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The cost of failure: The economic impact of failed primary closure in classic bladder exstrophy

Christopher A. Hesh [a,*], Ezekiel Young [a], Paul Intihar [b], John P. Gearhart [a]

[a] The Johns Hopkins School of Medicine, Department of Urology, Division of Pediatric Urology, Baltimore, MD, United States
[b] The Johns Hopkins Medical Institutions, Financial Analysis Unit, Baltimore, MD, United States

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ABSTRACT

Purpose: Failure of primary closure in classic bladder exstrophy (CBE) is a significant cause of morbidity, and yet its relative economic impact has not been well characterized. The authors aim to determine whether CBE patients who underwent failed primary closure incur greater economic burden in the year following their successful closure than those patients who underwent a successful primary closure.
Materials and Methods: After institutional review board approval CBE patients who were successfully closed between 1993 and 2013 were identified in an institutional exstrophy–epispadias database. Patients who were never closed at the study institution and those who had no documented successful closure were excluded. Inpatient hospital charges, hospital costs, and professional fees were collected for the year following successful closure.
Results: 162 patients met the inclusion and exclusion criteria and accounted for 312 inpatient admissions in the year following and including their respective successful bladder closures. 62 of the patients failed their primary closure and the remaining 100 succeeded. Adjusting for covariates, patients who underwent successful primary closure experienced a reduction in inpatient hospital charges of $8497, hospital costs of $9046 and professional fees of $11,180 in the year following their successful closure compared to those patients who failed their primary closure.
Conclusion: Apart from the self-evident financial advantages of a successful primary closure, namely the avoidance of reclosure, there appears to be a lasting negative financial impact of failed primary closure even after these patients undergo successful reclosure at the study institution.

1. Background and significance

Classic bladder exstrophy (CBE) is a rare congenital malformation with an incidence between 1 in 10,000 and 1 in 50,000. It is characterized by deformation of the bladder, lower abdominal wall, pelvis and genitalia with a splayed vesicourethral template and a defect within the anterior lower abdominal wall. The genitalia in males is characterized by epispadias, with a short, broad, dorsally curved, open urethral plate and shortened and proximally divergent corpora. In females the vagina is typically short and anteriorly displaced, often with a stenotic os, a bifid clitoris and divergent labia and mons [1]. Not only do these patients require special attention by pediatric urologists and general pediatric surgeons, but they also frequently require pelvic osteotomy at the hands of skilled pediatric orthopedic surgeons to encourage closure success. Because CBE requires complex intervention by such a large interdisciplinary team of surgical and non-surgical specialists it results in an economic burden totaling at least $10 million each year in hospital charges in the United States alone [2].

Costs incurred during the first year following primary bladder closure have been estimated, as have the relative cost advantages of neonatal primary closure as compared to both delayed primary closure and reclosure [3]. In this study the authors use an institutional exstrophy–epispadias database to identify CBE patients and explore the relative impact of failed primary closure on the inpatient total inflation-adjusted hospital charges (TIAHC), total inflation-adjusted hospital costs (TIAC) and total inflation-adjusted professional fees (TIAPF) in the first year following, and including, the patient's ultimate successful reclosure. The authors hypothesize that when comparing the ultimate successful reclosures of those patients who experienced a failed PC to patients who had a successful PC the TIAHC, TIAC and TIAPF will be significantly larger in those patients who failed their PC.

2. Methods

2.1. Study design

A retrospective cohort design was employed using the author's institutional exstrophy–epispadias database and this was approved by the study institution's review board (IRB00051221). Patients were separated into two groups based on whether their PC was successful, and were subsequently followed for one year to determine accumulated inpatient

Abbreviations: CBE, classic bladder exstrophy; PC, primary closure.
* Corresponding author at: 1800 Orleans Street, Suite 7302, Baltimore, MD 21287, United States.
E-mail address: chesh1@jhmi.edu (C.A. Hesh).

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TIAHC, TIAC and TIAPF in the year following their primary successful or ultimate successful closure (Fig. 1).

2.2. Data sources and setting

Three sources of data were used. Clinical data were obtained from an institutional database of exstrophy–epispadias patients treated at the study institution to date. This database includes, but is not limited to, date of all closures, age of patient at each closure, location of all closures at study vs nonstudy institution, use of pelvic osteotomy during closure, and success or failure of all closures. Hospital charges and costs were retrieved from the study institution's administrative CaseMix DataMart database which houses inpatient hospital financial data. These data were available on a per-admission basis, and included the following financial variables: hospital charges, direct costs, and total costs. In addition, length of stay was indicated for each admission. Charge and cost data were available from 1993 and 1998, respectively. Finally, professional fees were extracted from the study institution's physician billing system. Like the hospital's inpatient database, professional fees were reported on a per admission basis and were matched to each inpatient admission. Professional fee data were available from 2007.

The study institution is a large, tertiary referral center that has treated over 1280 patients with the exstrophy–epispadias complex. CBE patients at the study institution are closed primarily using the modern staged repair of exstrophy (MSRE) and all closures during the study period were overseen by a single surgeon. Patients referred to the institution for reclosure, however, include patients closed using the MSRE, complete primary repair of exstrophy (CPRE), Kelly closure, as well as other less common closure techniques.

2.3. Subjects

Patients were included in the study based on a diagnosis of classic bladder exstrophy (CBE) as determined by the primary surgeon at the time of first admission, or by nonstudy institutional medical records available to the primary surgeon at that time. Patients were excluded if they were closed before July 1st, 1993 because of available institutional financial data, and after July 1st, 2013 because of lack of adequate follow-up time. Similarly, patients who never underwent a bladder closure at the study institution were excluded because of lack of available financial data.

2.4. Definition of variables

2.4.1. Patient-level variables

Patient level variables include all those variables heretofore described as present in the institutional database. Bladder closures were deemed to have failed if, within a year after closure, the patient suffered bladder dehiscence, bladder prolapse, vesicocutaneous fistula or outlet obstruction. In addition to the variables available in the database a time variable was calculated to assess outpacing of inflation by taking the difference in days between a specific patient's date of successful closure and the beginning of the study period.

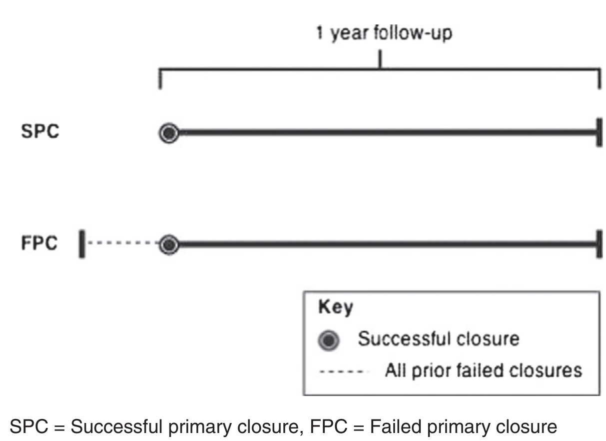

Fig. 1. Study design. SPC = successful primary closure, FPC = failed primary closure.

2.4.2. Hospital-level variables

Hospital charges, what the hospital expects to be paid for the services rendered, were provided on a per-admission basis. Per-patient total hospital charges for the year following successful closure were determined by summing individual per-admission charges. Total costs are what the hospital has paid to employees and vendors to be able to provide services during each admission. Direct costs, a portion of total costs, represent those costs directly involved in providing patient care (e.g., nursing salaries, pharmaceuticals, contrast fluids for CT scans, etc…) while indirect costs, the remainder of total costs, represent those which are not related to specific patient care but which are needed to support the direct care departments (e.g., security, utilities, housekeeping). Per-patient total and direct costs were determined by summing individual per-admission total and direct costs, respectively. Professional fees are billed according to services rendered or the procedure preformed and are reported on a per-admission basis, and, like the hospital costs and charges, were summed to arrive at total fees incurred. All financial variables were inflation-adjusted to 2014 dollars using the medical care consumer price index.

2.5. Statistical analysis

Descriptive statistics for both patient and hospital-level covariates were compared for patients with and without successful primary closure. Hospital-level covariates were collected for the year following ultimate successful reclosure. In the case of those patients with a successful primary closure this would represent the year following their primary closure. In the case of those with failed primary closure this would represent the year following their second, or in some few cases third or fourth, closure, whichever closure was successful. Where appropriate, Welch's two sample *t* test and Person's chi-squared test with Yates' continuity correction were utilized to compare groups.

Separate multivariable linear regression models were used to analyze the relationship between total inflation-adjusted hospital charges, total inflation-adjusted costs, and total inflation-adjusted professional fees as separate outcomes while controlling for clinically relevant covariates incorporated based on Akaike information criterion (patient-level variables). Each outcome variable was analyzed first with an unadjusted single linear regression on outcome of primary closure and, separately, on utilization of pelvic osteotomy at successful closure to assess confounding in the multivariable model. The model was then adjusted by adding patient-level covariates. Additionally, TIACs were specifically assessed by use of single and multivariable regression to identify patterns in the proportion of TIAC constituted by direct costs.

An alpha of 0.05 was utilized for null hypothesis significance testing. Statistical analysis was performed using R 3.1.2.

3. Results

349 CBE patients closed between 07/01/1993 and 07/01/2013 were identified in the authors' institutional exstrophy–epispadias database. Two patients lacked documentation of a successful closure and 185 patients were successfully closed at an outside hospital; both groups were excluded from the study. The remaining 162 patients accounted for 312 inpatient admissions in the year following their respective successful bladder closures. Selected descriptive statistics can be found in Table 1. 62 of the patients failed their primary closure while the remaining 100 underwent successful primary closure. A majority of the patients were male (n = 110), and while a smaller portion of males than females failed their primary closure this difference was insignificant

($\chi^2 = 1.55$, $p = 0.21$). By design, all patients who received a successful primary closure were closed at the study institution. Two of the patients who failed initial closure were primary closed at the study institution and the remainder, 60 patients, were primarily closed at a nonstudy institution ($\chi^2 = 149.58$, $p < 0.001$). Of those patients who failed their primary closure the median age in days at successful closure was 418 as compared to 1 for those patients who experienced a successful primary closure. A larger portion of the patients who failed their primary closure required a pelvic osteotomy at successful closure compared to those who had a successful primary closure ($\chi^2 = 12.97$, $p < 0.001$). In addition, not noted in the table, patients who received an osteotomy at successful closure had a total length of stay of 49.81 days in the year following successful closure compared to 39.44 days in those patients who did not have an osteotomy ($p = 3.63e{-}05$). Following successful closure those who had failed their primary closure experienced no difference in number of inpatient admissions, though they did have a significantly longer mean length of stay than those who experienced a successful primary closure (49.81 vs 43.45, $p = 0.01$, 95% CI [1.32, 11.39]).

Table 2 contains the single linear regressions of each economic outcome on success vs failure of primary closure. A statistically significant reduction in both total inflation-adjusted hospital charges (TIAHC), by \$18,831, and total inflation-adjusted professional fees (TIAPF), by \$13,791, but not total inflation-adjusted costs (TIAC) is observed. Fig. 2 displays the relationship of TIAHC to each primary closure group graphically. In addition, Table 2 contains data on the relationship of each economic outcome on the presence or absence of pelvic osteotomy at the time of successful closure, and demonstrates a statistically significant increase in both TIAHC and TIAC by \$17,691 and \$25,701, respectively, with no statistically significant difference in TIAPF.

Of the 162 patients in the study sample, all had available hospital charge data. As demonstrated in Table 3, the baseline TIAHC for a patient who failed their primary closure and did not have an osteotomy at their ultimate successful reclosure, with the mean total inpatient LOS, total number of inpatient admissions, and time since start of the study period, is \$129,1000 (95% CI \$120,144, \$138,122) for the year following successful closure. These charges are statistically significantly reduced by \$8497 (95% CI −\$16,493, −\$500) in those patients with a successful primary closure compared to those who failed their primary closure. Of note, use of a pelvic osteotomy at the time of successful closure does not statistically significantly change the TIAHC (Beta = −\$5302, 95% CI [−\$14,003, \$3400]), and addition of an interaction term including osteotomy at successful closure and total length of stay did not impact this conclusion and was itself statistically insignificant. In addition, each additional inpatient day increases TIAHC by \$2552 (95% CI \$2226, \$2878), each inpatient admission increases TIAHC by \$10,400 (95% CI \$6515, \$14,291), and passage of each day since start of the study period – a surrogate for outpacing of inflation – increases TIAHC by \$5.93 (95% CI \$4.14, \$7.72).

Of the 162 patients in the study sample 131 had available hospital cost data. As demonstrated in Table 3, the baseline TIAC for a patient who failed their primary closure and did not have an osteotomy at their ultimate successful reclosure, with the mean total inpatient LOS, total number of inpatient admissions, and time since start of the study period, is \$84,769 (95% CI \$77,623, \$91,915) for the year following successful closure. These costs are statistically significantly reduced by \$9046 (95% CI −\$15,558, −\$2533) in those patients with a successful primary closure compared to those who failed their primary closure. Just as in TIAHC, TIACs are not altered by use of an osteotomy at successful closure (Beta = \$3023, 95% CI [−\$4060, \$10,106]) and this conclusion is not confounded by addition of an interaction term including osteotomy at successful closure and total length of stay and was itself statistically insignificant. In addition, each additional inpatient day increases TIAC by \$2147 (95% CI \$1825, \$2469), each inpatient admission increases TIAC by \$5946 (95% CI \$2927, \$8965), and time since the start of the study period increases TIAC by \$17.08 (95% CI \$15.33, \$18.83). In addition, a single linear regression of the proportion of TIACs that are direct costs on TIAC as an outcome found a small, but statistically significant decrease in the proportion of TIAC made up by direct costs as TIAC increases (Beta = −1.97e−07, 95% CI −3.16e−07, −7.82e−08, $p = 0.001$).

Of the 162 patients in the study sample 56 had available professional fee data. As demonstrated in Table 3, the baseline TIAPF for a patient who failed their primary closure and did not have an osteotomy at their ultimate successful reclosure, with the mean total inpatient LOS, total number of inpatient admissions, and time since start of the study period, is \$60,760 (95% CI \$51,199, \$70,313) for the year following successful closure. These fees are statistically significantly reduced by \$11,180 (95% CI −\$17,851, −\$4514) in those patients with a successful primary closure compared to those who failed their primary closure. Unlike TIAHC and TIAC, use of a pelvic osteotomy at successful closure led to a statistically significant reduction in overall TIAPF by \$7320 (95% CI −\$14,116, −\$524). This relationship, however, ceased to exist when the same model was fit with an additional interaction term between osteotomy at successful closure and total length of stay (Beta = −\$5737, $p = 0.16$; 95% CI −\$13,720.54, \$2247.00). Each

Table 2
Single linear regressions of financial variables on successful primary closure and osteotomy at successful closure.

Single linear regressions	Beta [95% CI]
Economic impact of successful PC	
Total inflation-adjusted hospital charges	−18,831 [−35,383, −2279], $p = 0.026$
Total inflation-adjusted costs	−9754 [−27,473, 7964], $p = 0.278$
Total inflation-adjusted professional fees	−13,791 [−24,210, −3371], $p = 0.010$
Economic impact of osteotomy at successful PC	
Total inflation-adjusted hospital charges	17,691 [658, 34,723], $p = 0.042$
Total inflation-adjusted costs	25,701 [8253,43,149], $p = 0.004$
Total inflation-adjusted professional fees	−1051 [−12,718,10,615], $p = 0.857$

Results reported as Beta [95% CI] with p values. Each economic outcome is regressed on one value, either success versus failure of PC or presence versus absence of osteotomy at successful closure.

Table 1
Demographics and clinical outcomes of patients by success versus failure of primary closure.

	Failed primary closure	Successful primary closure	
Gender			
Male	38	72	$\chi^2 = 1.55$, $p = 0.21$
Female	24	28	
Location of primary closure			
Study institution	2	100	$\chi^2 = 149.58$, $p < 0.001$
Nonstudy institution	60	0	
Median days old at successful closure	418	1	
Osteotomy at successful closure			
Yes	52	45	$\chi^2 = 12.97$, $p < 0.001$
No	10	55	
Mean inpatient days in year following successful closure	49.81	43.45	$p = 0.01$ [1.32, 11.39]
Mean number of admission in year following successful closure	1.81	2.00	$p = 0.287$ [−0.55, 0.16]

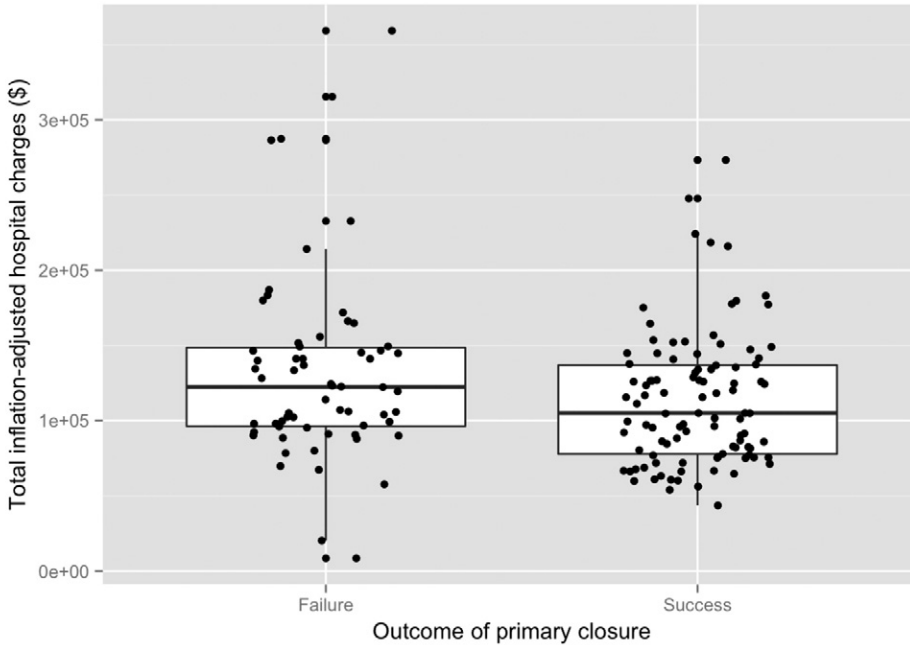

Fig. 2. Total inflation-adjusted hospital charges as they relate to outcome of primary closure. The points overlapping the boxplots represent individual patients and their respective total inflation-adjusted hospital charges for the year following successful closure.

additional inpatient day increases TIAPF by $989 (95% CI $658, $1320), each inpatient admission increases TIAPF by $4544 (95% CI $1464, $7625), and, unlike TIAHC and TIAC, time since the start of the study period had no impact on TIAPF (Beta = $.21, 95% CI [−$3.67, $4.09]).

4. Discussion and conclusion

The clinical impact of failed PC is well documented in the literature. Not only do these patients have significantly lower rates of urethral continence, but also they maintain smaller bladder capacities and exhibit slower bladder growth than their successful PC counterparts which reduces their ability to achieve a bladder capacity sufficient for bladder neck reconstruction [4]. All these after undergoing a failed PC that alone carries an estimated hospital charge ranging anywhere from $53,291 to $75,742 [6]. The self-evident advantages of successful PC are reducing these poor clinical outcomes and in addition avoiding a costly reclosure. And while the cost of primary closure and the subsequent year of follow-up has been previously estimated, the relative economic impact of failed PC had yet to be quantified [3].

Herein, the authors have demonstrated that after controlling for length of inpatient stay, number of inpatient admissions, days since the start of the study period and use of pelvic osteotomy, simply having failed a PC confers an increase in inpatient hospital charges by $8497, costs by $9046, and professional fees by $11180 on the patient's ultimate successful reclosure and subsequent year of follow-up, or in other words an increase in total charges of $19,677 — the sum of hospital charges and professional fees. While the source of this relative impact is not specified, the authors suspect both operative time and complications associated with reclosing a patient that could have been primarily closed using one of any number of approaches. In addition, surgery and perioperative care may have been more cautious in reclosures. Indeed, these aspects of care delivery are important stepping off points for future studies into the economic impact of exstrophy care. Regardless, the existence of these increased charges, costs and fees at a high-volume exstrophy center begs the question of how this relative impact varies across practice settings, particularly in light of past findings that high-volume exstrophy centers have lower hospital charges than their mid-volume and low-volume counterparts [2].

Of additional importance, while pelvic osteotomy at successful closure increased TIAHC by $17,691 and TIAC by $25,701 when assessed as a sole predictor, after controlling for the covariates its financial impact was negligible. This is important, particularly in reclosures, in that these patients fare better in their opportunity for bladder neck reconstruction or augmentation cystoplasty having had a pelvic osteotomy with immobilization at the time of reclosure [5]. That our data demonstrate the use of such a procedure does not negatively impact the overall cost of treatment in the year following and including reclosure is not insignificant.

The limitations of this study are first and foremost the unique referral pattern and patient population of a single, large-volume exstrophy

Table 3
Multivariable linear regression of financial variables.

Multivariable linear regression	TIAHC	TIAC	TIAPF
Intercept	129,100 [120,144, 138,122], $p < 2e{-}16$	84,769 [77,623, 91,915], $p < 2e{-}16$	60,760 [51,199, 70,313], $p < 2e{-}16$
Total inpatient LOS	2552 [2226, 2878], $p < 2e{-}16$	2147 [1825, 2469], $p < 2e{-}16$	989 [658, 1320], $p = 2.2e{-}07$
Total number of inpatient admissions	10,400 [6515, 14,291], $p = 4.17e{-}07$	5946 [2927, 8965], $p = 0.000157$	4544 [1464, 7625], $p = 0.00466$
Time since start of study period	5.93 [4.14, 7.72], $p = 8.10e{-}10$	17.08 [15.33, 18.83], $p < 2e{-}16$	.21 [−3.67, 4.09], $p = 0.91441$
Success of primary closure	−8497 [−16,493, −500], $p = 0.0374$	−9046 [−15,558, −2533], $p = 0.006864$	−11,180 [−17,851, −4514], $p = 0.00146$
Osteotomy at successful closure	−5302 [−14,003, 3400], $p = 0.2306$	3023 [−4060, 10,106], $p = 0.399940$	−7320 [−14,116, −524], $p = 0.03532$
Adjusted R^2	0.8026	0.8862	0.7106
Number of observations	162	131	56

Results reported as Beta [95% CI] with p values. Total inpatient LOS, total number of inpatient admissions, and time since start of study period are all centered at their respective means for ease of model interpretation.

referral center that sees patients from throughout the world. The study institution has a very specific postoperative management course that may not be reflective of exstrophy care elsewhere. Because of these factors the results may be limited in generalizability, and are a point of emphasis in light of past demonstration that perceived outcomes by pediatric urologists differ from those in the published literature [7]. Finally, this is a retrospective cohort study and carries with it all the limitations of a retrospective study.

In an effort to address these limitations, the authors intend to conduct future research into the heretofore mentioned hypotheses regarding the cost, charge and fee gap between failed and successful PC, specifically addressing the potential contributions of operative time, immobilization time after osteotomy, and immobilization technique. In addition, in order to determine whether the results herein are generalizeable to exstrophy care elsewhere the authors recommend establishment of a multicenter, prospective registry to investigate optimal management of exstrophy patients in an effort to both improve outcomes and reduce costs to families and the healthcare system as a whole.

Ethical Approval

The study outlined herein has been approved by the study institution's institutional review board (IRB00051221).

Conflicts of Interest

The authors claim no conflicts of interest.

Acknowledgments

None.

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