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A NEW SYNDROME OR AN EXTENSION OF VATER SYNDROME?

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Background: Esophageal atresia (EA) with distal fistula, also called III type, represents the most common form of EA, with a frequency of 1 case in 4000, often associated with other congenital anomalies. VATER syndrome represents a non-random etiologically diverse association of vertebral defects, anal atresia, tracheo-esophageal fistula and radial or renal defects. We describe a novel multiple malformation pattern, characterized by tracheal stenosis, type II laryngeal web, esophageal atresia (EA) with distal fistula, vestibular fistula (VF), atrial septal defect (ASD) and patency of the ductus arteriosus (PDA), never reported in literature

Case report: Newborn with normal delivery at the end of pregnancy with polydramnios seen by an ultrasonographic examination, birth weight 2.80 Kg and length 45.6 cm. At birth Apgar score was 7 and 8 at 1' and 5' min, respectively. Insertion of a nasal-gastric probe was immediately tried, with an obstacle to progression. An immediate X-ray thorax-abdomen showed 'a stop of the radiopacal nose-gastric probe at level of D4, with a back-banding, arousing suspection of EA'. Moreover, the presence of air in the digestive tube induced to suppose the presence of a tracheo-esophageal fistula. The examination of the perineal region showed an anorectal malformation (VF) with regular canalization and immediate emission of meconio. An ultrasonographic exam of the hearth showed the presence of ASD and PDA. An optic fibers laryngscope, used for repeated episodes of crisis of apnea with cyanosis and bradycardia, showed the presence of an anterior II type laryngeal web. A nuclear magnetic resonance (MRI) pointed out the presence of a tracheal stenosis involving a wide feature of tracheal lume. Cariotype asset was examined without finding any chromosomic alteration.

Treatment: On the second day of life, the patient underwent surgery in order to correct the EA through a right posterolateral toracotomy on the fourth intercostal space. An extrapleuric surgical approach was performed and a distal tracheoesophagel fistula was isolated during the surgical exploration. A resection of the laryngeal web was performed by means of laser CO2. Unfortunately, this procedure did not give appreciable improvements of the respiratory dynamics and the patient died in sixtyfourth day of life, during a serious respiratory crisis.

Discussion: It could be considered an original syndrome like an expansion of VATER association. In fact, the concomitant presence of tracheal stenosis and EA takes place rarely, and the correlation between these two defects is little studied. If it is taken into account the concomitant presence of a laryngeal web of II type, such an association appears unique. More observations will be necessary, along with in-depth genetic studies, to determine the cause and the genetic transmission of this multiple malformation syndrome.

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SPONTANEOUS IDIOPATIC INTESTINAL PERFORATION (SIPI): A DISTINCT CLINICAL ENTITY IN THE PRETERM INFANT

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Background: The most common cause of intestinal perforation in newborns is necrotizing enterocolytis (NEC). A novel condition has been increasingly described of idiopathic spontaneous intestinal perforation (SIPI), which does not show the typical clinical and diagnostic features of NEC. The aim of the present study is to define clinical peculiarities and potential risk factors in newborns by comparing SIPI and NEC patients.

Methods: In the last 4 years 85 preterm infants necessitated surgical counselling for acute abdominal conditions. Among them, 13 underwent emergency surgery, 6 for SIPI and 7 for NEC respectively. Patients were then subdivided into 3 groups: Group 0, 72 infants, who did not need surgery; Group I,6 patients with SIPI; Group II 7 patients with NEC. Some variables were retrospectively analyzed and were compared by means of CHI 2 test with a significative value of $p<0.05$.

Results: No association emergerd between patient's sex gender ($p=0.691$), membrane ruptures ($p=0.400$) and maternal infection ($p=0.415$). A correlation was observed between pneumoperitoneum and SIPI/NEC ($p=0.000$). The study on Group I and II showes a lower gestational age (27.3 as compared to 30.28 weeks) a higher number of membrane ruptures (4/6 as compared to 3/7) and of maternal infections (4/6 as compared to 3/7). Comparative analysis showed that patients under 28 weeks of gestetional age have a 17 folds higher risk of SIPI (96%, $p=0.001$) as compared to patients with older gestational age.

Conclusions: Our study shows that a determining factor for the etiopathogenesis of SIPI may be neonatal stress consequent to preterm birth. Infants under 28 weeks of gestational age, with low birth weight (<1.200 g) show a particular predisposition to SIPI, with a risk accounting to 96% of our case series.

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ADMISSION TEMPERATURE AFTER TRANSPORT FROM DELIVERY TO NURSERY: A RANDOMISED TRIAL COMPARING RADIANT WARMER AND INCUBATOR IN PRETERM INFANTS

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Introduction: Low admission temperature has been associated with increased morbidity and mortality in preterm infants.

Aim: To compare admission axillary temperatures of preterm infants <31 weeks transported from delivery to nursery on either radiant warmers (RW) or in incubators (INC). Target axillary temperature on admission was 36.5 to 37.5 degrees C.

Method: Following delivery infants were wrapped in plastic and resuscitated under radiant warmers. When stable (5–10 minutes) they were transported on either RW (with power source) or in a pre-warmed INC with power source and air temperature 39 degrees C. Allocation for transport was by randomisation and use of sealed envelopes; randomisation was stratified by gestation. Numbers allowed for detection of 35% difference in proportion of infants with admission temperature in target range; $p<0.05$, study power 80%.

Results: Of 71 eligible infants, 64 were analysed (32 in each group). 25 were 23–25 weeks gestation, 29 were 26–28 weeks; 10 were 29–30 weeks. Median birth weight (900g) and gestation (26.3wks) were similar in the two groups ($p>0.05$). Median time to admission was 16 minutes (RW group) and 17 minutes (INC group; $p>0.05$). The 2 groups were comparable with regard to antenatal steroid use, delivery mode, number of males and 5 minute Apgar score ($p>0.05$). Median (interquartile range) of admission temperatures was 36.8 (36.5 - 37) deg C for the RW group and 36.8 (36.2–37.3) deg C for the INC group ($p>0.05$). Overall, 7 infants in the RW group and 11 in the INC group had temperatures outside the target range ($p>0.05$). Rates of other neonatal outcomes were similar ($p>0.05$) in the 2 groups.

Conclusion: Both radiant warmer and incubator transport provided satisfactory admission temperatures in most infants. The use of plastic wrap probably helped achieve these results.

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BRAIN MITOCHONDRIAL MEMBRANE DAMAGE AFTER ASPHYXIA IN FOETAL LAMBS

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Background: Mitochondria play a crucial role as the main intracellular source of energy. Mitochondria contain cardiolipin, a phospholipid mainly located in the internal membrane of these organelles, which is essential for the functionality of several mitochondrial proteins and is important for ATP synthesis.

Aim: To analyse morphofunctional alterations that occur at the mitochondrial level in a perinatal asphyxia model induced by partial umbilical cord occlusion in foetal lambs. Methods: 15 foetal lambs (80–90% GE) were used. Asphyxia was performed by partial occlusion of umbilical flow during 1h. Lambs were randomly assigned to: Control group, lambs were sacrificed immediately (0h), or managed on intermittent mandatory ventilation (IMV) for 3h; Hipoxic-isquemic (H-I) group, lambs were sacrificed immediately after asphyxia (0h) or managed on IMV for 3h. Mitochondrial membrane damage was determined by measuring the concentration of cardiolipin using the fluorochrome nonyl acridine orange (NAO) by flow cytometry. Cerebral regions were divided into: Cortex, inner zones (striatum, thalamus, hypothalamus, hippocampus), cerebellum and pons. One-factor ANOVA, $p<0.05$.

Results: Cell stained percentage is summarised in the table:(*)vs. all group

BRAIN ZONES	CONTROL		H-I	
	0h	3h	0h	3h
CORTEX	98.4 ± 2.2	95.8 ± 9.1	96.0 ± 5.6	66.9 ± 17.2 (*)
INNER	96.0 ± 7.2	95.3 ± 8.4	98.6 ± 0.9	76.8 ± 23.1 (*)
CEREBELLUM	96.9 ± 3.6	96.4 ± 2.6	94.0 ± 5.4	64.1 ± 15.9 (*)
PONS	92.7 ± 9.6	95.1 ± 4.1	96.4 ± 3.1	77.1 ± 23.4

Conclusion: In H-I animals, we can observe a significant decrease in the mitochondrial viability after 3h of resuscitation. Mitochondrial alteration is an early finding of cell damage in brain of asphyctic animals.

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OLIGOSACCHARIDES REDUCE STOOL VISCOSITY AND ACCELERATE GASTROINTESTINAL TRANSPORT IN PRETERM INFANTS

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Objective: Feeding intolerance is common in preterm infants. A mixture of prebiotic non-digestible oligosaccharides (GosFos; referring to galacto- and fructo-oligosaccharides) has been suggested to reduce stool hardness and increase stool frequency. The aim of the current study was to investigate whether GosFos would improve feeding tolerance in preterm infants on full enteral nutrition. We hypothesized that GosFos would: 1) reduce stool viscosity and 2) accelerate gastrointestinal transport.

Methods: In a placebo-controlled double-blinded trial 20 preterm infants on full enteral nutrition (gestational age 27(24–31) weeks, postnatal age 42 (11–84) days, and weight at study entry 1570 (1080–2300) g were randomly allocated to have their feedings supplemented with either GosFos (1g/100mL) or placebo for 14 days. Stool viscosity was measured by high pressure capillary rheometry. Gastrointestinal transport time was assessed as the time from feeding carmine red to its appearance in the diaper . The hypotheses were tested as a-priori-ordered hypotheses. Data are shown as median (rangeminimum-maximum).

Results: Birth weight, gestational age, postnatal age, and weight at study entry did not differ between groups. GosFos significantly reduced both stool viscosity, as measured by extrusion force (32 (2–67) vs. 158 (24–314) N), and gastrointestinal transit time (12 (4–33) vs. 26 (5–52) h).

Conclusion: Formula supplementation with GosFos improved feeding tolerance measured by reduced stool viscosity and accelerated gastrointestinal transport. No adverse effects were observed. Further trials are required to investigate whether GosFos facilitates enteral feeding advancement and early enteral nutrition thereby eventually reducing the incidence of catheter-related nosocomial infections and improving long term outcome.

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EXTREMELY LOW BIRTH WEIGHT INFANTS HAVE IMPAIRMENTS OF VISUAL PERCEPTION

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Background: Extremely low birth weight infants (ELBWI) have a high risk for cognitive dysfunction.

Objective: To characterize neuropsychological disabilities at preschool age.

Methods: In a national cohort of all ELBWI survivors, n= 203 (mean;SD: 27.3;2.1 gestational weeks, GW, birth weight 806g;136 g) born in Finland in 1996–1997, 156 children were assessed at age five with neuropsychological test, the NEPSY. Mean scores were calculated from the following domains: attention and executive function (Auditory Attention, Statue), language (Phonological Processing, Comprehension of Instructions, Speeded Naming), sensorimotor function (Imitating Hand Positions, Visuomotor Precision, Manual Motor Sequences), visuospatial perception (Design Copying, Block Construction), and memory and learning (Memory for Names, Narrative Memory, Sentence Repetition). The normal population mean test score is 10+/-3 SD.

Results: In the visuospatial main domain 84 children (54%) had a mean test score below the normal average (equal or less than 7=-1SD); in sensorimotor domain 78 children (50%); and in attention domain 72 children (47%); in language domain 45 children (29%); and in memory and learning domain 48 (31%). In the cohort, the means of subtests poorly performed were: design copying (mean score 6.4), auditory attention (mean 6,6), imitating hand positions (mean 6.8) and visuomotor precision (mean 6.9). Males performed significantly ($P<.05$) worse than females in attention and sensorimotor tasks. The means of attention and language tasks were lower ($P<.05$) in children born <27 GW compared to children born after 27 GW. In 20 children (13%) with cerebral palsy, the attention domain ($P=.001$), the sensorimotor scores ($P=.002$) and the visuospatial perception ($P<.001$) were lower than in non-CP cases.

Conclusions: Impairments of visuospatial, sensorimotor and attention functions were common in ELBW children. Visual perception and motor dysfunction were associated, which may reflect the vulnerability of the visual and motor nerve pathways located close to each other.