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ORIGINAL ARTICLE

Outcomes of complex abdominal wall reconstruction in patients with connective tissue disorders: a single center experience

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Abstract

Introduction Individuals diagnosed with connective tissue disorders (CTD) are known to be predisposed to incisional hernia formation. However, there is a scarcity of data on outcomes for these patients undergoing hernia repair. We sought to describe our outcomes in performing abdominal wall reconstructions in these complex patients.

Methods Adult patients with CTD undergoing open, elective, posterior component separation with permanent synthetic mesh at our institution from January 2018 to October 2022 were queried from a prospectively collected database in the Abdominal Core Health Quality Collaborative. We evaluated 30-day wound morbidity, perioperative complications, long-term hernia recurrence, and patient-reported quality of life.

Results Twelve patients were identified. Connective tissue disorders included Marfan's n = 7 (58.3%), Loeys-Dietz syndrome n = 2 (16.7%), Systemic Lupus Erythematosus n = 2 (16.7%), and Scleroderma n = 1 (8.3%). Prior incisions included three midline laparotomies and nine thoracoabdominal, mean hernia width measured 14 cm, and 9 were recurrent hernias. Surgical site occurrences (SSOs) were observed in 25% of cases, and 16.7% necessitated procedural intervention. All twelve patients were available for long-term follow-up, with a mean of 34 (12–62) months. There were no instances of reoperation or mesh excision related to the TAR procedure. One patient developed a recurrence after having his mesh violated for repair of a new visceral aneurysm. Mean HerQLes scores at 1 year were 70 and 89 at ≥ 2 years; Mean scaled PROMIS scores were 30.7 at 1 year and 36.3 at ≥ 2 years.

Conclusion Ventral hernia repair with TAR is feasible in patients with connective tissue disorder and can be a suitable alternative in patients with large complex hernias.

Keywords Connective tissue disorders · Marfan's syndrome · Ventral hernia repair · Transversus abdominis release · Surgical site occurrences · Hernia recurrence

Introduction

Qualitative or quantitative deficiencies in collagen formation have been proposed as a common factor in abdominal wall hernia formation and recurrence [8]. Twenty types of collagen molecules exist within the human extracellular matrix, with type I and III collagen comprising 95% of the total [17]. Imbalances in collagen I to III ratio are theorized to contribute to hernia development. Individuals with connective tissue disorder exhibit a reduced collagen I/III ratio and thus are at a heightened risk of developing ventral hernias [2, 10, 19].

Connective tissue disorders (CTD) constitute a heterogeneous group of systemic disorders characterized by the degradation and impairment of the extracellular matrix,

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predominantly composed of collagen and fibrillin. These disorders can be broadly classified into hereditary and acquired CTD. Hereditary CTD encompasses various inherited conditions, with Marfan's and Ehlers-Danlos syndrome being prominent examples. Acquired CTD primarily arises from autoimmune and inflammatory reactions. Among the commonly encountered acquired disorders are systemic lupus erythematosus (SLE) and scleroderma.

Individuals diagnosed with CTD are known to be predisposed to various surgical complications, including poor wound healing, dehiscence, excessive scarring, perioperative bleeding, and an increased risk of developing hernias, including recurrent ventral hernias [1, 7, 8]. Many of these patients can go on to develop complex incisional hernias with large defects often encompassing prior thoracoabdominal incisions. There is a scarcity of data evaluating the outcomes of CTD patients undergoing complex ventral hernia repair. As a result, surgeons are often reluctant to offer repair, and these patients can suffer from poor hernia related quality of life. In this paper, we describe our short and long-term outcomes of performing open, elective, ventral hernia repair (VHR) with concurrent transversus abdominis release (TAR) and permanent synthetic mesh in patients with CTD.

Methods

Following approval from the institutional review board (IRB) at the Cleveland Clinic Foundation, adult patients with the diagnosis of CTD who underwent open, elective, VHR with concurrent TAR and permanent synthetic mesh at the Cleveland Clinic Center for Abdominal Core Health (Cleveland, Ohio) between January 2018 to October 2022 were identified from the Abdominal Core Health Quality Collaborative (ACHQC). The ACHQC is a hernia-specific nationwide registry aimed at improving the quality of hernia care through patient-centered data collection, performance feedback to clinicians, and collaborative learning. Surgeons enter patient data prospectively in real-time during routine clinical care, including patient demographics, hernia characteristics, operative details, patient-reported outcomes (PROs), and postoperative follow-up information [7]. Data for patients undergoing the aforementioned repair were exported and abstracted to confirm the previous diagnosis of CTD.

We evaluated 30-day wound morbidity, consisting of surgical site infections (SSI), surgical site occurrences (SSO), and surgical site occurrences requiring procedural intervention (SSOPI) [9]. SSIs were further categorized based on the CDC definition into superficial, deep, or organ space infections [15]. SSOs incorporated SSIs and other conditions such as wound cellulitis, non-healing incisional wounds, fascial disruption, skin or soft tissue ischemia, necrosis, serous or purulent wound drainage, stitch abscess, seroma, hematoma, infected or exposed mesh, or the development of an enterocutaneous fistula [9]. SSOPI incorporated any SSO that requires opening of the wound, wound debridement, excision of sutures, percutaneous drainage, or partial or complete mesh removal [9]. Additionally, 30-day all morbidity including ileus, bowel obstruction, pulmonary embolism (PE), stroke, deep venous thromboembolism (DVT), myocardial infarction (MI), cardiac arrest, sepsis, septic shock, pneumonia, urinary tract infection (UTI), acute kidney injury (AKI), renal failure, re-intubation, and other complications were also reported. Additionally, other outcomes evaluated included reoperation, mesh excision, pragmatic hernia recurrence and patient-reported quality of life at a minimum of 12 months (±4 weeks) after hernia repair or for longest available follow up.

Pragmatic hernia recurrence was determined based on prior work published by Krpata et al. [12]. In brief, clinical follow-up comprising surgeon evaluation and/or CT scans was employed to determine recurrence. If clinical and imaging evaluation did not occur, recurrence evaluation was carried out utilizing the Hernia Recurrence Inventory (HRI) [4]. In cases where a bulge was reported on HRI, the imaging could overrule that finding. If no other evaluation occurred, a bulge was considered a recurrence.

Long term quality of life was assessed utilizing the hernia-related quality of life score (HerQLes) and National Institutes of Health Patient-Reported Outcome Measurement Information System (PROMIS) Pain Intensity 3A T-Score. HerQLes is a validated patient-reported outcomes measure consisting of 12 questions focused on the impact of an individual's abdominal wall on their quality of life [13]. Modified HerQLes scores are calculated on a scale of 0 to 100, wherein higher HerQLes summary scores signify an enhanced quality of life. The minimum clinical important difference (MCID) of the HerQLes score has been determined at 15.6 [18]. The PROMIS Pain Intensity Short Form 3A is a validated assessment tool encompassing three questions about patients' pain over the preceding 7 days. It is rated between 30.7 and 71.8, with a value of 30.7 signifying an absence of pain, while higher scores correspond to escalated pain levels [6]. MCID of PROMIS score has not been determined yet for ventral hernias. Change from baseline scores for these measures was calculated at 30 days and at a minimum of 12-month (±4 weeks).

Clinical follow-up included surgeon evaluations at 30 days and 12 months, accompanied by CT scans at the 12-month follow up point. Furthermore, we tracked all patients whose follow-up period exceeded 24 months (±2 months) to gather long-term follow-up data through direct phone calls, chart abstraction, and imaging review. Descriptive statistics were used to characterize the study population.

Results

A total of 12 patients with the diagnosis of CTD underwent open, elective VHR with concurrent TAR and permanent synthetic mesh between January 2018 to October 2022. Demographics are detailed in Table 1. The mean age was 52 years (± SD 14.2 years), 8 (66.7%) were female and mean BMI was 27 kg/m^2 (± SD 4 kg/m^2). Nine patients (75%) had hereditary CTD, with Marfan's syndrome being the most prominent comorbidity in 7 patients (58.3%), while 2 patients (16.7%) presented with Loeys-Dietz syndrome. Three patients (25%) had acquired CTD, consisting of 2 patients with SLE (16.7%) and one with scleroderma (8.3%). Hypertension and Diabetes Mellitus were the most common comorbidity affecting 11 patients (84.6%). Six patients (50%) were on immunosuppression therapy, 12 (100%) were on antiplatelet medications, and 5 (41.7%) were on combined antiplatelet medications and anticoagulant medications at the time of surgery. Most patients had an ASA class 3 (11, 91.7%).

Hernia and operative characteristics are demonstrated in Table 2. Among the entire cohort, all cases involved incisional hernias, of which 9 patients (75%) exhibited recurrent hernias. Nine patients (75%) had previously undergone surgery with thoracoabdominal incisions, while 3 (25%) had a history of midline laparotomies. Seven patients (58.3%) had a midline hernia, one (8.3%) had a flank hernia, four (33.3%) had a combined midline and flank hernia, and 1 (8.3%) had concurrent parastomal hernia. The predominant symptoms reported by most patients included pain (11, 91.7%) and/or hernia enlargement (10 patients, 83.3%), with 2 patients (16.7%) having experienced episodes of bowel obstruction. The mean hernia width was 14 cm (± 5 cm) and length was 20 cm (± 4 cm). Eleven cases (91.7%) were classified as clean (CDC wound class I), while 1 (8.3%) was classified as contaminated (CDC wound class III). This patient underwent simultaneous parastomal hernia repair, had a stoma repositioned, and had retromuscular Sugarbaker repair. Mean operative time was 183 min (± 48). The mean length and width of the meshes were 36.7 cm (range 30–50 cm). Complete fascial closure was achieved in all patients, and no skin flaps were raised. Retromuscular drains were used

Table 1 Demographics and comorbidities

Demographics and comorbidities	n = 12
Age, y (mean ± SD)	52 (± 14.2)
Female	8 (66.7%)
BMI (mean ± SD)	27 (± 4.0)
Active smoking	2 (16.7%)
Type of collagen vascular disorder	
Marfan's syndrome	7 (58.3%)
Loeys-Dietz syndrome	2 (16.7%)
Systemic Lupus Erythematosus	2 (16.7%)
Scleroderma	1 (8.3%)
Diabetes Mellitus	11 (84.6%)
Hypertension	11 (84.6%)
COPD	1 (7.7%)
Dialysis	1 (7.7%)
History of Abdominal Aortic Aneurysm	9 (75%)
IBD	1 (8.3%)
Immunosuppressants	4 (33.3%)
Anti-platelet medications	12 (100%)
Anti-coagulant medications	5 (41.7%)
ASA Class	
3	11 (91.7%)
4	1 (8.3%)

Table 2 Hernia and operative characteristics

Hernia and operative characteristics	n = 12
Incisional hernia	12 (100%)
Previous incision	
Thoracoabdominal	9 (75%)
Midline laparotomy	3 (25%)
Hernia location	
Midline hernia	7 (58.3%%)
Midline + Flank	4 (33.3%)
Flank hernia	1 (8.3%)
Concurrent parastomal hernia	1 (8.3%)
Recurrent hernia	9 (75%)
Hernia width, cm (mean ± SD)	14 (± 5)
Hernia length, cm (mean ± SD)	20 (± 4)
VHWG grade	
1	1 (8.3%)
2	9 (75%)
3	2 (16.7%)
4	0
Surgical indication	
Pain	11 (91.7%)
Hernia enlargement	10 (83.3%)
History of bowel obstruction	2 (16.7%)
Wound status	
Clean	10 (83.3%)
Clean-contaminated	0
Contaminated	1 (8.3%)
Fascial Closure	12 (100%)
Skin flaps raised	0
Mesh length, cm (mean, range)	36.7 (30–50)
Mesh Width, cm (mean, range)	36.7 (30–50)
Concomitant procedure	0
Operative time minutes (mean ± SD)	183 (± 48)
Intraoperative complication	0

in all cases. There were no concomitant procedures and no intraoperative complications.

Table 3 outlines the 30-day outcomes. Within this period, two patients (16.7%) experienced SSO. One (8.3%) experienced superficial SSI managed by wound opening and IV antibiotics, while another (8.3%) had cellulitis treated with oral antibiotics. Both of these patients were on immunosuppression therapy. The 30-day SSOPI rate was 8.3%, with one patient necessitating wound opening, as mentioned. Postoperative ileus was the most prevalent outcome, affecting 4 patients (33.3%), followed by urinary tract infection (UTI) in 1 patient (8.3%). No other postoperative complications were recorded. The mean length of stay was 4 days (±2.1 days), and no readmissions were required. The patient who experienced a superficial SSI was a 61-year-old female diagnosed with SLE, treated with immunosuppressants, and had previously undergone six ventral hernia repairs. During the TAR procedure, all old meshes were excised with the exception of an onlay mesh in the right lower quadrant at an old ostomy site. Along with superficial SSI, her surgery was complicated by paralytic ileus and UTI. She was successfully managed through wound opening and intravenous antibiotics, exhibiting a satisfactory recovery. The 12-month follow-up was uneventful, with a normal CT scan showing no abnormalities. However, after 22 months, localized contamination of the old onlay mesh was observed with a subcutaneous abscess formation and draining sinus without the involvement of the retromuscular mesh on imaging. This patient is awaiting surgery for local removal of onlay mesh.

Table 4 provides details of the long-term follow-up. All patients had 12-month (±4 weeks) assessments, with nine patients (75%) having follow-up periods extending beyond 24 months (±2 months). Among these, three patients were evaluated through HRI and CT scans, while the remaining six relied solely on the HRI. The mean follow-up duration was 34 months, ranging from 12 to 62 months. Throughout that period, three patients required reoperation. One experienced mesh contamination of an old onlay mesh implanted years before the TAR procedure, as previously noted. Two patients with Marfan's syndrome required subsequent laparotomies for reasons not related to the AWR surgery. The first patient underwent an emergent splenectomy 2 months after AWR surgery due to spontaneous intrasplenic aneurysm rupture and ongoing bleeding. A year later, a CT scan

Table 3 30 Days clinical outcomes

Hernia and operative characteristics	30 days n = 12
SSO	2 (16.7%)
Superficial SSI	1 (8.3%)
Deep SSI	0
Organ space SSI	0
Wound cellulitis	1 (8.3%)
Infected mesh	0
Seroma	0
Hematoma	0
Non-healing incisional wound	0
Fascial disruption	0
Skin or soft tissue necrosis	0
Exposed mesh	0
Enterocutaneous fistula	0
SSOPI	1 (8.3%)
Wound opening	1 (8.3%)
Percutaneous drainage	0
Mesh removal	0
Reoperation	0
Ventilator > 48 h	0
Re-intubation	0
Post-op bleeding transfusion	0
Pneumonia	0
UTI	1 (8.3%)
Sepsis	0
Ileus	4 (33.3%)
DVT	0
Pulmonary embolism	0
Stroke	0
Myocardial infarction	0
Acute renal failure	0
Length of stay, d (mean ± SD)	4 (2.1)
Readmission	0
Hernia recurrence	0

Table 4 Long term clinical outcomes

Hernia and operative characteristics	12 months n = 12	> 24 months n = 9
SSO	0	1 (8.3%)[d]
Superficial SSI	0	0
Deep SSI	0	0
Organ space SSI	0	0
Infected mesh	0	1 (8.3%)[a]
Fascial disruption	0	0
Enterocutaneous fistula	0	0
Exposed mesh	0	0
SSOPI	0	1 (8.3%)
Percutaneous drainage	0	0
Mesh removal	0	1 (8.3%)
Reoperation	1 (8.3%)[b]	1 (8.3%)[b]
Readmission	0	0
Hernia recurrence	0	1 (8.3%)[b]

[a]Previously implanted mesh from prior hernia surgery

[b]Reoperation for reasons unrelated to hernia recurrence

[d]PDS - PolyDioxanone Suture

confirmed no recurrence. The second patient underwent elective surgery three years after AWR surgery for a mycotic SMA aneurysm. A year later, a small asymptomatic recurrence was evident on a CT scan, and reoperation has not been performed.

Quality of life assessment is detailed in Table 5. Mean HerQLes scores at baseline were 43 (n = 12), 70 at 12 months (n = 12), and 89 at ≥ 24 months (n = 9). Median change in HerQLes from baseline to 12 months is + 21.5, and from baseline, to > 24 months is + 54. Mean scaled PROMIS scores were 40.2 at baseline (n = 12), 30.7 at 12 months (n = 12), and 36.3 at ≥ 24 months(n = 9). Median change in scaled PROMIS scores from baseline to 12 months is -11.6, and from baseline to > 24 months is -9.5.

Discussion

This is the first case series describing outcomes of VHR with TAR among patients with connective tissue disorder. Our findings highlight the complexity of this patient population with three quarters of the patients having prior thoracoabdominal incisions and recurrent incisional hernias. Despite these complexities, CTD patients undergoing open posterior component separation procedures had remarkably similar outcomes to our other complex abdominal wall patients. However, underlying vascular issues should be thoroughly evaluated preoperatively, particular in Marfan's patients, as postoperative complications related to aneurysmal disease can necessitate reoperation and division of the prior mesh. Finally, these patients did benefit with significant improvement in long term quality of life and reduction of pain. Given these findings, we do believe that it is safe and feasible to offer patients with CTD complex abdominal wall reconstruction in high volume hernia centers.

Current literature regarding VHR in patients with CTD is limited, comprising primarily case reports and a handful of case series. While knowledge about surgical outcomes in hernia repair for patients with CTD is scarce, studies focusing on GI and other abdominal surgeries have indicated increased perioperative bleeding and delayed wound healing. This has prompted many surgeons to restrict surgical intervention to life-threatening events [5, 20]. However, our study, although derived from a modest sample size, challenges this notion. A recent publication from our group focused on long-term outcomes following TAR in the general population revealed SSOPI, reoperation, and composite hernia recurrence rates of 8.4%, 4.7%, and 10% (respectively) at one year [21]. Although lacking statistical power to determine significance, our study aligns with these findings (16.7% SSOPI, with no instances of reoperation or hernia recurrence attributed to the hernia repair during the entire follow-up period). This is supported by a case series by Kroese et al., which tracked 14 patients with Ehlers-Danlos undergoing open VHR. Within this series, 10 patients (71%) exhibited incisional hernias, with 9 undergoing open retromuscular repair. Over a mean follow-up of 50 months, 3 patients (21%) experienced postoperative wound complications (two SSIs and one seroma). No other complications were recorded, and the recurrence rate was 7.1% (one patient) [11].

Within the context of this study, pinpointing the precise factors behind the complications identified remains challenging. In addition to being evident in the general population [21], these complications are further confounded by variables like CTD, age, hernia characteristics, anticoagulation therapy, and immunosuppression. We also believe that one of the factors leading to our relatively low hernia recurrence rate in this collagen deficient population is the wide overlap achieved with our retromuscular approach utilizing synthetic mesh. While the use of other biologic or biosynthetic meshes have been evaluated extensively in prior literature, we do believe that in patients with inherent collagen disorders, these prosthetics are not appropriate. If permanent synthetic mesh was not an option due to contamination, or other findings, we would not perform this operation in this patient population.

Another interesting finding in our series is the long term reoperation rate for vascular issues not related to the hernia repair. We believe that surgeons should cautiously consider performing such a definitive hernia repair as described here with complete dissection of the retromuscular and preperitoneal planes. As seen in our series, these hernias were large and complicated with a mean defect size of 14 cm and the majority involving a prior hernia repair and or thoracoabdominal incisions. Given that individuals with CTD are particularly predisposed to requiring abdominal surgeries due to their inherent vasculopathy [1, 3, 14] it is wise to avoid disrupting the retromuscular planes unless required. We have previously presented our rate of re-laparotomy post TAR procedure in 1337 patients who underwent TAR procedure at our institution from January 2014 to September

Table 5 Hernia recurrence and life quality assessment

Hernia recurrence and life quality assessment	Baseline n = 12	30 days n = 12	12 months n = 12	> 24 months n = 9
HerQLes Score (mean)	43	43	70	89
PROMIS Pain 3A T-Score (mean)	40.2	52.1	30.7	36.3

2022. We noted a 4% rate of re-laparotomy unrelated to hernia recurrence or repair complications. The consequences of hernia recurrence after TAR procedure are significant. A recent study by our group reported redo-TAR outcomes, highlighted the technical challenges and potential morbidity associated with redo-TAR surgeries [16].

Our study bears limitations. The sample size is small, comprising 9 patients with inherent CTD and 3 with acquired CTD. Despite being one of the most comprehensive series to date, the modest numbers preclude definitive conclusions. Hence, other surgeons should exercise caution when applying our findings to their patients. Another limitation concerns the duration of clinical follow-up, which is not extensive. Our standard clinical follow-up involves surgeon assessments at 30 days, 12 months, and 5 years, complemented by CT scans at 12 months and 5 years. The study population includes patients from 2018 onward, as CTD documentation was not explicitly available in the ACHQC database before then. Given the potential long-term evolution of recurrence rates and mesh complications, we continue routine follow-up with registry surveillance and as this is an uncurable disease, we expect further findings. Ultimately, our results must be viewed in the context of these procedures being conducted within a high-volume hernia center, where TAR stands as the predominant reconstructive operation. Despite a high level of expertise with complex retromuscular repairs, operating on this specific population of patients suffering from complex hernias that may expand to the chest and involving prior thoracoabdominal incisions is one of the more challenging operations performed by our group. We recommend that patients with CTD be managed at specialized hernia centers by individuals with an extremely high volume of complex abdominal wall reconstruction.

Conclusion

Ventral hernia repair with TAR is feasible in patients with connective tissue disorder and yields acceptable complication and recurrence rates. However, we believe this procedure is best suited for large complex hernias accompanied by moderate to severe symptoms and should be conducted at specialized hernia centers with high volume of complex reconstructions.

Supplementary Information The online version contains supplementary material available at https://doi.org/10.1007/s10029-023-02957-y.

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Declarations

Conflict of interest I declare that there are no conflicts of interest related to this manuscript. The study was conducted impartially, and the findings presented in the manuscript are based solely on the analysis and interpretation of the data.

Ethical approval The research was conducted in accordance with the ethical principles outlined in Institutional Review Board. The use of data and materials was carried out with proper authorization and in compliance with all applicable laws and regulations.

Informed consent As this study involved the use of existing data and did not involve direct interaction with human subjects, no patients were enrolled, and therefore, no informed consent was required. The data used in this research were anonymized and de-identified, ensuring the privacy and confidentiality of individuals in accordance with the Institutional Review Board.

Human and Animal Rights In adherence to ethical standards, this chart review study, though devoid of directhuman or animal involvement, maintained patient data confidentiality and ethical standards as required by theinstitutional review board.

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