turner Institute in St. Petersburg, Russia. Olga Agranovich, Head of the Arthrogryposis Department of the Turner Institute, organized this special meeting. Care providers from multiple disciplines from all over the world representing 18 nations attended. Participants included: Pediatric orthopedic specialists, rehabilitation physicians, occupational therapists, physical therapists, medical geneticists, neurologists, craniofacial physicians, psychologists, developmental biologists, as well as representatives from parent support groups. The first symposium established the need for a collaborative and interdisciplinary approach to the treatment of arthrogryposis, engagement of parent support organizations, and the aim for more research. The second symposium highlighted the continuing need for more research on various therapies, identification of different types of arthrogryposis, standardized descriptions of severity, development of new orthotics, improved prenatal diagnosis, and studying adult outcome. Major progress has been made on both upper and lower limb treatments. © 2015 Wiley Periodicals, Inc.

How to Cite this Article:

Olga Agranovich, Head of the Arthrogryposis Department of the Turner Institute, organized this very special meeting.

Care providers from multiple disciplines from all over the world attended. Eighteen nations were involved. Attendees included: pediatric orthopedic specialists, rehabilitation physicians, occupational therapists, physical therapists, medical geneticists, neurologists, craniofacial physicians, psychologists, developmental biologists, as well as representatives from parent support groups.

The first International Symposium established the need for a collaborative and interdisciplinary approach to the treatment of arthrogryposis, engagement of parent support organizations, and the need for more research. Those issues were highlighted again, as well as an emphasis on uniqueness of each affected individual’s situation, the need for knowledge about outcomes in adults with arthrogryposis, and a re-emphasis on a definition of arthrogryposis as including multiple congenital contractures in two or more body areas (even though familial types of arthrogryposis may have great

Key words: arthrogryposis; multiple congenital contractures; international symposium; treatment; therapy; classification; support group; adults; children; newborns; Ponseti method; prenatal diagnosis; genetic basis; natural history; epidemiology

INTRODUCTION

...
SPARC-Network

Harold van Bosse

Judith Hall

Tanya Bédard

Bonita Sawatzky

Betty Darsaklis

Noémi Dahan-Oliel

Reggie Hamdy
SPARC-Network

Objectives:

• Create core group of experts
• Align research priorities with experts and funding opportunities
• Plan multi-site studies to promote evidence-based practice across the lifespan
SPARC-Network
Activities:

• Monthly conference calls
• Knowledge Exchange day SHC-Canada May 22\textsuperscript{nd}
  ➢ Research priority setting activity
  ➢ Focus Groups

Experts
Research idea
Funding opportunity
SPARC-Network

Research themes:

– Genetics: early diagnosis & screening
– Identification of outcome measures to evaluate interventions
– Longitudinal studies to document adult outcomes
– Development of an AMC registry

Patient engagement framework
Patient engagement in research is the «interactive, bi-directional exchange between stakeholders and researchers» in order to develop research that is most important to patients and to improve the quality and relevancy of research.

(Curtis et al., 2012)
The more engaged patients are in managing their own care, the better their health outcomes and more positive their experience.
SPARC-Collaboration

Core values for engagement:

- Support
- Mutual Respect
- Co-Build
- Inclusiveness

# SPARC-Collaboration

<table>
<thead>
<tr>
<th>Your contribution</th>
<th>Research Translation</th>
</tr>
</thead>
<tbody>
<tr>
<td>Determining what do you want to know, what is important to you</td>
<td>• Background</td>
</tr>
<tr>
<td></td>
<td>• Research idea</td>
</tr>
<tr>
<td>Generating ideas for research</td>
<td>• Research question</td>
</tr>
<tr>
<td></td>
<td>• Research design</td>
</tr>
<tr>
<td>Verifying the research process is realistic and do-able</td>
<td>• Validation of methodology</td>
</tr>
<tr>
<td></td>
<td>• Procedures</td>
</tr>
<tr>
<td></td>
<td>• Recruitment</td>
</tr>
<tr>
<td></td>
<td>• Requirements from patients and families</td>
</tr>
<tr>
<td>Verifying that research requirements are respectful</td>
<td>• Ethics (autonomy, informed consent, confidentiality, respect)</td>
</tr>
<tr>
<td>Identifying meaningful anticipated outcomes</td>
<td>• Expected contributions</td>
</tr>
<tr>
<td>How do you want to know the results</td>
<td>• Knowledge translation</td>
</tr>
<tr>
<td></td>
<td>• Knowledge dissemination</td>
</tr>
</tbody>
</table>
Engagement is successful when…

• Involvement at all levels
• Capacity building
• Communication and collaboration
• Your experience is valued
• Recognition of diversity
• Shared sense of purpose
SPARC-Collaboration

Focus on questions and outcomes important to you
Objectives:

• Collaboratively establish research priorities with youth and family members

• Form working groups in order to address the three most pressing concerns
Webinars to plan:

• Content
• Format
• Activities

Face-to-face meeting:

• Logistics
# An Arthrogryposis Exchange Day

## Meeting Agenda

<table>
<thead>
<tr>
<th>Time</th>
<th>Activity</th>
<th>Speaker/Panel Member</th>
</tr>
</thead>
<tbody>
<tr>
<td>8:00-9:00</td>
<td>Breakfast</td>
<td>Dr. R. Hamdy &amp; N. Dahan-Oliel, PhD</td>
</tr>
</tbody>
</table>
| 9:00-9:30  | Word of welcome                               | Dr. J. Hall, University of British Columbia  
Dr. H. van Rosse, SHC-Philadelphia  
B. Sawatzky, PhD, University of British Columbia  
M. Donohoe, PT, Nemours/Alfred I. duPont Hospital for Children |
| 9:30-10:30 | Clinician and researcher panel featuring      |                                                                                      |
| 10:30-11:00| Health break and networking                   |                                                                                      |
| 11:00-12:00| Health priority activity                      | Moderator: N. Dahan-Oliel, PhD                                                      |
| 12:00-1:00 | Lunch and networking                          |                                                                                      |
| 1:00-1:45  | Exchange panel                                | J. Carrier, parent  
T. Dick, young adult  
AM. Davison, PhD, adult  
A. Samargian, AMC Support Inc. |
| 1:45-2:30  | Plenary sessions                              | M.-H. Bolduc, Muscular Dystrophy Canada  
R. M. Wakil, Independent Living  
Dr. Moreau & Dr. Marleau, MCH, Dentistry  
V. Handfield Bigué, CIVA / Centre d'intégration à la vie active |
| 2:30-3:00  | Health break and networking                   |                                                                                      |
| 3:00-4:00  | Parallel discussion groups                    | Parent groups  
Young female group  
Young male group  
Clinician group |
| 4:00-5:00  | Closing remarks                               | Dance and music featuring L. "LaBylez" Patuelli & Rodolphe Gagnon                   |
| 5:00       | Networking Cocktail                           |                                                                                      |

**Sunday**  
December 4th 2016  
8:00 am-5:00 pm
SPARC-Collaboration
Cue card sorting activity
SPARC-Collaboration

How do we educate a wide range of doctors (those in the front line when babies are born/during ultrasounds) to understand AMC and the long term outcomes to avoid worrying new families? (28)
Given pain is such a long term and significant issue affecting AMCers, are there ways to complete surgeries and interventions that can reduce pain (as a result of the surgery or intervention) 27
SPARC-Collaboration

• Physiotherapy for older adults exercise programs
• What are the different difficulties as adults age?
• If my child who has inherited AMC from their (distal) and have a bit worse case, risk passing the gene, and will It be worse in their children’s case.
SPARC-Rehab

To develop practice recommendations for rehabilitation

Session by Caroline Elfassy, MSc OT.
Friday 2:00-3:00.
Grand Ballroom B
SPARC-Rehab

- Expert rehabilitation practitioners
- Experiential knowledge
- More research in AMC is being funded
- No rehabilitation guidelines
- No AMC-specific assessment tools
- Local clinicians may feel unprepared
Objectives:
To generate multiple future research avenues to enhance current care and establish new therapies

(Dahan-Olien, Hamdy, vanBosse, Bédard, Hall)
AMC Pediatric Registry

How:

• Create the datasets
• Evaluate the validity of the datasets
• Pilot the registry in Montreal and Philadelphia
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• Evaluate the validity of the datasets
• Pilot the registry in Montreal and Philadelphia
AMC Pediatric Registry

Utility:

- Estimate the # of children with AMC & distribution across North America
- Identify risk factors
- Platform for further studies
AMC Pediatric Registry

Procedures:
- Recruitment by health care team
- Consent to access medical chart and interview
AMC Adult Registry

Adult Arthrogryposis Registry: For patients by patients

• PI: Bonita Sawatzky, PhD
• Research team composed of investigators with and without AMC (6:6)

Session by Ann-Marie Davison, PhD.
Friday 3:15-3:45
Grand Ballroom B
SPARC Research Program

- Video created by Sabrina Shemie
- Content validated with youth, parent, adults with AMC, clinicians, researchers
- Voice-over
• video
AMC Interdisciplinary Clinic

- Team
- Format
- Research
## Mes buts / My Goals

<table>
<thead>
<tr>
<th>Buts identifiés pour mon enfant</th>
<th>Importance (1-10)</th>
<th>Performance (1-10)</th>
<th>Satisfaction (1-10)</th>
</tr>
</thead>
<tbody>
<tr>
<td>1 Brushing her teeth independently</td>
<td>10</td>
<td>1</td>
<td>1</td>
</tr>
<tr>
<td>2 Feeding herself</td>
<td>10</td>
<td>1</td>
<td>1</td>
</tr>
<tr>
<td>3 Having a better understanding of the lower limb surgery and what the outcome will be</td>
<td>10</td>
<td>5</td>
<td>5</td>
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</tbody>
</table>
THE NEW FACE OF RESEARCH

IS...YOU
ASK QUESTIONS, BE ENGAGED

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