

Dear Colleagues,

A warm welcome to the CDH meeting in Liverpool as part of an event programme linking closely also with the Peter Paul Rickham Symposium which celebrates the life-time achievements of an eminent paediatric surgeon who was co-founder of the World's 1st Neonatal Surgical Unit at Alder Hey Children's Hospital in 1953. At this two day workshop, a faculty of leading experts will deliver state-of-the-art lectures pertinent to clinical care and management of CDH (prenatal/postnatal/long-term follow up) including the latest advances in the basic sciences/developmental lung biology of particular relevance to the clinician/investigator(s).

Professor Paul Losty

On behalf of the Local Organising Committee

# Timetable

TIME	TUESDAY 14 NOVEMBER	
09.00 – 09.10	Welcome and opening remarks	Paul Losty (UK) / Beverly Power (UK)
09.10 – 09.35	Fetal CDH : Prenatal evaluation update and developments	Alexandra Benachi (France)
09.35 – 10.00	Fetal CDH : FETO / Latest news	Jan Deprest (Belgium)
10.00 – 11.00	Fetal CDH : Free Papers (5)	
11.00 – 11.20	Coffee Break	
11.20 – 11.50	Fetal CDH : Free Papers (3)	
11.50 – 12.20	Advanced Genetics and CDH	Mauro Longoni (USA)
	Lunch	
13.30 – 13.55	Perinatal Management and Stabilisation : Cardiac evaluation in CDH – Current status	Neil Patel (UK)
13.55 – 14.20	Perinatal Management and Stabilisation : CDH ECMO Mannheim experience	Thomas Schaible (Germany)
14.20 – 14.45	Perinatal Management and Stabilisation : CDH and ECMO service – the Glasgow experience	Carl Davis (UK)
14.45 – 15.55	Perinatal Management and Stabilisation : Free Papers (7)	
15.55 – 16.15	Coffee Break	
16.15 – 17.25	Perinatal Management and Stabilisation : Free Papers (7)	
17.25 – 18.00	CDH – British Association of Perinatal Medicine Guidelines	David Field (UK)

# Timetable

TIME	WEDNESDAY 15 NOVEMBER	
08.30 – 08.40	Surgery and CDH	Paul Losty (UK)
08.40 – 09.40	Surgery and CDH : Free Papers (6)	
09.40 – 10.05	Science and CDH : Science CDH and Nanoparticles as novel therapies	Richard Keijzer (Canada)
10.05 – 10.30	Science and CDH : Pulmonary vascular developmental biology	Richard Rottier (Netherlands)
10.30 – 11.10	Science and CDH : Free Papers (4)	
11.10 – 11.20	Coffee Break	
11.20 – 11.45	CDH Patient Outcomes : Long term outcomes	Hanneke Ijsselstijn
11.45 – 12.30	CDH Patient Outcomes : Free Papers (3)	
12.30 – 13.00	Lunch	
13.00 – 13.30	CDH Patient Outcomes : Free Papers (3)	
13.30 – 13.55	Science and CDH : Regenerative medicine	Paolo De Coppi (UK)
13.55 – 14.20	Science and CDH : New ways to support the patient	Alan Flake ( USA)
14.20 – 15.50	Science and CDH : Free Papers (9)	
15.50 – 16.05	Coffee Break	
16.05 – 16.30	Boston Children’s Hospital – A lifetime personal experience	Jay Wilson (USA)
16.30 – 16.50	CDH Registries and Networks : CDH International Registry	Kevin Lally (USA)
16.50 – 17.10	CDH Registries and Networks : CAPsNET CDH Network	Erik Skarsgrad (Canada)
17.10 – 18.00	CDH Registries and Networks : Free Papers (5)	

**CONGENITAL DIAPHRAGMATIC HERNIA  
INTERNATIONAL WORKSHOP SYMPOSIUM  
NOVEMBER 2017 – LIVERPOOL**

**List of abstracts**

**Fetal CDH**

01

**Congenital diaphragmatic hernia and diaphragmatic eventration: separate entities or two sides of the same coin?**

Francesca M Russo<sup>1</sup>, Luc De Catte<sup>1</sup>, Lennart Van der Veecken<sup>1</sup>, Michael Aertsen<sup>2</sup>, Mary Patrice Eastwood<sup>1</sup>, Anne Debeer<sup>3</sup>, Roland Devlieger<sup>1</sup>, Jan Deprest<sup>1,4</sup>

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3. Department of Neonatology, University Hospitals of Leuven, Leuven, Belgium
4. Institute for Women's Health, University College London, London, UK

02

**Measurement of pulmonary vascular volume using virtual reality three-dimensional ultrasound in fetuses with congenital diaphragmatic hernia**

N.C.J. Peters<sup>1</sup>, A.J. Eggink<sup>1</sup>, A.H.J. Koning<sup>3</sup>, E.A.P. Steegers<sup>1</sup>, D. Tibboel<sup>2</sup>, T. E. Cohen-Overbeek<sup>1</sup>

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2. Pediatric Surgery and Intensive Care, Erasmus University Medical Centre, Rotterdam, Netherlands.
3. Pathology, Erasmus University Medical Centre, Rotterdam, Netherlands

03

**Ultrasonographic assessment of mediastinal shift angle in isolated left congenital diaphragmatic hernia for the prediction of postnatal survival**

Anita Romiti,<sup>1</sup> Milena Viggiano,<sup>1</sup> Andrea Conforti,<sup>2</sup> Laura Valfré,<sup>2</sup> Francesco Morini,<sup>2</sup> Lucia Aite,<sup>2</sup> Leonardo Caforio,<sup>1</sup> Pietro Bagolan<sup>2</sup>

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04

**Imaging the fetal pulmonary vasculature in CDH**

Matthew T. Harting, MD, MS<sup>1</sup>, Jennifer Johnston, MD<sup>2</sup>, Anthony Johnson, DO<sup>3</sup>, Pamela A. Lally, MD<sup>1</sup> and Kevin P. Lally, MD, MS<sup>1</sup>

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05

**THE CARDIAC COMPRESSION INDEX AS A NEW METHOD OF PRENATAL PREDICTION OF POSTNATAL CONDITION OF NEWBORNS WITH CONGENITAL DIAPHRAGMATIC HERNIA**

A.A. Burov, Y.L. Podurovskaya, N.V. Mashinets, V.N. Demidov, A.I. Gus, V.V. Zubkov, D.N. Degtyarev, E.N. Baybarina

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06

**Outcome after fetoscopic tracheal occlusion in neonates with congenital diaphragmatic hernia – a single –center-analysis and review of literature**

K.Zahn, T.Schaible, L.Wessel – Mannheim , Germany

07

**Comparison of two case series of Congenital Diaphragmatic Hernia cases in the era of Fetal Endoluminal**

**Tracheal Occlusion: what differences?**

A. Di Cesare, A. Morandi, F. Macchini, A. Zanini, V. Gentilino, G. Farris, E. Leva

Paediatric Surgery Unit, Fondazione IRCCS Ca' Granda - Ospedale Maggiore Policlinico, Milan, Italy

08

**TRACHEAL IMPACT OF FETO PROCEDURE (FETAL ENDOSCOPIC TRACHEAL OCCLUSION) FOR CONGENITAL DIAPHRAGMATIC HERNIA: TRACHEOMEGALY AND RESPIRATORY OUTCOMES.**

A.Morandi<sup>1</sup>, F.Macchini<sup>1</sup>, M.Ophorst<sup>2</sup>, I. Borzani<sup>3</sup>, F.Ciralli<sup>4</sup>, A.Farolfi<sup>2</sup>, I.Fabietti<sup>5</sup>, N.Persico<sup>5</sup>, E.Leva<sup>1</sup>

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## **Perinatal Management and Stabilisation**

01

**INDICATORS IN-DEPTH EVALUATION OF THE OXYGEN STATUS IN CHILDREN WITH CONGENITAL DIAPHRAGMATIC HERNIA IN THE PERIOPERATIVE PERIOD**

T.A. Sokratova, A.A. Burov, Y.L. Podurovskaya, D.N. Degtyarev

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02

**TARGETED THERAPY OF PULMONARY HYPERTENSION IN NEWBORNS WITH CONGENITAL DIAPHRAGMATIC HERNIA**

A.A. Burov, D.V. Nikiforov, Y.L. Podurovskaya, V.V. Zubkov, D.N. Degtyarev, E.N. Baybarina  
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03

**Pharmacokinetics and cardiovascular tolerance of intravenous Sildenafil in newborns with congenital diaphragmatic hernia.**

Suzan CM Cochijs – den Otter, MD<sup>1</sup>, Florian Kipfmueller, MD, PhD<sup>2</sup>, Birgit Koch, MD, PhD<sup>3</sup>, Brenda de Winter, MD, PhD<sup>3</sup>, Karel Allegaert Prof MD PhD<sup>1</sup>, Dick Tibboel Prof, MD, PhD<sup>1</sup>

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04

**Prognostic role of echocardiographically measured pulmonary blood flow in neonates with CDH during ECMO support.**

Lukas Schroeder, Katrin Heindel, Suemeyra Akkas, Peter Bartmann, Andreas Mueller, Florian Kipfmueller

Department of Neonatology and Pediatric Critical Care Medicine, University of Bonn Children's Hospital, Germany

05

**Intravenous sildenafil for management of persistent pulmonary hypertension in congenital diaphragmatic hernia: a single centre experience**

Purvis P, Liaw F, Coutts J, Patel, N

Royal Hospital for Sick Children, Glasgow , UK

06

**Mortality patterns and use of extracorporeal membrane oxygenation (ECMO) in congenital diaphragmatic hernia (CDH)**

Tim Jancelewicz<sup>a</sup>, Mary E. Brindle<sup>b</sup>, Pamela A. Lally<sup>c</sup>, Matthew T. Harting<sup>c</sup>, on behalf of the Congenital Diaphragmatic Hernia Study Group

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c) University of Texas McGovern Medical School and Children's Memorial Hermann Hospital, Houston, TX, USA

07

**LUNG VOLUME OPTIMIZATION STRATEGY WITH OPTOELECTRONIC PLETISMOGRAPHY IN THE MANAGEMENT OF CONGENITAL DIAPHRAGMATIC HERNIA**

Lista G <sup>a</sup>, Bresesti I <sup>a</sup>, Cavigioli F <sup>a</sup>, Castoldi F <sup>a</sup>, Lupo E <sup>a</sup>, Lo Mauro <sup>Ab</sup>, Aliverti <sup>Ab</sup>

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b. Dipartimento di Elettronica, Informazione e Bioingegneria, Politecnico di Milano, Milan, Italy.

08

**The soluble receptor of advanced glycation end products is associated with disease severity in neonates with congenital diaphragmatic hernia.**

Florian Kipfmüller <sup>a</sup>, Katrin Heindel <sup>a</sup>, Stefan Holdenrieder <sup>b</sup>, Andreas Müller <sup>a</sup>

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09

**Flexible Bronchoscopy in Congenital Diaphragmatic Hernia**

MacLaren A, Walker G, Coutts J

Royal Hospital for Sick Children, Glasgow , UK

010

**Noninvasive measurement of ventilation perfusion mismatch for infants with pulmonary hypertension secondary to congenital diaphragmatic hernia**

Yasser N Elsayed MD, PhD<sup>1</sup>, Mary M Seshia MBChB<sup>1</sup>, Richard Keijzer MD, PhD<sup>2</sup>

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2. Pediatric Surgery, University of Manitoba, Canada

011

**High-frequency vs. conventional ventilation at the time of CDH repair is associated with, but does not contribute to higher mortality and BPD rates - A case-control study**

Gabrielle Derraugh, Matt Levesque, Suyin Lum Min, Daryl Schantz, Melanie Morris, John Minski, John Baier, Molly Seshia, Richard Keijzer - , University of Manitoba, Canada

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**Heliox Adjunct Therapy for Neonates with Congenital Diaphragmatic Hernia**

Audra Wise, MD; Mallory Boutin, MPH; Ellen Knodel, RCP; James Proudfoot, M.Sc; Brian Lane, MD; Marva Evans, MD; Denise Suttner, MD; Amy Kimball, MD

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013

**NEURALLY ADJUSTED VENTILATION IN PATIENTS WITH CONGENITAL DIAPHRAGMATIC HERNIA**

Ana Sanchez Torres – Department of Neonatology La Paz University Hospital -Madrid , Spain

014

**EPIDURAL ANALGESIA AS PART OF A THERAPY OF PULMONARY HYPERTENSION IN NEONATES WITH CONGENITAL DIAPHRAGMATIC HERNIA**

A.A. Burov, T.A. Sokratova, Y.L. Podurovskaya, E.A. Filippova, V.V. Zubkov, D.N. Degtyarev

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## **Surgery and CDH**

01

### **Pitfalls in CDH-repair: are they evitable?**

L. Wessel, T. Schaible, K. Zahn  
University Hospital Mannheim, Germany

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### **THORACOSCOPIC OPERATIONS IN NEONATES WITH CONGENITAL DIAPHRAGMATIC HERNIA IN A PERINATAL CENTER. THE RUSSIAN EXPERIENCE**

Y.L. Podurovskaya, E.I. Dorofeeva, A.A. Burov, V.V. Zubkov, D.N. Degtyarev  
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03

### **Evaluation of differences between thoracoscopic and open surgery in neonates with congenital diaphragmatic hernia with matched-pair-analysis according to relative fetal lung volume on prenatal MRI**

K.Zahn, U.Jaenicke, T.Schaible, L.Wessel – Mannheim Germany

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### **A novel technique of minimally invasive patch-implementation reduces the risk of recurrence in neonatal repair of congenital diaphragmatic hernia**

K.Zahn, T.Schaible, L.Wessel – Mannheim Germany

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### **Abdominal muscle flap repair for recurrent congenital diaphragmatic hernia in our institution**

Kina Miyoshi, Takuya Kondo, Tsuyoshi Iwanaka, Genshiro Esumi, Yoshiaki Kinoshita, Tomoaki Taguchi  
Department of Pediatric Surgery, Kyushu University

06

### **THE RISKS FACTORS OF SMALL-BOWEL OBSTRUCTION AFTER CDH SURGERY AND THE EFFECTS OF USE OF ANTI-ADHESION BARRIERS.**

Julia Boubnova<sup>\*1</sup>, Virginie Fouquet<sup>2</sup>, Guéno­lée de Lambert<sup>2</sup>, Géraldine Héry<sup>1</sup>, Catherine Jacquier<sup>3</sup>, Elisabeth Carricaburu<sup>4</sup>, Anne Schneider<sup>5</sup>, Dominique Forgues<sup>6</sup>, Marie-Odile Marcoux<sup>7</sup>, Benoît Parmentier<sup>8</sup>, Alexis Arnaud<sup>9</sup>, Quentin Ballouhey<sup>10</sup>, Sabine Irtan<sup>11</sup>, Therry Petit<sup>12</sup>, Jean-Luc Michel<sup>13</sup>, Aurélien Scalabre<sup>14</sup>, Jean-François Lecompte<sup>15</sup>, Audrey Guinot<sup>16</sup>, Guillaume Podevin<sup>17</sup>, Frédéric Auber<sup>18</sup>, Alexandra Benachi<sup>19</sup>, Laurent Storme<sup>20</sup>

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20. Lille, France

## Science and CDH

01

### **Placenta transfer of sildenafil in the ex-vivo human cotyledon perfusion model.**

Francesca M Russo<sup>1,2</sup>, Sigrid Conings<sup>2</sup>, Pieter Annaert<sup>3</sup>, Tim Van Mieghem<sup>4</sup>, Jaan Toelen<sup>2</sup>, Karel Allegaert<sup>2,5</sup>, Kristel Van Calsteren<sup>1,2</sup>, Jan Deprest<sup>1,2,6</sup>

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02

### **Amniotic fluid stem cell derived exosomes as a new therapy for lung regeneration in congenital diaphragmatic hernia**

Linda Antounians, Vincenzo D Catania, Alyssa Belfiore, Qi Ma, Adrienne Sulisty, Bo Li, Amy Wong, Janet Rosant, Augusto Zani. Hospital For Sick Children Toronto Canada.

03

### **Amniotic fluid stem cell derived exosomes promote fetal lung growth in the nitrofen model of CDH**

Linda Antounians, Vincenzo D Catania, Alyssa Belfiore, Qi Ma, Adrienne Sulisty, Bo Li, Yukhi Kioke, Augusto Zani. Hospital For Sick Children Toronto Canada

04

### **Impaired FOXF1 function causing pulmonary vascular developmental disorders; a model for CDH?!**

E. Slot<sup>a, b</sup>, D. Tibboel<sup>a</sup>, R. Rottier<sup>a</sup> & A. de Klein<sup>b</sup>

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b) Department of Clinical Genetics, Erasmus MC-Sophia Children's Hospital, Rotterdam, The Netherlands

## **CDH Patient Outcomes**

01

### **Management and Outcomes of Congenital Anomalies in Low-Middle-and High-Income Countries: Protocol for a Multi-Centre Prospective Cohort Study**

Naomi Wright RCS (Eng) Research Fellow King's Centre For Global Health and Health Partnerships London ( UK )

02

### **Prenatal versus postnatally diagnosed congenital diaphragmatic hernia – side, stage, And outcome.**

Carmen Mesas Burgosa\*, Björn Frencknera, Matias Lucob, Matthew T Hartingc, Pamela A Lally c And Kevin P Lallyc for The Congenital Diaphragmatic Hernia Study Group

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03

### **Prognostic markers and pulmonary hypertension severity in congenital diaphragmatic hernia survivors.**

Matthew Wong<sup>1</sup>, Janette Reyes<sup>2</sup>, Eveline Lapidus-Krol<sup>1</sup>, Monping Chiang<sup>1</sup>, Tilman Humpl<sup>2</sup>, Malukah Al-Faraj<sup>3</sup>, Greg Ryan<sup>3</sup>, Priscilla PL Chiu<sup>1</sup>. Hospital For Sick Children Toronto, Canada

04

### **CHARACTERIZATION OF ESOPHAGEAL MOTILITY IN INFANTS BORN WITH CONGENITAL DIAPHRAGMATIC HERNIA USING HIGH RESOLUTION MANOMETRY AND PRESSURE FLOW ANALYSIS**

Maissa Rayyan<sup>1</sup>, Taher Omari<sup>2</sup>, Karel Allegaert<sup>3</sup>, Herbert Decaluwe<sup>4</sup>, Anne Debeer<sup>1</sup>, Jan Deprest<sup>5</sup>, Nathalie Rommel<sup>6</sup>

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<sup>6b</sup> Experimental ORL, Neurosciences, KU Leuven, Belgium

05

### **Longterm Cardiopulmonary Health Following Congenital Diaphragmatic Hernia**

**Brunton A<sup>(1)</sup>, Burns P,<sup>(2)</sup> Liddell M,<sup>(1)</sup> Davis C<sup>(3)</sup>, Coutts J<sup>(1)</sup>**

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3. Department of Surgery, Royal Hospital for Children, Glasgow, Scotland

06

### **Perinatal Predictors for Self-Reported Respiratory and Gastrointestinal Outcomes in Children Managed at Fetal Treatment Centers with Isolated Congenital Diaphragmatic Hernia**

Mary Patrice Eastwood, Luc Joyeux, Laura Salazar, Francesca Maria Russo, Olga Gomez, Jordi Prat MD, Eduard Gratacos, Jan Deprest. University of Leuven, Belgium.

## **Science and CDH**

01

### **ATTENUATION OF THE REACTIVE OXYGEN SPECIES PRODUCTION BY APOCYNIN AND EPIGALLOCATECHIN-GALLATE IN THE LUNGS OF RATS WITH NITROFEN-INDUCED DIAPHRAGMATIC HERNIA**

Aras-López R<sup>1</sup>, Almeida L<sup>2</sup>, Andreu V<sup>3</sup>, Tovar JA<sup>1,4</sup>, Martínez<sup>1,4</sup>

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02

### **Profiling circular RNAs in normal and nitrofen-induced hypoplastic lung development due to Congenital Diaphragmatic Hernia (CDH)**

Shana Kahnamoui, Thomas Mahood, Richard Keijzer. University of Manitoba, Canada

03

### **Development of pulmonary neuroendocrine cells in CDH nitrofen-induced mouse model**

E. Eenjes, H.M. Kool, P. Burgisser, A. Boerema-de Munck, M. Buscop- van Kempen, D. Tibboel and R.J. Rottier. Sophia Children's Hospital Rotterdam, The Netherlands

04

### **Enhanced Pulmonary Vascular and Alveolar Development via Prenatal Administration of a Slow-Release Synthetic Prostacyclin Agonist in Rat Fetal Lung Hypoplasia**

Satoshi Umeda, Hiroomi Okuyama

Department of Pediatric Surgery, Osaka University Graduate School of Medicine  
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05

**DECELLULARIZED MATRICES DO NOT IMPROVE ON TRADITIONAL ALTERNATIVES FOR DEFECT CLOSURE IN A RABBIT MODEL OF CONGENITAL DIAPHRAGMATIC HERNIA**

<sup>1</sup>Mary Patrice Eastwood, <sup>1</sup>Luc Joyeux, <sup>4</sup>Luca Urbani, <sup>4</sup>Koichi Deguchi, <sup>1,2</sup>Savitree Pranpanus, <sup>1,3</sup>Rita Rynkevic, <sup>1</sup>Lucie Hympanova, <sup>5</sup>Eric Verbeken, <sup>4</sup>Paolo De Coppi, <sup>1,6</sup>Jan Deprest

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06

**Decellularized Piglet Diaphragm as Acellular Biologic Scaffold for Diaphragm Repair.**

Koichi Deguchi<sup>1</sup>, Luca Urbani<sup>1,2</sup>, Patrice Eastwood<sup>1,3</sup>, Martina Piccoli<sup>4</sup>, Jan Deprest<sup>3,5</sup>, Alessandro Pellegata<sup>1</sup>, Paolo De Coppi<sup>1</sup>

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07

**Muscular satellite cell and diaphragm development in congenital diaphragmatic hernia**

Nagata K<sup>1,2</sup>, Stok M<sup>3</sup>, Burgisser PE<sup>1</sup>, Eenjes E<sup>1</sup>, de Munck AB<sup>1</sup>, Taguchi T<sup>2</sup>, Tibboel D<sup>1</sup>, Wijnen RM<sup>1</sup>, Rottier RJ<sup>1</sup>

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08

**Synergistic effect of maternal sildenafil and fetal tracheal occlusion improving pulmonary development in the rabbit model for congenital diaphragmatic hernia**

Francesca M Russo<sup>1,2</sup>, Marina G Monteiro Carvalho Mori Da Cunha<sup>1</sup>, Julio Jimenez<sup>1</sup>, Mary P Eastwood<sup>1</sup>, Flore Lesage<sup>1</sup>, Tim Van Mieghem<sup>3</sup>, Jaan Toelen<sup>1</sup>, Jan Deprest<sup>1,2,4</sup>

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## **EXPERIENCE OF CLINICAL APPLICATION OF MULTIPOTENT MESENCHYMAL STROMAL CELLS IN THE TREATMENT OF NEWBORN WITH CONGENITAL DIARRHAGMAL HERNIA**

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## **CDH Registries and Networks**

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### **Towards consensus in the management of congenital diaphragmatic hernia (CDH): an overview of North American clinical practice guidelines**

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### **What we have done, and will do. From the research conducted by Japanese congenital diaphragmatic hernia study group.**

Tomoaki Taguchi, Kina Miyoshi, Kouji Nagata, Shoichiro Amari, Taizo Furukawa, Keita Terui, Tadaharu Okazaki, Naoto Urushihara, Masahiro Hayakawa, Akiko Yokoi, Kouji Masumoto, Mikihiro Inoue, Hiroomi Okuyama, Noriaki Usui. Japanese congenital diaphragmatic hernia study group

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### **Comparison of previous (2006-2010) and newly constructed (2011-2016) registry data of Japanese congenital diaphragmatic hernia study group.**

Keita Terui, Yuko Tazuke, Shoichiro Amari, Masahiro Hayakawa, Akiko Yokoi, Kouji Masumoto, Mikihiro Inoue, Hideo Yoshida, Tomoaki Taguchi, Hiroomi Okuyama, Noriaki Usui.  
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### **The role of hospital staff in educating patient families of diagnoses and how that affects patient outcome**

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Founder of CHERUBS - The Association of Congenital Diaphragmatic Hernia Research, Awareness and Support

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**Association of deprivation and CDH occurrence in a 45-year cohort in the West of Scotland**

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