

(S01) OXYGENATION AND VENTILATION INDICES WITH MILD PULMONARY HYPERTENTION PARAMETERS IN SELECTING THORACOSCOPIC REPAIR FOR CONGENITAL DIAPHRAGMATIC HERNIA

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Background: Thoracoscopic repair of congenital diaphragmatic hernia (CDH) have been reported in many series of neonates and infants in the last two decades with proven safety and feasibility of the technique. Selection criteria for CDH thoracoscopic repair were based on author's personal experience.

Purpose: We aim to settle selection criteria to help in selecting CDH cases for thoracoscopic repair in limited resources countries.

Methods: Between Jan 2014 to June 2019, 54 patients with posterolateral CDH were admitted and were repaired thoracoscopically in the neonatal period .

Results: Primary repair was done in 47 cases, with 7 cases were converted to open laparotomy. The mean Peak inspiratory pressure (PIP) was 20.84 ± 2.35 (17-24 CmH₂o). The mean positive end expiratory pressure (PEEP) was 5.80 ± 0.82 (4-7 CmH₂o). The mean respiratory rate (R.R) was 37.60 ± 9.29 (27-50 Cycle/min). The mean ventilation index was 483.75 ± 110.01 (325-730). The range of oxygenation index was 4 -20. Eight cases died, all of them had P.H. (42 - 45 mmHg). On the other hand all cases (46 cases) that did not have P.H. survived. From our series, we found that there is a relation of V.I. and OI with mortality where all cases that died have high V.I. between 595 to 620 and OI more than 12.

Conclusion: Thoracoscopic repair for Bochdalek CDH is feasible and safe. Even in countries with limited infra-structure, It could be done safely with good selection of neonates. These selection data from our results for neonates with CDH with ventilation index below 550, oxygenation index below 10 and pulmonary hypertension below 40 mm.Hg can be used as parameters for excellent outcome following thoracoscopic CDH repair. These results and parameters need to be validated on a wider scale by larger series of patients.

(S02) THORACOSCOPY VS THORACOTOMY IN THE REPAIR OF ESOPHAGEAL ATRESIA WITH DISTAL TRACHEO-ESOPHAGEAL FISTULA

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AIM OF THE STUDY: Thoracoscopic repair of esophageal atresia is gaining popularity among pediatric surgeons all over the world but it is a highly technically demanding procedure. The aim of our study is to compare the surgical results and the early outcome between the thoracoscopic and the open (thoracotomy) approach for esophageal atresia repair.

METHODS: This is a retrospective bi-centric study, reviewing all the patients operated for esophageal atresia with distal tracheo-esophageal fistula. To have a homogenous population, only patients who underwent primary anastomosis were included. From 2008 to 2018, 128 patients were included. Comparison was made between the open and the thoracoscopic approaches regarding the patients' demographic data, operative time, postoperative ventilation time, length of hospital stay, postoperative complications, outcome and 1-year-follow up. Data were compared using non-parametric Mann Whitney u test, p value < 0.05 was considered significant.

RESULTS: Among the 128 patients, 41 were operated thoracoscopically (group A) and 87 were operated by the open approach (group B). Mean gestational age was 38 ± 2.36 weeks in group A and 36 ± 3.62 weeks in group B ($p=0.0008$) with a mean birth weight of 2787 ± 653 grams and 2354 ± 700 grams, in groups A and B respectively ($p=0.003$). The rate of associated congenital anomalies was comparable ($p=0.29$), but cardiac anomalies were higher in thoracotomy group ($p= 0.01$). Mortality rates during the neonatal or follow up period were 4.1% and 9.2% in groups A and B respectively ($p=0.49$).

In group A, 96% were operated within the first 72 hours of life. In group B, 78% underwent surgical intervention within the first 72 hours of life ($p=0.38$). The mean operative time was 119 ± 29 minutes in group A and 105 ± 23 minutes in group B ($p=0.0002$). No peri-operative surgical complications occurred in both groups.

The mean post-operative ventilation time and the mean length of stay were significantly shorter in the thoracoscopic group ($p=0.002$ and $p=0.001$ respectively).

The rate of post-operative complications was comparable in the both group ($p=0.25$). The incidence of anastomotic leak was 8.9% in group A vs 17.7% in group B ($p=0.18$). Anastomotic Stenosis occurred in 28.9% of group A and in 27.9% of group B ($p=0.9$). A mean of three sessions of endoscopic dilatation was needed for patients in group A during the 1st year of life compared to 2 sessions in group B ($p= 0.16$). During the 1st year of life, 13.3% patients in group A and 10.1% patients in group B required antireflux surgery for significant reflux ($p=0.59$).

CONCLUSION: Our results showed that there was a significant difference between patients in the two groups regarding the body weight, prematurity and associated cardiac malformations. However, the thoracoscopic repair resulted in uneventful post-operative outcome and short hospital stay. The results of the esophageal reconstruction are comparable to the ones of the open procedure, with the expected long-term benefits of the minimally invasive approach on the prevention of chest deformities.

(S03) SINGLE INSTITUTION REVIEW AND MANAGEMENT OF PHRENIC NERVE-DIAPHRAGM PACEMAKER FAILURES FOLLOWING THORACOSCOPIC PLACEMENT FOR CONGENITAL CENTRAL HYPOVENTILATION SYNDROME

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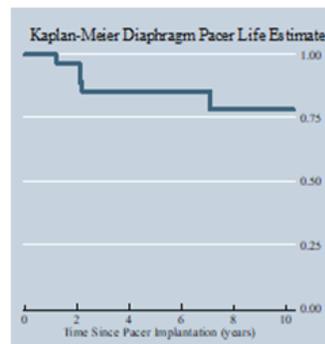
Background: Thoracoscopic placement of bilateral phrenic nerve-diaphragmatic pacer systems as an alternative means of artificial ventilation in tracheotomized pediatric patients with Congenital Central Hypoventilation Syndrome (CCHS) is now considered the standard approach to implantation. Ventilator-dependent patients benefit from such pacers as it allows negative pressure respiration and freedom from a mechanical ventilator during wakefulness. There has been no study to determine the failure rate and longitudinal management of implanted 3-part pacer components (electrode, connecting wire, receiver) in the era of thoracoscopic implantation in children.

Methods: A single institution identification of all thoracoscopically implanted phrenic nerve-diaphragm pacemaker systems (Avery Biomedical Devices, Commack, New York) in children with CCHS was performed. A subset of patients who presented with the inability to successfully pace following implantation was identified. Patient demographics, presentation of failure, radiographic imaging, age of component replaced, operative findings, component replaced, and revision results were collected. Student t-tests, Kaplan-Meier survival curves, and logistic regression were used for analyses.

Results: Fourteen patients (28 initial pacemaker insertions) were identified who met study requirements. During our study period, 5 of 14 patients presented with inability to pace due to shoulder pain or absent contraction of a diaphragm for a 17.9% overall failure rate for implanted systems (n=5/28). Mean age at implantation for children without failure was 6.6±5.0 yrs (n=9) vs. those requiring replacement of 6.1±4.9 yrs (n=5, *p*-value=0.9). Mean age of the pacer system without failure was 6.3±2.5 yrs (n=23) where the failed cohort pacer system mean age was 2.9±2.3 yrs (n=5; Figure 1, *p*-value=0.01). Adjusted for age of patient at pacemaker implantation (*p*=0.43), pacer failure was associated with pacer component age (OR 0.53 (95% CI 0.29, 0.95), *p*=0.03). Four patients had primary non-function of their pacemaker and one presented with pain that prohibited pacemaker use. Two patients had broken pacing wires identified on radiographic imaging. Six procedures were performed on these 5 patients. Two patients had replacement of their receiver and another 2 patients of their connecting wire. One patient required 2 procedures with an initial receiver change, followed by a complete replacement of the pacemaker system and scar release. All patients resumed pacing successful pacing without need for thoracotomy.

Conclusion: Patients undergoing diaphragmatic pacemaker insertion for CCHS may present with inability to adequately pace their diaphragms following insertion. Component failure is the leading cause of non-pacing. Radiographic imaging may localize the source of failure with discontinuity of the pacing wire. Receiver failure is another source of failure and isolated replacement may lead to successful pacing. Failure rate in this series is estimated at 17.9% of inserted systems without a statistical difference of failure given age of patient when implanted. Age of system has statistically higher likelihood for failure, with the older systems 47% less likely to fail with each additional year after placement. The modular pacer system allows component replacement without need for risky dissection intra-thoracically or around the phrenic nerve. Component replacement and successful diaphragm pacing may be safely accomplished with a minimally-invasive technique.

Figure 1. Kaplan-Meier Survival Curve for Diaphragmatic Pacemaker Failure



(S04) TO FREEZE OR NOT TO FREEZE: THE PECTUS SURGEON'S QUESTION.

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Introduction: Recent studies have demonstrated rapid hospital discharge and low requirements of pain control medications after bilateral intercostal nerve cryoablation for minimally invasive repair of pectus excavatum (MIRPE). However, since few surgical groups have assumed this strategy, we aim to report our experience after 44 cases with this technique in order to show its potential.

Methods: We performed a prospective registry of all patients submitted to intraoperative cryoanalgesia during MIRPE in our institution since September 2018. All patients with pectus excavatum operated during this period were included.

Technique: Selective orotracheal intubation was performed. A cryosurgery system was used to cool a probe to -70°C . Then, under thoracoscopic control, the probe was directly applied to the intercostal nerve for 2 minutes. This was done bilaterally along five intercostal spaces, from the 3rd to the 7th space with special care to avoid freezing adjacent tissues. Postoperative pain control was assessed with a Visual Analogue Scale (VAS).

Results: Forty-four patients were included. Ninety-three percent were males, the mean age at surgery was 15.8 ± 3.2 years and the mean weight was 56.2 ± 10.0 kg. The mean Haller Index was 5.6 ± 2.3 , the mean Correction Index was $41.2\pm 11.5\%$ and $16.7\pm 10.5\%$ had sternal rotation, 61% to the right and 32% to the left. Sixty-five percent had heart displacement, all to the left. Sixty-one percent of the patients received 3 bars and 39% received 2.

The mean duration of cryoanalgesia was 38.1 ± 14.7 minutes. None received epidural anesthesia. There were no intraoperative complications. The mean length of stay was 1.6 ± 0.9 days postoperative. Analysis of the median VAS in postoperative days 1, 2, 7 and 21 were 2, 4, 2, 1 and 0.

Conclusions: Cryoanalgesia during MIRPE allowed a rapid hospital discharge with very good pain control in all cases. Cryoanalgesia has become our first choice of treatment for pain control in the thoracoscopic correction of pectus excavatum.

(S05) THORACOSCOPIC REPAIR OF LONG GAP ESOPHAGEAL ATRESIA. WHEN SHOULD WE TRY? WHEN CAN WE SUCCEED?

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Introduction: Esophageal atresia (EA) occurs in 1/2500 to 1/400 live births. According to the available literature patients diagnosed with long gap esophageal atresia (LGEA) comprise about 10% of this population. Treatment of this group of patients remains one of the greatest challenges in paediatric surgery. Due to the continuous development of minimally invasive surgical techniques, we can restore the native esophagus via the thoracoscopic approach.

The aim of the study was to analyze available video records of thoracoscopic repair of LGEA performed by one of the authors (D.P). We focused on anatomical factors that may impact on the final success defined as thoracoscopic anastomosis.

Materials and methods: We performed a retrospective review of video and medical records of 25 patients diagnosed with LGEA in five centers in Poland from June 2010 to August 2019. All recorded thoracoscopies were meticulously analyzed to assess the anatomy of the right hemithorax and determine the localization of esophageal ends and the gap between them in relation to vertebral bodies. Patients were divided according to the type of atresia and outcome (group 1 - success, group 2 - failure). Gross classification was used to define the type of atresia. Mann Whitney U test and Fisher exact test were used for statistical analysis.

Results: All patients except one underwent staged thoracoscopic repair with internal traction technique to elongate the esophageal ends.

Among 25 patients 15 were diagnosed with "pure" type A atresia and 10 were classified as type B. In 20 (11 type B and 9 type A) patients we succeeded with thoracoscopic anastomosis. Four out of five patients who failed thoracoscopic treatment had type A atresia. The success rate was similar in both types 73% type A vs 90% in type B. Every video record was suitable for accurate assessment of thoracoscopic anatomy. We found no significant difference in esophageal end localization and the gap between types A and B. There was a significant difference between lower end position and gap between failure and success group ($P < 0,02$ and $p < 0,01$ respectively). We achieved primary anastomosis in every patient with a lower end located above Th9. In the failure group, each patient's gap was longer than 7 vertebral bodies. Median time from birth to anastomosis was 49 days. Median time from the first surgery to anastomosis was 28 days.

Conclusions: Assessing the anatomy of esophageal atresia is feasible with thoracoscopy and can be a basis for a further tailor-made treatment plan. Lower end position, as well as relative gap length, seem to be good predictors of final outcome. The thoracoscopic approach enables to repair the esophagus in a short period of time. Thoracoscopy can be performed in every case of LGEA, at least as a diagnostic procedure in the first stage. Further studies on a larger group of patients are necessary to define a group of patients feasible for successful thoracoscopic treatment.

(S06) BACTERIAL AIRWAY MICROBIOME, VOLATILE ORGANIC COMPOUNDS AND EXERCISE PERFORMANCE OF PATIENTS AFTER SURGICAL REPAIR OF CONGENITAL DIAPHRAGMATIC HERNIA

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Introduction: Even after surgical repair many patients with congenital diaphragmatic hernia (CDH) suffer from lifelong pulmonary sequelae. Recurrent respiratory infections and reduced physical performance capacity have been reported. Over many years the peripheral respiratory tract was assumed sterile. However, recent microbiological advancements demonstrated that the lung harbors a diverse array of microbes, whose dynamic composition is influenced by host and environmental factors. Volatile organic compounds (VOCs) can be detected in exhaled breath samples and allow reflections on bacterial and human host metabolism. In CDH patients airway microbiome, VOC profile, lung function and performance capacity in CDH patients have never been compared healthy age and sex-matched controls.

Patients and Methods: After ethical approval all patients treated for CDH at our department (age 6-18 years at follow-up) were invited to participate in this study. A total of 9 patients (median age 11 years) could be recruited. Lung function testing was performed as nitrogen multiple breath washout body plethysmography. The physical performance was evaluated by exhausting bicycle spiroergometry. Airway microbiome was determined from deep induced sputum by 16S rRNA gene sequencing. VOCs were sampled by needle trap microextraction in the end-expiratory phase and measured by GC-MS.

Results: There was no difference in size, body weight and BMI between the two groups. CDH patients showed significantly reduced Tiffenau Index ($p=0.028$). Body plethysmography revealed significantly increased residual volume/total lung capacity (RV/TLC; $p=0.008$) and significantly decreased mean expiratory flow (MEF25-75%; $p=0.036$). Additionally, there was a trend towards higher RV and Scnd in the CDH group. Spiroergometry showed no statistically significant differences between the groups. Microbiome analysis revealed no statistically significant differences for alpha-diversity and beta-diversity. CDH patients exhibited significantly lower relative abundances of Pasteurellales ($p=0.038$) and Pasteurellaceae ($p=0.038$) compared to healthy controls. Exhaled VOC profile showed significantly higher levels of Cyclohexane ($p=0.004$) and significantly lower levels of Acetone ($p=0.002$) and 2-Methylbutane ($p=0.038$) in CDH patients.

Conclusion: This is the first study to report on airway microbiome and VOC profile in CDH. Alterations of the microbiome were minor and the clinical consequence of reduced Pasteurellaceae remains unclear at present. Elevations of Cyclohexane observed in our CDH group have also been reported in cases of lung cancer and pneumonia. The majority of our CDH patients showed signs of an obstructive pulmonary disease. CDH patients had no signs for impaired physical performance capacity adding further data to controversial reports in the literature in this regard. Future larger scale multi-center studies will be required to confirm these first results.

**(S07) LONG-TERM OUTCOME AFTER MINIMAL-INVASIVE REPAIR OF CONGENITAL
DIAPHRAGMATIC HERNIA CAN BE IMPROVED BY TECHNICAL ADJUSTMENTS**

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Purpose: In literature higher recurrence-rates have been reported for minimally invasive surgery (MIS) compared to open surgery (OS) already during the first hospital-stay. Also, higher recurrence-rates have been reported after MIS-patch-implantation and therefore the longterm-efficacy has been questioned. The purpose of this study was to critically review all patients operated within 10 years to draw conclusions how to improve long-term-efficacy.

Methods: Follow-up-data was collected prospectively within a structured follow-up program and regular radiologic imaging was performed to screen recurrence. Results of patients with a minimal follow-up of two years are presented. Fisher's exact test was used for analysis.

Results: Between 2008 and 2017 355 patients were operated by OS, 101 by MIS and 29 converted from MIS to OS. After exclusion of patients, who deceased, received ECMO-therapy or were lost to follow-up, 95 MIS-patients were compared to 173 nonECMO OS-patients. Patch-rate was 25% in MIS and 81% in OS ($p < 0.000001$). In MIS recurrence-rate was 15.5% after primary and 12.5% after patch-repair ($p = 1.0$). In OS it was 3.1% after primary and 7.1% after patch-repair ($p = 0.7$). Nevertheless the difference in recurrence-rate for primary and patch repair between MIS and OS was not significant (primary repair: $p = 0.1$; patch-repair: $p = 0.4$). Due to the high recurrence-rate in MIS a few technical adjustments were made since 2014. Comparing 58 MIS-patients before to 37 MIS-patients after these changes, recurrence-rate dropped from 20.7% to 5.4% ($p = 0.07$). We observed only one in-hospital recurrence. In MIS most recurrences developed within the first year of life and all were symptomatic (67%). Older children showed no or unspecific symptoms and were diagnosed on follow-up-visits. One child deceased due to unrecognized recurrence with bowel gangrene and lethal septicaemia. No small bowel obstruction due to adhesions was observed after MIS, one patient developed a partial volvulus requiring surgery.

Conclusion: Careful patient-selection is essential for favourable long-term outcome in MIS-repair of congenital diaphragmatic hernia. Hernial sacs should be resected. It is crucial to reduce tension on the diaphragm and preferable to implant a patch in patients with missing lateral diaphragmatic rim. It is essential to promote scarring of the diaphragm in primary repair and adhesions between prosthetic material and diaphragm to prevent recurrence, because patients do not develop intestinal adhesions. Thus small recurrences bear the risk of intestinal incarceration, bowel gangrene and lethal septicaemia. Long-term follow-up with regular radiologic imaging and detailed parent-counseling is therefore mandatory. With rising experience and meticulous technique recurrence-rates similar to those in open surgery seem to be achievable. Yet longterm follow-up until adulthood has to be awaited for final judgement on efficacy.

(S08) HAVE YOU EVER HEARD ABOUT ARCHAEA? PROBABLY NOT! SO WHAT ARE THESE MICROBES DOING IN THE DEEPER AIRWAYS OF CHILDREN FOLLOWING ESOPHAGEAL ATRESIA REPAIR?

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Introduction: Archaea, formerly termed archaeobacteria, constitute a separate domain of single cell organisms that are biologically different to bacteria. One of their unique features is methanogenesis. Until now there are no known pathogens in the domain of archaea. While archaea have been known to inhabitate the gastrointestinal tract for several decades, their occurrence in the pulmonary tract especially in children remains unknown. For the first time ever the archaeome was assessed in patients following esophageal atresia (EA) repair and compared to a healthy age- and sex-matched control group.

Methods: Patients (>12years) following correction of EA were invited for a long-term follow-up examination. Deep induced sputum samples were collected. Samples were subjected to DNA extraction, archaea-specific amplicon generation (16S rRNA gene) and next generation sequencing (Illumina Miseq; Pausan et al., 2019). Raw reads were bioinformatically processed (Qiime2), and results were compared to an age- and sex-matched control group consisting of healthy patients recruited from the families of the medical staff.

Results: 19 patients (9 female, 10 male) following EA with a mean age of 24.7 years (range 14-40) could be recruited. The control group consisted of 19 healthy patients (9 female, 10 male). While archaea could be found in 6 out of 19 EA patients (32%), archaeal signatures were present in only one out of 19 control patients (5%). In the EA patient the signatures belonged to Methanobrevibacter, a group of Euryarcheota, in five patients and a mixture of Methanobrevibacter and Candidatus Nitrosotenuis (a genus of Thaumarchaeota, potentially aerobic ammonia-oxidizing archaeon) in one patient. In the control patient Candidatus Nitrosotenuis was found.

Conclusion: This is the first study to reveal archaea in the airway microbiome of almost a third of the patients following correction of EA. Our findings and their clinical implication have to be further examined in larger scale multi-centric studies.

(S09) THE USE OF INDOCYANINE-GREEN FLUORESCENCE IMAGING FOR PERFUSION ASSESSMENT OF COMPLEX PEDIATRIC ESOPHAGEAL ANASTOMOSIS

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Background: Blood flow is critical for healing of esophageal anastomoses. We sought to evaluate the use of indocyanine-green (ICG) fluorescence imaging to assess the perfusion of esophageal anastomoses in children.

Methods: We reviewed children who underwent a complex esophageal anastomosis at our institution and had an ICG perfusion assessment of their anastomosis. We evaluated changes in intra-operative decision making directly attributed to ICG perfusion assessments. From the video recordings of each assessment, we defined perfusion features to develop a scoring system. Anastomotic perfusion scores were compared between patients with favorable and poor anastomotic outcomes. Poor anastomotic outcome was defined as a clinically significant leak, need for ≥ 6 dilations in first year post-anastomosis, need for advanced endoscopic therapy (i.e. stenting, endoscopic-vacuum assisted therapy [e-vac]) or stricture resection.

Results: In a one-year period, 43 children (53% female), median age of 13 months (IQR: 5-31 months), with history of esophageal atresia (EA; n=40, 40% type A or B), caustic esophageal injury (n=1), esophageal peptic stricture (n=1), or acquired trachea-esophageal fistula (n=1), underwent 45 complex esophageal anastomoses. Procedures included primary (n=6) or delayed anastomosis after traction (Foker; n=15), stricture resection (n=10) or plasty (n=2), jejunal interposition (n=7) or primary repair of esophageal leak (n=1) or trachea-esophageal fistula (n=2). We encountered five instances of changes in intraoperative plan that were directly attributed to ICG assessments which ranged from imbrication or reinforcement of a poorly perfused area, to converting from a stricture resection to a jejunal interposition. Thirty-four (76%) videos were suitable for anastomotic perfusion scoring. Perfusion features identified included: strength and speed of perfusion signal uptake, extent of hypoperfusion, symmetry between each side of the anastomosis, and the width of the anastomotic ischemic penumbra (Figure). Median anastomotic perfusion score was 15, (range 8-20, possible scores 3-20). With a median follow up of 4 months (IQR: 2-10 months) 9 (20%) patients experienced poor anastomotic outcomes (6 leaks, 4 with ≥ 6 dilations, 4 stents, 4 e-vac, 4 stricture resections; patients often fell into more than one category). Anastomotic perfusion scores were significantly lower in patients who experienced a poor anastomotic outcome versus those who did not (mean \pm SD; 11.9 \pm 3 vs. 15.6 \pm 2.4, p=0.009). Greater anastomotic perfusion scores were significantly correlated with lesser risk for poor anastomotic outcome (r=0.29, p=0.001); similarly, greater perfusion scores were associated with fewer number of endoscopic dilations in the first year post-anastomosis (p=0.02). Patients with an anastomotic perfusion score of >12 had a 70% decrease in risk (relative risk 0.3, p=0.02) of a poor anastomotic outcome.

Conclusions: The use of indocyanine-green fluorescence imaging is useful to assess the perfusion of complex pediatric esophageal anastomoses. This technology provides real-time information that can help identify anastomotic areas of hypoperfusion which can be addressed intraoperatively to decrease the risk of anastomotic complications. Furthermore, our novel anastomotic perfusion scoring system helps identify anastomoses at risk for poor outcome. Future validation efforts of this scoring system in other settings and types of anastomoses are needed to corroborate these findings.

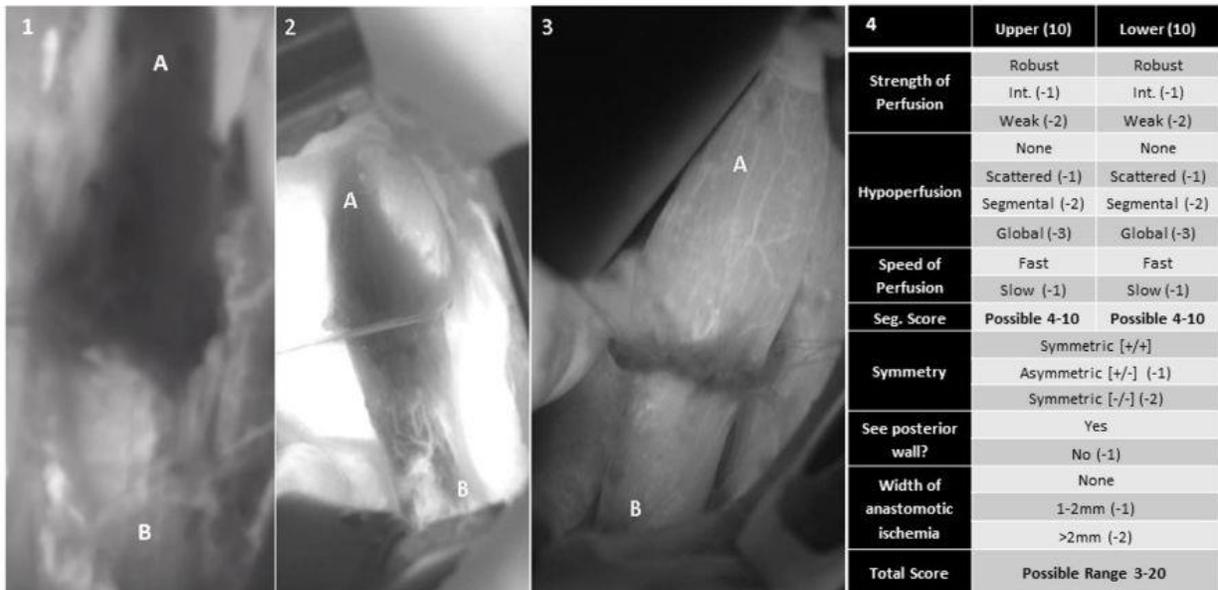


Figure (Panels 1, 2, 3, 4): Examples of different ICG perfusion assessments. Panel 1= patient with history of caustic esophageal stricture undergoing a stricture resection with primary anastomosis with resulting global hypoperfusion of upper esophageal segment (A), perfusion score of 10. Panel 2= Patient with a history of long gap esophageal atresia (LGEA) with anastomosis after Foker procedure with a segmental hypoperfusion area of the upper esophageal segment (A) and scattered hypoperfusion of the lower segment (B), perfusion score of 8. Panel 3= Patient with a history of LGEA after Foker procedure with excellent perfusion throughout, perfusion score of 19. Panel 4= Perfusion assessment scoring system, each segment starts with a max score of 10 and as signals of hypoperfusion are identified these detract negative points to generate an upper and lower esophageal segment score, these segments are then compared with regards to their degree of symmetry to generate a total anastomotic perfusion score (range 3 to 20).