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Gastroschisis In Consecutive Siblings: A Rare Occurrence

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Introduction: Gastroschisis presents most commonly as a right lateral umbilical abdominal wall defect and occurs at an estimated 1 in approximately 3000 births. Environmental factors such as socioeconomic status, lack of prenatal care, young maternal age, and illicit drug use or alcohol consumption have been implicated as causative factors. Excluding genetic syndromes, familial predisposition has not been observed. In this case report, we present the unique case of brothers born consecutively with gastroschisis without a genetic disorder.

Case presentation: 32-week-old male, born at 2.0 kg in weight, was found to have right lateral gastroschisis upon birth and transferred to the neonatal intensive care unit. Upon examination with the pediatric surgery team, the bowel was pink, healthy and without evidence of stricture. Of note, the mother did not have prenatal care prior to birth. A 5cm silo was placed bedside in sterile fashion to protect the bowel. Patient underwent fascial closure 1 week later. . The patient's clinical course was complicated by intestinal failure with a jejunal stricture who underwent exploratory laparotomy, stricture resection, appendectomy, G-tube placement and made full recovery. Upon the mother's next pregnancy by the same father, she was encouraged to have proper prenatal care. At this time, fetal ultrasound demonstrated another right sided gastroschisis. Postpartum, the baby was evaluated by the pediatric team on day 0 of life, which demonstrated healthy, pink bowel and had a silo placed. He underwent formal closure of fascia on day 6 of life, recovered well and was discharged on ad lib feeds. Our mother denied use of tobacco, alcohol, or illicit drug use during both pregnancies.

Discussion: Gastroschisis has been noted in both identical and dizygotic twins but is rarely reported in siblings consecutively. Considering our mother denied any teratogenic factors that may have influenced the development of this pathology, it may suggest an occult genetic predisposition or an unknown environmental factor contributing to the development of gastroschisis in these siblings. However, this mother and father have five children and the other three did not suffer from gastroschisis. This raises the question of counseling for future pregnancies that may be complicated by this pathology and necessity for prenatal care.

Impact Of Frailty On Complications And Length Of Stay After Minimally Invasive Adrenalectomy Surgery

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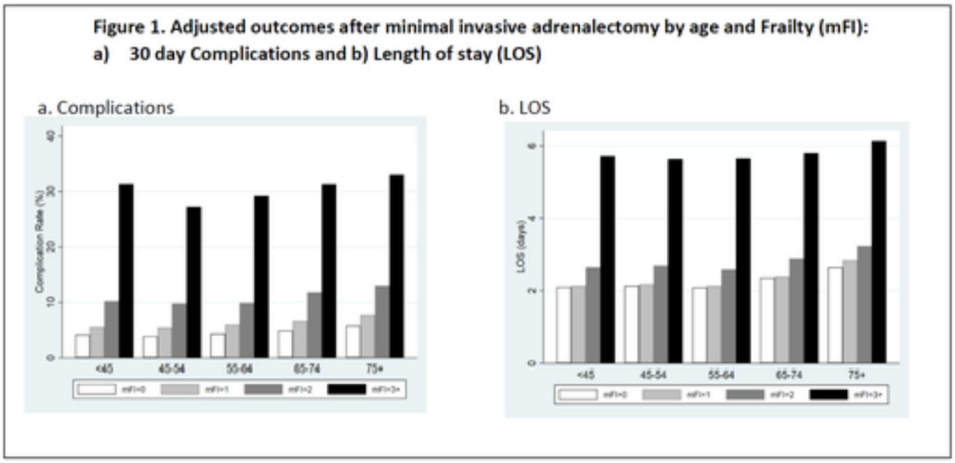
Introduction: The incidence of adrenal disease requiring surgery increases with age and a minimally invasive procedure is a reasonable treatment option for the elderly population. Frailty, independent of age, is an important predictor of operative risk among surgical patients. This study evaluates the perioperative outcomes of elective adrenalectomies when performed in elderly patients and how frailty score affects such outcomes.

Methods: Patients undergoing minimally invasive adrenalectomy (MIA) were identified using ACS NSQIP targeted adrenal surgery PUF files years 2005-2020(n=10,197). The surgical indication was categorized: benign disease, endocrine disorder, and malignant disease. Frailty was defined using the 5-item modified frailty index (mFI). Simple and multivariable regression was used to model the relationship of age and frailty to 30-day mortality, complications, postoperative length of stay(LOS), and readmissions, after adjusting for race, sex, year of surgery, ASA, emergency case status, and surgical indication.

Results: MIA accounts for 85.4% of all cases (n= 8,710), has been stable from2005-2020, did not vary by race, was least used in patients over 75 years old (78.3%), and most used in those <55 years old (86.9%) (p<0.001). In MIA patients,5,291 (60.8%) were female, 5,037 (57.8%) were white, and 1,928 (22.1%) over 65years old. Indications for surgery were benign disease (63.1%), endocrine disorder (32.8%), and malignancy (4.1%). Patients age<65 compared to those age>65 years old were more likely to have a mFI = 0 (25.8% vs 13.5% respectively), and less likely to have a mFI = 3+ (1.8% vs 3.7% respectively) (p< 0.001). Outcomes: 30-day mortality 0.2%, complications 5.8%, return to OR 0.8%, and median LOS 2days. 30-day mortality was associated with frailty (p=0.009) and endocrine disease(p=0.006), but not with age. Complications were associated with frailty (p<0.001) and malignant disease (p=0.012), but not with age. Return to OR was associated with age 75+ (p=0.020), but not with frailty. Post-operative LOS was associated withage 65+ (P=0.009), frailty (P<0.006), and malignant disease (p<0.001). Figure 1shows the adjusted relationships between age and frailty with complications and LOS after MIA procedure.

Conclusion: MIA has low 30-day mortality and complication rates that do not increase with age but do vary with frailty. Frailty is a better predictor than the age of most adverse outcomes after MIA, except for the return to OR which is only associated with age 75+.

Figure 1. Adjusted outcomes after minimal invasive adrenalectomy by age and Frailty (mFI): a) 30 day Complications and b) Length of stay (LOS)



Effect of Right Ventricle to Pulmonary Artery Conduit Size on Outcomes Following Primary Repair of Congenital Heart Defects

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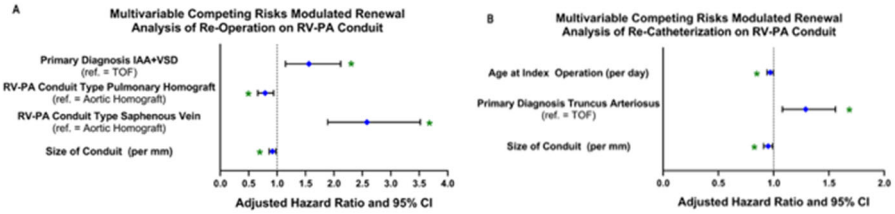
Introduction: Reconstruction of right ventricle to pulmonary artery (RV-PA) continuity is an integral part of commonly performed surgical procedures in neonates and young infants with congenital heart defects. The aim of this study was to assess differences in clinical outcomes among patients with congenital heart defects who underwent RV-PA conduit placements during initial repair using multivariable time-to-event competing risks analysis with modulated renewal for repeated interventions.

Methods: Surgical and follow-up data were retrospectively collected from medical records and/or follow-up from referring cardiologists in patients who underwent RV-PA conduit placements during initial biventricular repair of congenital heart defect between January 1980 and September 2021. A modulated renewal Fine and Gray competing risks regression analysis was applied to account for multiple conduit replacements/reinterventions from the same patient and mortality as a competing risk.

Results: In the four-decade period of 1980-2021, 874 patients underwent an RV-PA conduit placement. Median age at implantation was 116 (IQR 11.4-846.3) days with a primary diagnosis of tetralogy of Fallot in 436 (50%) and truncus arteriosus in 214 (25%). Median follow-up was 7.4 years and overall mortality was 14%. Conduit types included aortic homograft (n=469, 54%), pulmonary homograft (n=286, 33%), Gore-Tex, (n=23, 3%), Contegra (n=15, 2%), and saphenous vein graft (n=8, 1%). Overall reoperation occurred in 543 cases (62%) and reoperation on the RV-PA conduit occurred in 454 patients (52%). With death as a competing risk and modulated renewal for repeated conduit reinterventions, larger conduit size (hazard ratio [HR] 0.92; 95% CI 0.89-0.94, P<0.001) and pulmonary homograft (HR 0.8; 95% CI 0.7-0.9, P=0.008) had a protective effect, while use of saphenous vein (HR 2.6; 95% CI 1.9-2.5, P<0.001) and diagnosis of interrupted arch (HR 1.6; 95%CI 1.1-2.1, P=0.004) were significantly associated with reoperation on the RV-PA conduit. A diagnosis of truncus arteriosus (HR 1.3; 95% CI 1.1-1.6, P=0.006) was significantly associated with re-catheterization on the RV-PA conduit, while larger conduit size (HR 0.95; 95% CI 0.93-0.98, P=0.001) and older age at index operation (HR 0.97; 95% CI, 0.95-0.99, P=0.003) were associated with significantly lower risk.

Conclusions: Right ventricle to pulmonary artery conduit reoperations/reinterventions are common. Larger conduit size and use of pulmonary homograft were protective while use of saphenous vein conduit was associated with repeated reoperation; older age and smaller conduit size were associated with catheterization interventions of the conduit. Appropriate sizing of conduit and use of pulmonary homograft may delay time to first reintervention and reduce the number of reinterventions.



For MALS Patients, Reoperation is Safe and Effective

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Introduction: Median arcuate ligament syndrome (MALS) is due to congenital variation in the anatomic position of the median arcuate ligament. The low-lying median arcuate ligament exerts compression on the celiac artery and surrounding celiac plexus. Patients with MALS have significant epigastric pain, which may be associated with nausea, vomiting, and weight loss. Long-term success rate of pain relief after median arcuate ligament release varies greatly, and recurrence of pain is frequent. The surgery we perform is unique in that we resect the celiac plexus and median arcuate ligament, in contrast to division of the median arcuate ligament alone. Our goal in this study is to assess the long-term impact of this procedure and to determine if the results we are noting immediately postoperatively are durable. Herein we will be analyzing operative outcomes in patients who had previously undergone unsuccessful operative correction of MALS at different institutions.

Method(s): After institutional review board approval, a 17-item follow-up questionnaire was sent electronically to all patients who underwent resection of median arcuate ligament and celiac plexus at Stamford Hospital. We focus on outcomes for a unique sub-population of 22 patients who had undergone prior operations to treat MALS pain. Descriptive statistics and frequencies on outcomes of interest are presented.

Results: A total of 503 surveys were sent to patients who underwent operative correction of MALS at Stamford Hospital since January of 2019. Patients who underwent operation within the past 3 months were excluded. Out of 222 patients who responded,

22 self-identified as having undergone prior operative interventions to treat MALS. 15 of these patients underwent laparoscopic repair prior to surgery at Stamford Hospital. All patients underwent celiac ganglion block prior to operation. 12 patients reported complete relief of pain after block, 8 patients reported partial relief of pain after block. After MALS corrective surgery, 77% reported a reduction in upper abdominal pain, with 68% reporting their ability to eat improved. In addition, almost 70% reported attendance at work and/or school improved, with 59.09% of patients reporting complete resolution of MALS pain. 8/22 patients reported recurrence of MALS related pain after operation, most reported pain recurrence within the first 6 months after surgery.

Conclusions: In reviewing our experience in treating MALS patients, we identified an important subset of patients who had previously undergone operative correction of MALS without symptom relief. Existing literature quotes success rates for MALS surgery to be approximately 30 – 60%. The operation we perform focuses on resection of the celiac nerve plexus which we believe is the cause of MALS pain. This initial analysis demonstrates pain relief rate of nearly 80% in a group that had failed prior median arcuate ligament release. This outcome is significant and reflects the importance of resection of celiac plexus to improve pain relief rates in these patients. This may indicate that the etiology of pain in MALS is neurogenic, not ischemic as previously thought.

Mechanical vs Chemical VTE Prophylaxis in Minimally-Invasive Adrenalectomy Patients: Which is best?

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Introduction: With the advent and subsequent mainstream use of minimally invasive techniques for adrenalectomies, there has been a significant decrease in the surgical complications previously associated with open procedures. However, data shows that in the percentage of patients that have significant post-operative adverse effects, venous thromboembolism (VTE) events and bleeding requiring transfusion are fairly common. The protocols for VTE prophylaxis are varied and often institution-dependent, and there is a paucity of current data comparing mechanical and chemical VTE prophylaxis in patients undergoing minimally-invasive adrenalectomies, particularly via a retroperitoneoscopic approach.

Methods: We performed a retrospective cohort study evaluating all minimally-invasive adrenalectomies performed at our institution from 2014-2022 by experienced adrenal surgeons, who primarily use a retroperitoneoscopic approach, with mechanical VTE prophylaxis as the preferred method of prophylaxis. All patients >18 years of age who underwent either a retroperitoneoscopic or transabdominal laparoscopic adrenalectomy were included. Exclusion criteria were patients with a diagnosis of Cushing's syndrome or adrenocortical carcinoma. The median patient age was 50.96 years, with a median BMI of 31.17 (26.47, 36.35). We compared our results to nationally available data through the ACS-NSQIP database, against cases where chemical prophylaxis was used for VTE prevention. We then evaluated the incidence of specific outcomes, including VTE, bleeding requiring transfusion, hematomas, and 30-day readmission and mortality rates.

Results: We found a VTE incidence rate of 0.2% and a bleeding rate of 0.7% in our cohort. There was no significant difference in VTE rate between our cohort and the ACS-NSQIP chemical prophylaxis cohort. The was a significantly higher incidence of bleeding complications in the ACS-NSQIP group. There were no differences in either 30-day readmission or mortality rates between the groups.

Conclusion: We conclude that both mechanical and chemical VTE prophylaxis are effective means of prophylaxis in minimally-invasive adrenalectomy procedures. We propose that surgeons tailor the decision to provide VTE prophylaxis based on the individual risk profile of the patient in question.

Neonatal Congenital Segmental Intestinal Dilatation Causing Gastric Outlet Obstruction

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Introduction: Congenital segmental intestinal dilatation (CSID) is a rare condition characterized by an isolated portion of the gastrointestinal tract that abruptly transitions between normal and dilated bowel. CSID most commonly affects the ileum and colon, and infrequently the duodenum and jejunum.[1] Neonates typically present with abdominal distention and bilious emesis

secondary to a lower gastrointestinal tract obstruction, while older patients may present with abdominal pain and failure to thrive. Associated anomalies include anorectal malformations[2] and cardiovascular defects.[3] The diagnosis of CSID is challenging due to the lack specificity of radiological imaging and its similar presentation to other pediatric disorders such as malrotation, intestinal atresia, and Hirschsprung's disease. In this case report, we present a unique patient who presented with CSID causing gastric outlet obstruction.

Case: The patient was a male neonate born at full term by uncomplicated vaginal birth to a 29-year-old G2P2 mother. Within the first day of life, the patient was transferred to the NICU due to frequent non-bilious emesis. He had passed meconium and no anorectal malformation was identified on exam; however, he exhibited a persistent inability to tolerate feeds. Abdominal radiography on day two showed a mildly distended stomach and a loop of dilated bowel in the right upper quadrant, likely a meconium-filled transverse colon, and air in the rectum.

Despite decompression with a Replogle tube, bowel rest and suppositories, the patient continued to experience persistent abdominal distention. Barium enema on day four demonstrated dilation of the colon at hepatic flexure without evidence of Hirschsprung's disease, small left colon syndrome, or distal obstruction. Follow-up serial abdominal films demonstrated persistence of a dilated air-filled segment of colon. After failure of progression by day six, the decision was made to bring the patient to the OR. An exploratory laparotomy was performed via a right upper quadrant transverse incision. A10cm segment of significantly dilated right colon was identified, without palpable stool or meconium, that was consistent with the dilatation seen on preoperative imaging. This dilated segment was resected, followed by a primary hand-sewn two-layer anastomosis of the proximal ascending to transverse colon. After determining that the anastomosis was widely patent, and that the remaining bowel appeared normal, the incision was closed and the patient was extubated.

Feeds were resumed at a low rate on post-operative day one and TPN was continued. The patient tolerated a slow diet advance over the coming days and began passing regular stools. On post-operative day seven, the patient was discharged home. Final pathology revealed normal ganglion cells at both margins of the resected bowel. The patient continued to do well on follow-up visit with appropriate feeding and stooling.

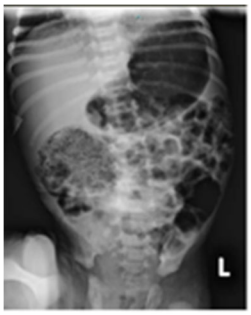

Figure 1: Abdominal Xray

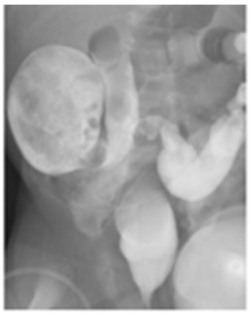
Figure 2: Barium Enema

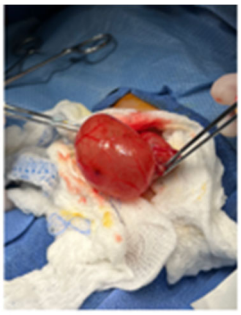
Figure 3: Dilated Colonic Segment

Discussion: To our knowledge, our patient is the first case in literature to present with CSID causing gastric outlet obstruction. The dilated colonic segment in our neonate was positioned in the RUQ at the hepatic flexure. Its location caused extrinsic compression on the proximal duodenum, resulting in a mechanical impediment to gastric emptying. Histopathologic analysis of the resected colon showed no masses, lesions, or evidence of aganglionosis. This is consistent with other reports of CSID which have predominantly found normal cells on histology.[4] The cause of CSID is unknown, though several hypotheses have been proposed. It has been postulated by some that prolonged intrauterine compression on a loop of bowel at both ends results in segmental dilatation[5,6], while other authors implicate a disturbance in the embryological division of the notochord from the ectoderm.[7] Fortunately, the treatment of CSID is a simple resection of the affected segment. Nevertheless, the etiology of CSID remains unclear, and further research investigating its underlying pathogenesis is necessary to better understand this rare developmental anomaly.

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Open Aortoiliac Vascular Surgery - A Dying Art: Successful Retrieval of a Retained Endovascular Sheath

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Introduction: Peripheral angiography is a widely used diagnostic modality to assess lower extremity arterial vasculature. The predominant access sites for peripheral vascular interventions are the common femoral artery, followed by the radial artery. Transfemoral approach remains the preferred access modality. The incidence of femoral access site complications varies widely, from 0.7% to 9%. Reports of catheter fracture and endovascular retrieval have been made in the context of coronary angiography and radial artery access. We present a case of a sheath that was torn while performing a lower extremity arterial intervention that was retrieved using an open retroperitoneal approach, since endovascular retrieval was not feasible. To our knowledge, this is the first report of open vascular sheath retrieval.

Method: An 83-year-old man with left leg critical limb ischemia underwent a left leg angiogram using the right common femoral artery approach. A 5 French x 90 cm Cook hydrophilic sheath would not advance beyond the left mid superficial femoral artery and it was decided to use a shorter sheath. During removal of the sheath, it unraveled and tore in the scarred right groin. Localized right groin cutdown was performed which confirmed the fractured sheath to be intravascular. Snaring the sheath was not a possibility. The sheath was neither advancing nor it was coming back over the wire. The patient was transferred to the operating room and a left retroperitoneal dissection was performed. Left common iliac, internal iliac and external iliac arteries were dissected but were found to be circumferentially calcified. Dissection was continued onto the infrarenal abdominal aorta up to the level of the inferior mesenteric artery. The aorta was densely calcified with no clampable site. The external iliac artery had 2 partially clampable sites that were not ideal. Left infraclavicular percutaneous axillary artery access was obtained but on angiogram it was discovered that his left subclavian artery was occluded which ruled out the possibility of balloon occluding the aortic bifurcation. Wire access through the broken sheath was maintained throughout the case. A 6-French sheath was inserted over the wire into the right common femoral artery. Fogarty clamps were applied to the proximal and distal left external iliac artery. After arteriotomy, the wire was pulled back into the left common iliac artery and then the sheath was taken out from the mid superficial femoral artery. The wire was advanced at this time and through and through wire access was established. Next, the proximal clamp was released and then the sheath was taken out over this wire. The proximal clamp was reapplied. The sheath was checked and it was confirmed that it was taken out in its entirety. A focal endarterectomy and primary repair of the left external iliac artery was performed. The patient made a full recovery and was discharged from the hospital.

Result: The sheath was confirmed to have been removed in its entirety through measurements and fluoroscopy. The patient has full function of his bilateral lower extremities and he will have his left lower extremity intervention using a left femoral antegrade approach.

Conclusion: Our case demonstrates successful retrieval of a damaged intravascular sheath by an open retroperitoneal approach. A history of scarred groins, tortuous iliac vasculature, acute aortic bifurcation angle, coral reef calcifications and severe atherosclerosis were anatomic factors not working in favor of the patient. Whenever possible, endovascular retrieval of damaged intravascular devices should be preferred, however, in some situations this is not possible and having adequate open aortoiliac surgical training becomes paramount in taking care of these complex vascular patients.

Rare Presentation of Labial Ulceration in a Female Patient after Hyperthermic Intraperitoneal Chemotherapy (HIPEC)

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Introduction: Cytoreductive surgery (CRS) and HIPEC is the mainstay of treatment for primary peritoneal mesothelioma, and peritoneal metastatic disease from appendiceal and colorectal cancers.[1] An estimated 55 cases per year are performed in high-volume centers, ranging from 23-123 cases annually.[2] Few case reports have demonstrated late post-operative skin complications.[3] These reports are frequently in male patients presenting with painful scrotal ulcerations.[4] One case report has even described full thickness penile necrosis in a young patient several weeks after HIPEC.[5] With the increase use of CRS and HIPEC, these complications are likely to become more frequent. To follow is a case report of a young non-Diabetic female patient that presented with painful labial ulceration with overlying skin discoloration more than two months after a repeat HIPEC.

Case Report: A 41-year-old with past medical history of stage II colon cancer presented initially in 2011 for which she underwent operative intervention. She developed a recurrence in 2013 which was managed by a total abdominal colectomy and adjuvant Folinic acid, 5-Fluorouracil, and Oxaliplatin (FOLFOX). Despite this treatment, the patient had recurrence in 2016 that progressed on FOLFOX and Avastin requiring a change to FOLFIRI with Avastin which then demonstrated stable disease. A diagnostic laparoscopy was performed on 11/2016 which showed peritoneal disease which was considered amenable for further surgical management. On 12/2016, the patient underwent CRS/HIPEC with Oxaliplatin to a Completeness of Cytoreduction (CCR) Score of 0 for a peritoneal cancer index (PCI) of 16.
On surveillance imaging, the patient was found to have an isolated recurrent implant in the splenic hilum and underwent a repeat CRS/HIPEC with Mitomycin-C to a CCR 0 for a PCI of 6 in 10/2017. On post-operative day 74, the patient presented with very painful labial ulcerations with overlying skin necrosis as seen on Figure 1. A Foley catheter was placed, and the patient was admitted to the surgical oncology service for pain control. Consultation by the Gynecology/Oncology and Plastic surgery team did not recommend any operative interventions and her skin was managed with local wound care. Patient went on to have a good recovery and healing of her wounds as demonstrated on Figure 2.

Discussion: Scrotal ulcerations is a rare complication of HIPEC that has been more commonly reported in male patients. At MD Anderson Cancer Center, there have been a total of 7 cases of inguinal skin changes with induration and scrotal sloughing in males after HIPEC. These patients presented with these skin complications up to 110 days post operatively. An example of the extent of male scrotal ulceration can be seen on Figure 3. All patients were managed non-operatively with local wound care and demonstrated good recovery with healed wounds over the next several months. This case report demonstrates the first female patient to present with a painful labial ulceration after a HIPEC. The etiology of this complication is unclear. Due to the analogous embryology of the scrotum and labia, it is possible that patients with patent process vaginalis may allow for sequestration of chemotherapy and prolonged contact resulting in skin changes and pain.
Despite the similarity of these presentations to Fournier's gangrene, most patients presenting with ulcerations secondary to HIPEC can be managed non-operatively with good outcomes. The only patient in literature review requiring an operative intervention for an excisional debridement with split thickness skin graft was the case report of full thickness penile necrosis. Further studies assessing for risk factors such as age, type of cancer, type of intraoperative chemotherapy, and complication found after initial or repeat HIPEC can help us better understand the association of these rare skin complications.

Conclusion: Scrotal ulceration is a late complication of HIPEC that can affect both men and women without Diabetes months after the procedure. We aim to raise awareness for providers that this rare complication ought to be mentioned in the consent prior to

the operation. Our goal is to also assist in recognition that most patients can be managed with local wound care with good results.

Sponsoring Institution: University of Texas MD Anderson Cancer Center

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Ruptured Silicone Breast Implant: A Rare Cause of Pleural Effusion & Trapped Lung

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Stamford Hospital

Introduction: Silicone pleural effusion is a rare complication of breast reconstruction utilizing silicone implants. We present a patient who developed a large silicone effusion with subsequent trapped lung and requiring thoracotomy and decortication.

Case Description: Our patient is a 69-year-old former 30 pack-year smoker with history of bilateral mastectomy with silicone implant reconstruction two decades previously, later diagnosed with a right upper lobe adenocarcinoma. She underwent uncomplicated thoracoscopic lobectomy. Surveillance imaging revealed a new right lower lobe nodule, requiring thoracoscopic wedge resection. At that time, a small anterior chest wall defect was noted intraoperatively, through which an intact silicone implant could be visualized. Nine months later, surveillance imaging revealed a worsening right pleural effusion with associated extracapsular rupture of her silicone implant. Contrast-enhanced MRI of the chest with silicone specific sequence demonstrated intrapleural silicone communicating with the implant capsule (Figure 1). She was taken to the operating room for washout and decortication, repair of chest wall defect, and implant removal. The silicone was extremely viscous (Figure 2) and unable to be suctioned or aspirated. It had be removed manually using a combination of techniques. The lung was entrapped within an especially thick inflammatory rind which extended homogeneously throughout the hemithorax. The lung was successfully decorticated resulting in complete reexpansion (Figure 3). The patient recovered well and was discharged on day six.

Discussion: To our knowledge, this is the first report of a patient developing a silicone effusion secondary to spontaneous implant rupture, and who developed trapped lung because of the tremendous inflammation generated by the silicone. Previously published case reports described pleural effusions treated with simple aspiration occurring after accidental or iatrogenic trauma. Silicone pleural effusion remains an unusual complication of breast implant rupture.

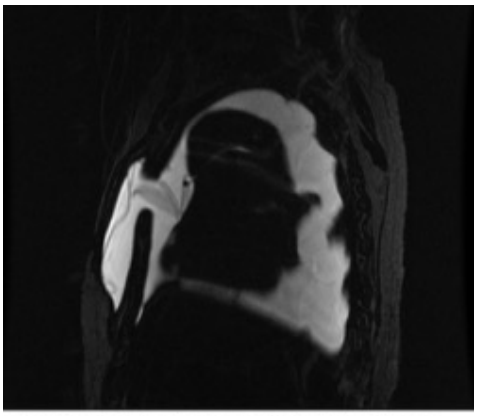

Figure 1: Contrast-enhanced MRI with silicone specific sequence, showing communication of the implant with the pleural space

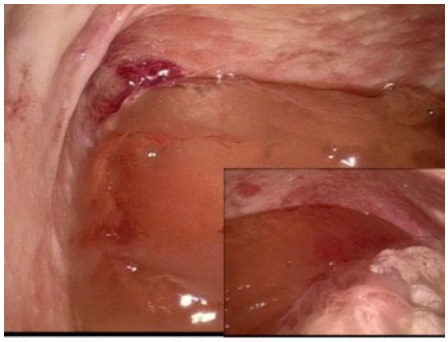

Figure 2: Thoracoscopic view of silicone pleural effusion

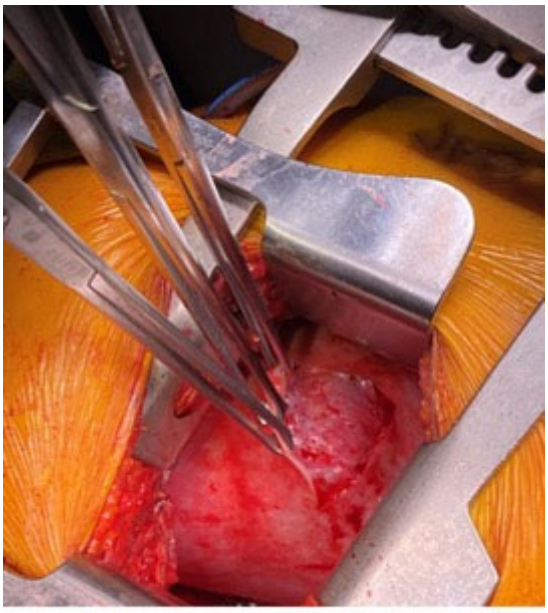

Figure 3: Decortication of trapped lung via thoracotomy