Journal of Pediatric Surgery 51 (2016) 622–625

Contents lists available at ScienceDirect

Journal of Pediatric Surgery

journal homepage: www.elsevier.com/locate/jpedsurg

Continence in the cloacal exstrophy patient: What does it cost?

Seth D. Goldstein [a], Brian M. Inouye [b], Sunil Reddy [b], Kathy Lue [b], Ezekiel E. Young [b], Mahmoud Abdelwahab [b], Mehnaj Grewal [b], Spencer Wildonger [a], Andrew A. Stec [b], John P. Gearhart [b,*]

[a] The Johns Hopkins University School of Medicine, Division of Pediatric Surgery, 600 N. Wolfe Street/Harvey 319, Baltimore, MD, USA, 21287-0005
[b] The Johns Hopkins University School of Medicine, James Buchanan Brady Urological Institute, Division of Pediatric Urology, 1800 Orleans St. Suite 7304, Baltimore, MD, USA, 21287

ARTICLE INFO

Article history:
Received 9 June 2015
Received in revised form 1 December 2015
Accepted 3 December 2015

Key words:
Cloacal exstrophy
Cost analysis
Continence

ABSTRACT

Background: Surgical advancements have made cloacal exstrophy (CE) a survivable condition, though management remains complex. Urologic, orthopedic, colorectal and gynecologic interventions are not standardized, and the cost of this care is high. While the importance of a successful primary closure in terms of outcomes is known, the economic consequences of failure remain uncharacterized.
Methods: A prospectively maintained institutional database of epispadias–exstrophy complex patients was reviewed for continent CE patients. Hospital charges for all inpatient admissions prior to achieving urinary continence were inflation-adjusted to year 2013 values using Consumer Price Index for medical care published by the United States Bureau of Labor Statistics. Records for which charge data were incomplete were completed by using single mean imputation, also inflation-adjusted. Descriptive data are presented as mean ± standard deviation (SD).
Results: Of 102 CE patients, 35 had available hospital charge data: 15 who underwent successful primary closure at the authors' institution and 20 who presented after previously failed primary closures at referring institutions. The mean ± SD hospital charges for primary closure in the success group were $136,201 ± $48,920. These patients then underwent subsequent additional surgeries that accrued charges of $59,549 ± $25,189 in order to achieve continence. Overall, successful primary closures accumulated hospital charges of $200,366 ± $40,071. In comparison, patients referred after prior failure required significantly more hospital admissions and additional charges of $207,674 ± $65,820 were required to achieve continence ($p < 0.001$). Patients who failed primary closure are estimated to accumulate 70% more total health care charges compared to the group following successful primary closure.
Conclusion: The cost of CE management until urinary continence is high, averaging more than $200,000 in inpatient hospital charges alone. Initial success is desirable from both an outcomes and economic perspective, as the cost of salvaging a failed primary closure at our institution is similar to the overall costs of a successful closure; this is in addition to the cost of any previous failed closures. Further studies will be required to determine the optimal timing of surgical management in terms of both patient outcomes and financial consequences.

Cloacal exstrophy (CE) is a rare multisystem congenital anomaly involving the gastrointestinal, nervous, musculoskeletal, and genitourinary systems that occurs with an incidence of 1:200,000 to 1:400,000 live births [1]. Although a historically incurable and fatal condition, advancements in surgical management have made CE a uniformly survivable disease with improved quality of life [2,3]. Although each patient has unique anatomic features, several universal principles exist for the management of CE. These include: neurologic evaluation for myelomeningocele, immediate omphalocele closure, intestinal diversion, posterior approximation of exstrophied hemibladders, and eventual abdominal wall closure with placement of the bladder deep into the pelvis. Nevertheless, the sequence comprising the modern staged repair of exstrophy remains complex and the timing of the urologic, orthopedic, and gastrointestinal interventions is not standardized [4,5].

The health care costs associated with multifaceted CE management are high. As health care spending is increasingly scrutinized in the modern era, quality of care is progressively considered a function of both outcomes and cost [6,7]. While the importance of a successful primary closure in terms of outcomes has been described, the economic consequences of failure remain uncharacterized. The purpose of this study is to describe the hospital charges associated with achieving urinary continence for children with cloacal exstrophy and the economic ramifications of failed initial closure.

1. Materials and methods

A prospectively maintained institutional database of 1211 epispadias–exstrophy complex patients seen by one experienced pediatric urologist at the authors' institution between 1975 and 2014 was reviewed for continent CE patients. Urinary continence was defined as dryness with voiding or catheterization at 3-h intervals. Inpatient hospital charges were available from 1983 onwards. Charges for all inpatient

* Corresponding author. Tel.: +1 410 955 2960; fax: +1 410 502 5314.
E-mail addresses: sgoldstein@jhmi.edu (S.D. Goldstein), jgearha2@jhmi.edu (J.P. Gearhart).

http://dx.doi.org/10.1016/j.jpedsurg.2015.12.003
0022-3468/

admissions prior to achieving urinary continence were inflation-adjusted to year 2013 values using the Consumer Price Index for medical care published by the United States Bureau of Labor Statistics. Records for which charge data were incomplete were completed by using single mean imputation if less than 10% of the hospital charge breakdown was missing. Hospital charges are reported as mean ± standard deviation (SD) when applicable. Patient characteristics and clinical outcomes were obtained from the hospital electronic medical record and from correspondence with referring physicians when applicable.

Patient data were divided into 2 groups for analysis: successful primary closure (group 1) and failed initial closure (group 2). Failed bladder closure was defined as either protrusion of the bladder through a dehisced abdominal wall closure, prolapse of a functionally or cosmetically significant portion of the bladder through the urethral opening, or bladder outlet obstruction. Using STATA IC12 (StataCorp, College Station, TX), select variables from patients in groups 1 and 2 were analyzed using chi-square analysis for categorical variables and Student's t-test for continuous variables.

At the authors' institution, patients with cloacal exstrophy will generally undergo immediate repair of their myelocystocele (if applicable) followed by omphalocele repair and intestinal diversion. During omphalocele repair, the posterior aspects of the exposed hemibladders are aligned to allow the bladder template to enlarge, which will facilitate later closure. Given the very wide diastases and multiple associated anomalies seen in CE, a staged approach is undertaken when performing exstrophy closure. Six months after omphalocele repair and intestinal diversion, the patient undergoes bilateral iliac osteotomies with an external fixation device, followed 3 weeks later by bladder closure and genital revision. In the rare event of no neurological defect, a small omphalocele, and a good bladder template, newborn closure with osteotomy is considered.

2. Results

Of 102 CE patients, 35 (34.3%) patients who achieved urinary continence had available hospital charge data: 15 (42.9%) who underwent successful primary closure at our institution and 20 (57.1%) who presented after previously failed primary closures at referring institutions. Median follow up time after final closure was 9 years, range 13 months to 29 years.

The mean ± SD hospital charges for the sequence of omphalocele closure, bladder closure, and osteotomy was $204,543 ± $56,364 (Table 1). The principle component of the hospital charges was the routine bed rate for inpatient admission, followed by operating room time and operating room supply charges. Table 2 compares patients who underwent successful primary closure to those who were referred to our center after failed closure at another institution. The failed group was significantly older at the time of definitive closure (65 versus 14 months, $p < 0.001$) and required more inpatient hospitalizations to achieve continence (mean 3.8 versus 2.7, $p = 0.001$), though continence was ultimately achieved at similar ages (mean 109 versus 103 months, $p = 0.75$). Thirteen of 15 (86.7%) in the successful closure group and 18 of 20 (90%) in the failed closure group underwent bladder augmentation ($p = 0.99$).

Table 1
Institutional inflation-adjusted hospital charges for modern staged repair of cloacal exstrophy.

Billing category (2013 inflation adjusted)	All patients (n = 35)
Bed rate charges, mean ± SD, USD	$118,309 ± $34,305
Operating room charges, mean ± SD, USD	$36,107 ± $11,719
OR supply charges, mean ± SD, USD	$19,472 ± $6863
Pharmacy charges, mean ± SD, USD	$9467 ± $4350
Radiology charges, mean ± SD, USD	$5910 ± $2361
Laboratory charges, mean ± SD, USD	$6440 ± $2833
Total hospital charges, mean ± SD, USD	$204,543 ± $56,364

In the successful closure group, the mean ± SD hospital charges associated with omphalocele and bladder closure was $136,201 ± $48,920. This figure is unknown for the failed group, as outside hospital charges could not be assessed. However, for subsequent calculations this is assumed to be equivalent for both groups. After presenting with prior failed closure, this group accrued additional hospital charges on average of $124,504 ± $59,487. Overall, successful primary closures accrued hospital charges of $200,366 ± $40,071, whereas the failed group is estimated to have been charged an average of $343,874 ± $65,821.

3. Discussion

Advances in pediatric surgical care have made previously fatal anomalies such as cloacal exstrophy into survivable conditions. The cost of health care for these children is high and is greatly affected by the occurrence of complications. Particularly for a staged surgical sequence such as modern CE management, failure of an index operation has drastic consequences for the patient and their family. When considering the CE population as a whole, the financial burden to the U.S. health care system is minor in comparison to 10% of the population with chronic conditions that consumes about 64% of health care expenditures [6]. Many physicians assume excessive amounts of health care dollars are spent on “million dollar babies”—patients who spend long periods in intensive care units and require a multitude of interventions. However, an analysis of almost 20 million insured patients revealed that only 255 patients had consumed more than $1 million each on health care expenditures in 2010, constituting 0.5% of all health care costs. Expanding this group to patients who consume more than $250,000 in health care expenditures per year would equate to 6.5% of health care costs [6]. In this study, we demonstrate that the cost of CE management from birth until achievement of urinary continence averages more than $200,000 in hospital charges alone, and that failure of primary closure nearly doubles those charges in addition to the detrimental effects on quality of life. Although this may not amount to a substantial yearly health care system cost owing to its rarer incidence and distribution of operations over the years, the individual burden far exceeds what most patient's families are prepared for.

Historically, the modern era of CE management began in 1959 when Rickham presented the first long-term survivor of exstrophy surgery [8,9]. Currently, patients may be undergo one-stage CE closure within the first days of life or alternatively, a staged approach as described above routinely practiced at the authors' institution [10]. In either case, failure to achieve primary closure of the bladder template is a catastrophic setback for patients. This has been previously described both in terms of detrimental outcomes associated with failure as well as investigation of the risk factors that may lead to such. Based on the authors' experience, delaying initial bladder closure 3–6 months after omphalocele repair, using a staged pelvic closure approach with a delay between osteotomy and bladder closure, utilization of an intersymphyseal bar to secure the pubic symphysis during closure, and secure immobilization following closure using Buck's traction and external fixation are practiced strategies to prevent bladder closure failure [11].

Unlike the increased cost of salvaging a failed classic bladder exstrophy closure, the cost of a failed cloacal exstrophy closure was similar to that of the primary closure. Nelson et al. found that there was significant variation in charges based on timing of surgery, with newborn primary repair being less costly than reclosure of failed primary or delayed primary repair ($p < 0.0001$). The most influential factors were operative times and inpatient hospitalizations for newborns [12]. The similarity of costs in closure of our primary versus failed CE repairs concurs with their findings, as most patients at our institution underwent delayed repair with osteotomy beyond the newborn period, reducing the variability in the time of surgery, procedures performed, and operative time. Additionally, pelvic osteotomy and postoperative immobilization have been shown to decrease the rate of failure of redo repairs in carefully selected bladder exstrophy patients, which could similarly