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Cost Determinants of Mandibular Distraction Osteogenesis in Infants With Robin Sequence

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Background: Robin sequence is an anomaly of micrognathia, glossoptosis, and airway obstruction. Mandibular distraction osteogenesis (MDO) performed in early infancy relieves airway obstruction and improves feeding difficulties. Though clinical outcomes data for MDO are strong, studies examining the cost drivers of the procedure are scarce.

Methods: A retrospective 10-year single-institution chart review examined medical and billing records of infants undergoing MDO at an urban tertiary care center. Data included hospital charges, patient characteristics, comorbidities/complications, intubation duration, and length of stay (LOS). Multivariate regression analysis determined significant cost contributors over the course of admission. Conclusions from this smaller sample were compared with analyses from a larger, less detailed, population-based inpatient registry using the Kids' Inpatient Database (2016 and 2019).

Results: In the single-institution analysis, 29 cases were identified with a mean age of 12 days at hospital admission. Mean postoperative and overall LOS were 19 ± 10 and 31 ± 13 days, respectively. Mean total charges were \$287K–\$118K. The most significant driver of total charges was floor charges ($P < 0.01$). LOS was a proxy for floor charges, as they were highly correlated ($r = 0.98$). LOS was significantly driven by intubation duration ($P = 0.01$). In the nationwide analysis, 165 weighted cases were identified; mean age was 2 months at hospital admission. Mechanical ventilation >96 hours was associated with increased LOS and hospitalization costs.

Conclusions: Of infants with Robin sequence undergoing MDO, the most significant driver of total charges was LOS. Intubation duration was highly associated with LOS. *(Plast Reconstr Surg Glob Open 2025;13:e6550; doi: 10.1097/GOX.0000000000006550; Published online 14 February 2025.)*

INTRODUCTION

Robin sequence (RS), a triad of micrognathia, glossoptosis, and airway obstruction, occurs in approximately 1 in 8500 live births, with an estimated 500 new cases annually in the United States.[1,2] Clinical manifestations include gasping or aspiration with feeding, gastroesophageal reflux, oxygen desaturations, apnea, and failure to thrive.[3,4] Conservative treatments for infants with RS include nasogastric tube placement, nasopharyngeal airway placement, or prone positioning.[4,5] However, it is estimated that 30%–70% of RS cases require surgical intervention with tongue-lip adhesion (TLA), tracheostomy, and/or mandibular distraction osteogenesis (MDO) for airway obstruction.[1,4,5,6]

MDO, an increasingly popular alternative to TLA and tracheostomy, is a surgical intervention aimed at pulling the tongue forward to relieve airway obstruction by lengthening the mandible.[5,7] MDO can be performed with either external or internal distraction devices, the latter facilitating wound care and reducing facial scarring in the long term.[8–10] Current literature suggests that MDO offers cost savings over tracheostomy and TLA in symptomatic infants with RS, with MDO having one-quarter of the complication rate of tracheostomy or TLA.[5,7,10] In addition, among patients with isolated RS, MDO results in a greater decrease in apnea/hypopnea index and a higher

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postoperative oxygen saturation compared with TLA.[9,11] However, other studies have shown that although MDO was more effective at reducing airway obstruction, it had higher complication rates than TLA.[12]

Despite fewer complications and secondary procedures compared with tracheotomy and TLA, MDO is more costly upfront, with long-term savings compounding over time via lower secondary and outpatient expenses.[3,5,13] Previous database studies have suggested that length of stay (LOS) is the primary cost driver of admission for surgical management of RS.[13] However, such reports may be limited by the accuracy of their data related to variations in coding, institutional billing practices, and regional differences in cost.[13] Therefore, various potential charges attributable to MDO must be examined on a more granular level to better understand the cost determinants of MDO-related admissions. This study evaluates the detailed charges associated with neonatal MDO care episodes at a single institution, with the primary aim of determining the key contributors to total costs. Our second aim was to corroborate the consistency of our findings by providing external validation of our relatively small sample through correlation with a national patient database to identify other potential contributors to inpatient costs during an MDO-related admission and broaden the applicability of our findings.

Takeaways

Question: What are the cost determinants of mandibular distraction osteogenesis–related admissions for infants with Robin sequence?

Findings: Based on single-institution chart review and nationwide database analysis, the most significant drivers of total charges were the length of intubation and length of stay (LOS), including intensive care unit LOS, which correlated with floor charges and room/board expenses.

Meaning: Clinicians and institutions may lower charges related to infant mandibular distraction osteogenesis hospitalization by reducing overall hospital and intensive care unit LOS, as well as minimizing the duration of mechanical ventilation in these patients.

MATERIALS AND METHODS

A retrospective case series was performed at a single tertiary urban academic medical center. Approval was provided by our institutional review board. All patients under 90 days old who underwent MDO as a primary intervention for isolated or syndromic RS between January 2011 and January 2021 were identified and evaluated for inclusion. Any patient admitted to our institution before undergoing MDO, either as a transfer from an outside hospital or an in-hospital birth admission, was considered eligible.

Infants included in the study underwent MDO with internal linear devices or multivector external devices. Only patients with internal devices underwent virtual surgical planning (VSP). Following MDO, patients were not discharged until deemed capable of independently sustaining oral or enteral intake and oxygenation. From our institution's electronic medical records, patient characteristics including demographics, comorbidities, medical or surgical complications, mechanical ventilation duration, MDO device type (internal versus external), use of total parenteral nutrition, nasogastric tube placement, peripherally inserted central catheter (PICC) placement, and LOS were collected.

Fees charged for each MDO-associated admission were obtained from our institution's billing database and were organized into fixed and variable cost categories (Table 1). Changes in billing structure occurred at our institution after 2017, resulting in an adjustment of billing rates for nursing staff and room/board in the neonatal intensive care unit, pediatric intensive care unit, and floors. We controlled for this by conducting significance testing on 2 separate samples: patients undergoing MDO between January 2011 and December 2017 and those undergoing MDO between January 2018 and January 2021. Statistical analyses, including significance testing and multivariate regression modeling, were performed (Microsoft Excel).

To examine broader nationwide trends, a population-based analysis was performed using the 2016 and 2019 Kids' Inpatient Database (KID), a national database created by the Healthcare Cost and Utilization Project and Agency for Healthcare Research and Quality.[14,15] Because this database extrapolates for nationwide estimates, all statistical analyses used the provided weighting. The International Classification of Diseases, 10th Revision, Procedure Coding System was used to identify patients admitted under 4 months of age undergoing procedures representing MDO (Table 2).[16,17] Admissions without International Classification of Diseases, 10th Revision, Clinical Modification codes suggestive of RS were excluded from the MDO cohort.[18] An independent 2-sample *t* test was used to compare continuous variables. Statistical significance of odds ratios was determined with a chi-square test. IBM Statistical Package for Social Sciences version 28 (IBM Corp, Armonk, NY) was used for multivariate analysis. Linear regression models were created to determine significant variables associated with hospitalization charges and LOS. The correlation between total

Table 1. Categories of Different Costs Associated With an Episode of Care in Which MDO Was Performed, Separated by Fixed and Variable Costs, the Latter Depending on Factors Such as LOS or Complications

Fixed Costs	Variable Costs
Sterile supplies	Anesthesia services
Nonsterile supplies	Audiology services
Imaging (MRI, CT)	Laboratory/cytology/microbiology testing
Laryngoscopy/diagnostic procedures	Medications/pharmacy
Cardiac evaluation (EKG, echocardiogram)	Operating room services
	ICU staffing and room/board
	Speech/language pathology
	Respiratory/pulmonary services
	Occupational/feeding therapy

CT, computed tomography; EKG, electrocardiogram; ICU, intensive care unit; MRI, magnetic resonance imaging.

Table 2. ICD-10-CM and ICD-10-PCS Codes Used for Nationwide Database Analysis

Associated Diagnosis	ICD-10-CM Codes
VSD	Q21.0
ASD/PFO	Q21.1
PDA	Q25.0
Preterm newborn	P07.2, P07.3
Procedure	ICD-10-PCS codes
Reposition of mandible with internal or external fixation device	0NST04Z, 0NST05Z, 0NST34Z, 0NST35Z, 0NST44Z, 0NST45Z, 0NSV04Z, 0NSV05Z, 0NSV34Z, 0NSV35Z, 0NSV44Z, 0NSV45Z, 0NST0ZZ, 0NSV0ZZ
Division of mandible	0N8T0ZZ, 0N8T3ZZ, 0N8T4ZZ, 0N8V0ZZ, 0N8V3ZZ, 0N8V4ZZ
Mechanical ventilation > 96 h	5A1955Z
Mechanical ventilation < 96 h	5A1935Z, 5A1945Z
Red blood cell transfusion	30233N1, 30243N1
TPN/PPN	3E0336Z, 3E0436Z
Insertion of infusion device into inferior vena cava, superior vena cava, or right atrium, percutaneous approach	06H033Z, 02HV33Z, 02H633Z
Insertion of feeding device into the stomach, via natural or artificial opening (nasogastric tube)	0DH67UZ
Tracheotomy	0B110F4, 0B110Z4, 0B113F4, 0B113Z4, 0B114F4, 0B114Z4
Gastrostomy tube placement	0DH60UZ, 0DH63UZ, 0DH64UZ
Airway endoscopy	0CJS4ZZ, 0CJS8ZZ, 0BJ04ZZ, 0BJ08ZZ, 0BJ14ZZ, 0BJ18ZZ

ASD, atrial septal defect; ICD-10-CM, International Classification of Diseases, 10th Revision, Clinical Modification; ICD-10-PCS, International Classification of Diseases, 10th Revision, Procedure Coding System; PDA, patent ductus arteriosus; PFO, patent foramen ovale; PPN, peripheral parenteral nutrition; TPN, total parenteral nutrition; VSD, ventricular septal defect.

hospitalization charges and LOS was calculated using the Pearson correlation.

RESULTS

Between January 2011 and January 2021, 29 infants under 90 days old underwent MDO at our institution, with a mean age at admission of 11.8 days (range 0–58 d). Seventeen (59%) infants had preoperative comorbidities, most of which were cardiopulmonary (59%), including laryngomalacia, intubation at birth, and congenital heart disease. In the KID nationwide analysis, 165 weighted cases were identified with a mean age of 2 months at hospital admission. Patient characteristics and other preoperative comorbidities are described in Table 3.

Eleven (38%) infants experienced postoperative complications: a majority were cardiopulmonary (64%), including atelectasis, mucous plugging, delay in extubation, hypercapnic respiratory failure, and cardiac arrest. Noncardiopulmonary complications (36%) included vocal cord trauma, occipital pressure wounds, and infectious concerns (facial cellulitis or aspiration pneumonia). There were no deaths, and all patients were discharged home. Hospital procedures, hospital course metrics, and postoperative complications are summarized in Table 4.

Six infants who underwent MDO (all occurring before 2018) had incomplete or missing charges data and were excluded from the cost analysis. Mean total charges for MDO-related admission were \$287.1K ± \$118.4K; Figure 1 depicts the distribution of total episodic costs across our sample. A breakdown of the mean charges for an MDO-related admission at our institution is depicted in Figure 2. Mean nursing staff and room/board before 2018 were \$3188 per day and were \$8524 per day for 2018 onward. This adjustment was made based on market rates for hospital charges among nearby academic medical centers. In the KID nationwide data, mean total charges were \$514.3K ± \$531.0K.

For both groups within our institution, the multivariate regression of total MDO episode charges versus individual cost drivers was significant ($F = 0.002$ before 2018, $F < 0.001$ for 2018 onward) (Table 5). Both models found that the primary determinants of overall charges were room/board and nursing staff expenses, accounting for approximately 62% of the mean overall charges ($P = 0.004$ before 2018, $P < 0.001$ for 2018 onward). Both models found that sterile and nonsterile supplies were proportionately associated with overall charges ($P = 0.007$ before 2018, $P = 0.005$ for 2018 onward), accounting for approximately 15% of mean overall charges. The largest component of sterile and nonsterile supplies were surgical instruments, equipment used in the procedure, and the mandibular distractors themselves.

LOS was highly correlated with cumulative daily nursing floor charges ($r = 0.97$ before 2018, $r = 0.99$ for 2018 onward) and was, therefore, assumed to be a strong predictor of total floor charges for a patient in our sample. Multivariate regression suggests that postoperative day of extubation was the only variable associated with LOS ($P = 0.01$); overall LOS increased by a mean of 4.2 days for every day a patient remained on a ventilator postoperatively (Table 6). No patients at our institution were reintubated after extubation. In the KID data, LOS and total hospital charges were also highly correlated ($r = 0.66$). Multivariate regression modeling using the KID data shows that mechanical ventilation for more than 96 hours was associated with both increased hospitalization charges ($P = 0.0016$) and increased LOS ($P = 0.025$). Preterm birth, patent ductus arteriosus, and total parenteral nutrition/peripheral parenteral nutrition were also associated

Table 3. Patient Demographics and Comorbidities for the Overall Cohort, Including Comparison Between Those Included and Excluded From the Episodic Cost Analysis

Patient Characteristics	Total (n = 29)	Included (n = 23)	Excluded (n = 6)	*P*	KID Results (n = 165)	*P*
	Mean (Min, Max) or n (%)				n (%)	
Mean age at admission, d	11.8 (0.0, 58.0)	13.3 (0.0, 58.0)	6.0 (0.0, 26.0)	0.22	—	
Mean age at procedure, d	24.0 (9.0, 65.0)	25.8 (9.0, 65.0)	17.3 (12.0, 29.0)	0.06	—	
Sex assigned at birth, female	14 (48)	11 (48)	3 (50)	0.92	80 (48.2)	0.98
Race/ethnicity						
White	19 (66)	13 (57)	6 (100)	0.046	88 (53.3)	0.23
Non-White	10 (34)	10 (43)	0 (0)	0.046	77 (46.7)	0.23
Black	—	—	—	—	—*	
Hispanic	—	—	—	—	22 (13.5)	
Asian/Pacific Islander	—	—	—	—	—*	
Native American	—	—	—	—	—*	
Other	—	—	—	—	11 (6.5)	
Preoperative intubation	4 (14)	3 (13)	1 (17)	0.82	—	
Admitted as a transfer	22 (76)	17 (74)	5 (83)	0.50	76 (46.1)	<0.01
Internal distraction device	10 (34)	10 (43)	0 (0)	0.046	—	
External distraction device	19 (66)	13 (57)	6 (100)	0.046	—	
Prematurity	6 (21)	6 (26)	0 (0)	0.16	25 (15.1)	0.41
Syndrome	9 (31)	6 (26)	3 (50)	0.26	—	
Patient comorbidities	17 (59)	13 (57)	4 (67)	0.65	—	
Cardiopulmonary	10 (34)	6 (26)	4 (67)	0.063	—	
VSD	2 (7)	1 (4)	1 (17)	0.989	13 (8.0)	0.855
ASD/PFO	17 (59)	16 (70)	1 (17)	0.039	70 (42.5)	0.110
PDA	4 (14)	4 (17)	0 (0)	0.481	32 (19.6)	0.863
Noncardiopulmonary (infectious, neurological, social work concerns, etc.)	7 (24)	7 (30)	0 (0)	0.12	—	

Nationwide database analysis for similar patient demographics and comorbidities within the MDO population.
*Value less than 10.
ASD, atrial septal defect; CI, confidence interval; max, maximum; min, minimum; OR, odds ratio; PDA, patent ductus arteriosus; PFO, patent foramen ovale; VSD, ventricular septal defect.

with both increased LOS and hospitalization charges (Table 7).

DISCUSSION

Our aim was to examine the associations among patient characteristics, hospital course, and costs associated with MDO. We investigated costs for unique episodes of care among patients undergoing MDO at a tertiary care center, as well as cost determinants nationwide for MDO-related admissions. Within our institution's dataset, we were able to analyze detailed aspects of each admission such as operating room services, room/board costs, staffing charges, and patient features such as days of ventilation, data that were not available in the nationwide dataset.

We found that overall charges for MDO were primarily driven by cumulative daily expenses of room/board, nursing staff charges, and surgical sterile and nonsterile supplies. Cumulative daily room/board and staffing expenses were highly correlated with LOS, and multivariate analysis showed a significant association with the duration of postoperative mechanical ventilation and overall LOS, which drove much of the episode of care charges. These findings were consistent with the results of the nationwide KID analysis. Notably, mechanical ventilation for more than 96 hours was also significantly associated with increased LOS in the KID dataset. As such, both our institutional data and national database analysis demonstrate strong concordance in findings that LOS and duration of mechanical ventilation are the primary drivers of total charges among young infants undergoing MDO. Thus, the findings of the 2 analyses are complementary and suggest applicability of these results beyond our institution.

Many infants with RS who undergo MDO require mechanical ventilation; however, the optimal duration of intubation to prevent adverse outcomes that may prolong hospitalization and incur additional costs is not well established. Several studies have investigated MDO and intubation duration, reporting varying results ranging from 1 to 46 days.[19–22] Zhang et al[22] found that intubation for a minimum of 5 days (6 for syndromic patients) was associated with successful extubation after MDO. Similarly, a retrospective analysis reported an optimal intubation duration of 6 days after MDO.[23] In addition, the amount of distraction may be an important factor in successful extubation as lengthening of the mandible seems to reduce extubation difficulties.[19,23] On the other hand, prolonged intubation following any form of surgery may increase the risk for complications including postoperative pulmonary infection and, among pediatric patients undergoing cardiac surgery, lengthier hospital stays and increased mortality.[24] Therefore, practitioners should work to tailor intubation duration based on individualized patient characteristics, balancing the risk of extubation failure with avoiding lengthy intubation periods, the latter of

Table 4. Hospital Metrics Including LOS and Charges Associated With an Entire Episode of Care Involving MDO for the Overall Cohort as Well as Differentiated by Those Included and Excluded From the Episodic Cost Analysis

	Total (n = 29)	Included (n = 23)	Excluded (n = 6)		KID Results (n = 165)	
	Mean ± SD or n (%)			*P*	n (%) or Mean (STD; Min, Max)	*P*
Hospital course						
Postoperative complications	11 (48)	10 (43)	1 (17)	0.22	—	
Cardiopulmonary	7 (24)	6 (26)	1 (17)	0.63	—	
Noncardiopulmonary (infectious, trauma, etc.)	4 (14)	4 (17)	0 (0)	0.27	—	
Additional interventions		42.7 ± 16.0				
pRBC transfusion	6 (21)	6 (26)	0 (0)	0.16	11 (6.5)	0.02
PICC line	23 (79)	21 (91)	2 (33)	0.002	31 (18.7)*	<0.01
NGT placement	19 (66)	15 (65)	4 (67)	0.95	—†	<0.01
Gastrostomy tube placement	2 (7)	2 (9)	0 (0)	0.80	32 (19.3)	0.12
TPN	18 (62)	14 (61)	4 (67)	0.79	25 (15.4)‡	<0.01
Tracheotomy	1 (3)	1 (4)	0 (0)	0.93	—*	
Cardiac evaluation		3.7 ± 2.7			—	
Medications/pharmacy		3.3 ± 1.9			—	
Hospital metrics, d						
Days on ventilator	5.4 ± 1.8	5.4 ± 2.0	5.2 ± 0.75	0.61	—	
Mechanical ventilation > 96 h	19 (66)	14 (61)	5 (83)	0.321	80 (48.1)	0.095
Mechanical ventilation < 96 h	9 (31)	8 (35)	1 (17)	0.406	28 (17.1)	0.081
Any mechanical ventilation	28 (97)	22 (96)	6 (100)	0.933	101 (61.2)	0.005
Hospital LOS	30.9 ± 12.8	31.1 ± 13.6	30.0 ± 10.5	0.84	44.4 (39.1; 1–277)	
ICU LOS	28.7 ± 14.1	28.3 ± 15.1	30.0 ± 10.5	0.76	—	
Preoperative LOS	12.3 ± 8.1	12.5 ± 8.6	11.3 ± 6.0	0.70	—	
Postoperative LOS	18.6 ± 10.2	18.6 ± 11.2	18.7 ± 6.1	0.98	—	

Nationwide database analysis for similar hospital metrics within the MDO population.
*Insertion of infusion device into inferior vena cava, superior vena cava, or right atrium, percutaneous approach.
†Value less than 10.
‡Peripheral parenteral nutrition.
CI, confidence interval; ICU, intensive care unit; max, maximum; min, minimum; OR, odds ratio; NGT, nasogastric tube; pRBC, packed red blood cell; TPN, Total parenteral nutrition.

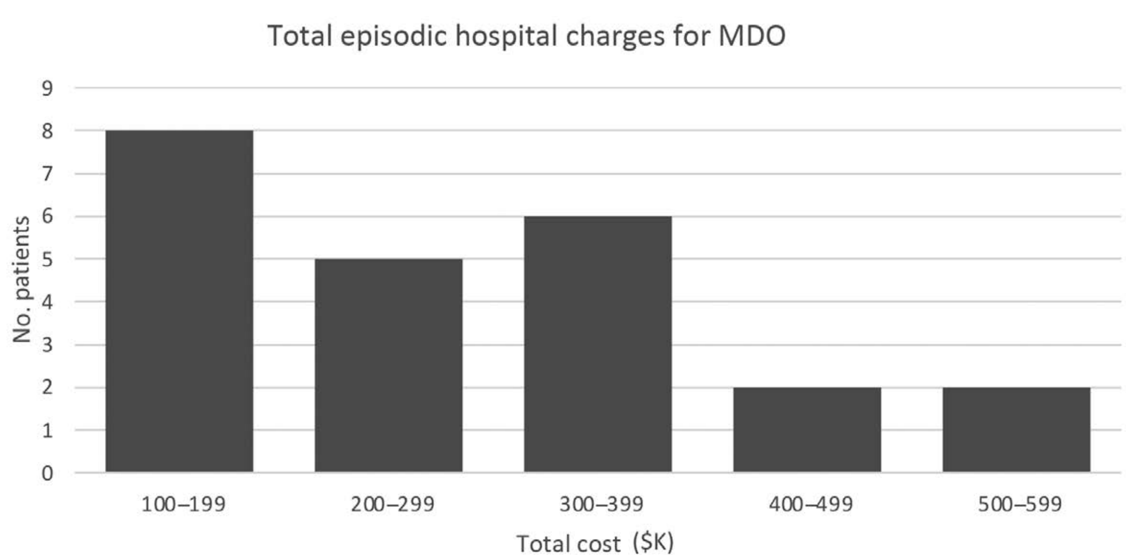

Fig. 1. Histogram displaying the distribution of total hospital charges for an episode of MDO for 23 patients with available billing records in our sample.

which seems to be an important contributor to overall LOS following MDO.

Despite overall congruence in findings, there were some notable differences between our institutional series and the national data presented herein. Although our institutional analysis found mean total charges for a neonatal MDO admission to be \$287.1K ± \$118.4K, the nationwide database analysis estimated mean total charges of twice that rate (\$514.3K ± \$531.0K), indicating a high level of variability. Application of our institution's Centers

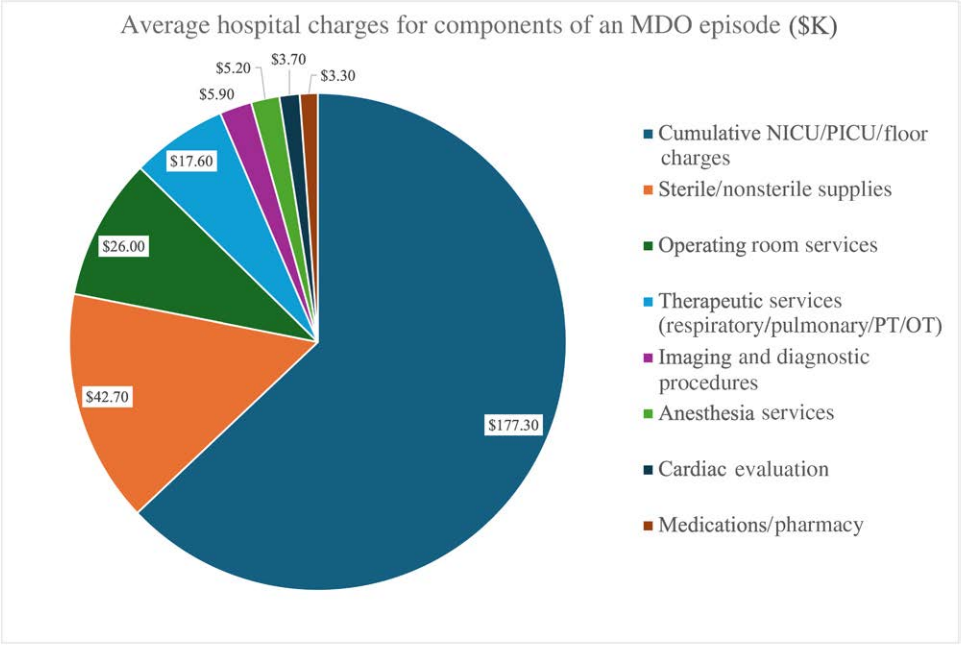

Fig. 2. Pie chart depicting the mean distribution for the majority of cost components for an episode of MDO. ICU, intensive care unit; NICU, neonatal intensive care unit; OT, occupational therapy; PICU, pediatric intensive care unit; PT, physical therapy.

Table 5. Linear Regressions of the Largest Cost Components of an Episode of MDO Against the Total Cost of an Episode of MDO, Controlling for Change in Billing Structure Made 2018 Onward

	Episodes of MDO From 2011 to 2017 (n = 17)			Episodes of MDO From 2018 to 2021 (n = 12)		
Effect on Total MDO Episodic Cost	**Estimate**	**95% CI**	***P***	**Estimate**	**95% CI**	***P***
Intercept	−663.98	−59,807 to 58,479	0.97	−3757.51	−65,022 to 57,507	0.86
Cardiac evaluation	1.07	−5.21 to 7.35	0.54	0.72	−2.66 to 4.10	0.54
Imaging and diagnostic procedures	1.56	−11.9 to 15.0	0.67	0.57	−1.50 to 2.64	0.44
Therapeutic services (respiratory/pulmonary/PT/OT)	1.32	−1.17 to 3.81	0.15	0.73	−0.01 to 1.46	0.05
NICU/PICU/floor R&B and staffing	1.04	0.75 to 1.32	<0.01	1.06	0.88 to 1.25	<0.01
Operating room services	0.98	−2.53 to 4.50	0.35	0.80	−0.76 to 2.36	0.20
Anesthesia services	0.89	−3.17 to 4.95	0.44	1.87	−6.41 to 10.2	0.52
Sterile/nonsterile supplies	1.10	0.71 to 1.49	<0.01	1.02	0.60 to 1.45	<0.01
Medications/pharmacy	0.95	−5.32 to 7.21	0.58	3.49	−3.92 to 10.9	0.23

CI, confidence interval; ICU, intensive care unit; NICU, neonatal intensive care unit; OT, occupational therapy; PICU, pediatric intensive care unit; PT, physical therapy; R&B, room and board.

for Medicare and Medicaid Services cost-to-charge ratio of 30% results in the KID estimating total admission costs of $154.3K ± $159.3K, which are also higher than what we observed at a granular level. When estimated cost data reported in the KID are controlled for LOS, daily charges and cost estimates were still higher than what we observed at the single-institution level. This is likely due to a longer mean LOS in the KID analysis compared with our institutional data (44.4 versus 31.1 d), as well as a higher rate of gastrostomy tube placement among patients in the national database.

In the nationwide analysis, 19.3% of patients received gastrostomy tubes compared with 9% of our patients. Although gastrostomy tube placement was not directly associated with increased charges, it was associated with longer LOS (P = 0.001), allowing more time for charges to be accrued. Indeed, Marston et al[25] found that a lower proportion of patients with RS who received MDO at children's specialty hospitals required nutritional intervention compared with non–children's hospitals. Furthermore, patients seen at larger institutions performed more nutritional interventions and had a higher likelihood of increased LOS and nonroutine charge. This observation may reflect a tendency for more conservative management strategies at non–children's hospitals ultimately leading to additional interventions and increased LOS.[25] Similarly, the wide variance in charges in the national analysis may be explained by both longer LOS and a wider array of comorbidities represented in the sample.

In discussing limitations, it is important to note that the cohort of 23 of 29 (79%) patients who were included

Table 6. Linear Regression of Patient Characteristics, Hospital Procedures, Comorbidities, and Additional Charges, Against Overall LOS for an Episode of MDO

Effect on Overall LOS	Estimate	95% CI	*P*
Intercept	11.41	−9.04 to 31.9	0.25
Age at admission	−0.27	−0.65 to 0.10	0.13
POD extubation	4.19	1.15 to 7.22	0.01
Sex assigned at birth, female (Y/N)	−7.55	−17.7 to 2.55	0.13
Race, non-White (Y/N)	8.42	−2.06 to 18.9	0.11
Transfer from OSH (Y/N)	2.27	−11.1 to 15.6	0.72
Premature (Y/N)	0.32	−14.2 to 14.8	0.96
Preoperative intubation (Y/N)	−1.36	−16.4 to 13.7	0.85
Postoperative complications (Y/N)	1.25	−8.60 to 11.1	0.79
Significant comorbidities (Y/N)	2.80	−8.01 to 13.6	0.59
Internal MDO (Y/N)	−3.61	−13.2 to 6.01	0.43
Syndromic RS (Y/N)	−7.09	−19.9 to 5.72	0.25
PICC line (Y/N)	−6.44	−24.2 to 11.4	0.45
NGT placement (Y/N)	1.10	−8.74 to 10.9	0.81
TPN (Y/N)	8.91	−1.83 to 19.6	0.10

CI, confidence interval; NGT, nasogastric tube; N, no; OSH, outside hospital; POD, postoperative day; TPN, total parenteral nutrition; Y, yes.

Table 7. Linear Regression of Patient Characteristics, Hospital Procedures, and Comorbidities Against Overall LOS and Total Charges for Hospitalization Among Weighted MDO Cases in Infants

	Total Charges for Hospitalization (Thousands of Dollars)		LOS (d)	
	Unstandardized Beta	*P*	Unstandardized Beta	*P*
Age, mo	169.286	0.244	−0.080	0.980
Sex, female	−1.757	0.971	−3.959	0.413
Race/ethnicity	6.043	0.822	−0.098	0.995
Transfer	35.989	0.653	3.24	0.532
VSD	221.299	0.123	11.590	0.211
ASD/PFO	87.459	0.309	−0.038	0.995
PDA	294.810	0.005	28.203	<0.001
Preterm newborn	296.471	0.008	26.775	<0.001
Mechanical ventilation > 96 h	200.668	0.016	12.052	0.025
Mechanical ventilation < 96 h	76.605	0.473	−1.275	0.854
Red blood cell transfusion	−385.096	0.017	−19.354	0.064
PICC/central line	51.255	0.608	0.414	0.949
NGT placement	−137.475	0.551	−1.006	0.946
TPN/PPN	517.555	<0.001	35.727	<0.001
Tracheotomy	307.546	0.150	49.659	<0.001
Gastrostomy tube placement	76.676	0.478	23.418	0.001

ASD, atrial septal defect; PDA, patent ductus arteriosus; PFO, patent foramen ovale; PPN, peripheral parenteral nutrition; TPN, total parenteral nutrition; VSD, ventricular septal defect.

in our institutional financial analyses had multiple potential confounding factors. This subset included all 10 of the patients who identified as non-White from the overall cohort ($P = 0.046$) and all 10 of the patients who underwent internal MDO ($P = 0.046$) (Table 3). Additionally, a significantly greater proportion of patients included in the subgroup (21 of 23 or 91%) underwent PICC line placement and likely incurred additional downstream charges—compared with patients in the excluded group (2 of 6 or 33%) ($P = 0.002$) (Table 4). It is reassuring, however, that in our institutional multivariate regression model examining the relationship between LOS and multiple patient and hospital course characteristics, LOS was not found to be significantly modified by race/ethnicity ($P = 0.11$), PICC placement ($P = 0.45$), or MDO modality ($P = 0.43$) (Table 6). This result is noteworthy, as theoretical cost differences among hardware types or use of VSP may be negated by the cost associated with the duration of hospitalization. Of note, the nationwide database analysis similarly found no association between race/ethnicity and LOS ($P = 0.995$) or PICC/central line placement ($P = 0.949$), suggesting the generalizability of the primary result. Data specific for each MDO hardware type were not available for the national cohort.

Another limitation is the use of VSP for internal but not external distraction hardware, which likely resulted in increased costs in the internal hardware cohort. Our study was unable to detect nuances among the costs of hardware type. For example, internal hardware requires a second hospital admission and surgical removal, whereas external hardware can typically be removed as an ambulatory procedure. Additional costs associated with VSP, along with the advantages and disadvantages of VSP, remain a subject of debate and are beyond the scope of this study.

Our study has additional limitations. Several patients had incomplete financial data for review and were excluded from the cost analysis. In addition, over the years our institution has become more efficient with the management of infants with RS, with a more predictable postoperative LOS.[1,26] However, creation of standardized postoperative management protocols has also introduced consistent utilization of procedures with fixed costs, such as PICC lines and a standardized duration of mechanical ventilation. Our institutional practice has been to discharge patients from the neonatal intensive care unit or pediatric intensive care unit rather than transferring infants to a lower cost step-down unit for monitoring before discharge. We, therefore, have no data to determine if earlier transfer out of the ICU would indeed result in lower charges. The data we have available for review are also subject to error. Some procedures may have been billed inappropriately or not accounted for altogether. In addition, RS is a rare condition with low annual incidence; this is reflected in our institution's average of only 3 neonatal mandibular distraction cases performed each year, potentially qualifying it as an "intermediate" or "low-volume" center. Nevertheless, presenting data from such institutions serves a valuable purpose, as increasingly, neonatal mandibular distraction is being performed at both high-volume and low-volume tertiary pediatric centers. Furthermore, our clinical outcomes and LOS compare favorably with large-volume centers and the KID overall.

Our nationwide KID data analysis is limited by our ability to extract charges for the course of care episodes on the same granular level we were able to for our institution (ie, sterile supplies, imaging, etc.). As such, we were not able to directly compare our institutional data to the KID national data through the same methodology, and this limited our ability to describe the proportion of costs accounted for by MDO alone on a national level. In addition, we were unable to decipher cost differences between individuals with and without syndromic RS. The use of weighted data for KID analysis carries inherent limitations, with the risk of overrepresenting certain populations and introducing biases into the dataset. However, weighted data allow for a large enough sample size to assess nationwide estimates, trends, and costs.

Our study also demonstrates multiple strengths, for example, the availability of robust institutional demographic and clinical data, which allowed thorough multivariate analyses across various timepoints and changes in billing practices. Although our institutional sample size was small, our results are corroborated by the large number of patients assessed in the KID. The findings in both analyses detected an association between the duration of mechanical ventilation and LOS, which in turn was correlated with increased costs. This highlights that cost drivers at our institution are likely representative of national determinants, making the results presented herein both specific and widely generalizable.

A future area for research, with a larger sample, would be to explore whether preoperative LOS differs significantly between direct admissions versus outside hospital transfers and how this contributes to overall cost. Cumulative expenses toward room/board and nursing staff charges varied significantly depending on whether the patient was in an ICU or the pediatric wards. It would be beneficial to determine the clinical manifestations of severe RS that determine which infants with RS must remain in the ICU and whether more medically stable infants can be managed once off mechanical ventilation and stable in a non-ICU setting.

In summary, clinicians and institutions may lower charges related to infant MDO hospitalization by reducing overall hospital and ICU LOS, as well as minimizing the duration of mechanical ventilation in these patients. Limiting the rates of gastrostomy tube placement in infants with RS may also indirectly reduce costs by decreasing LOS. Further studies are necessary to determine the effectiveness of implementing these strategies on clinical outcomes and cost containment.

CONCLUSIONS

Our study suggests that among infants with RS undergoing MDO as a primary intervention for obstructive airway symptoms, most charges associated with an episode of care were an accumulation of floor charges and room/board expenses in the intensive care unit. Surgical expenses related to sterile and nonsterile supplies used for the surgery itself were also important contributors. Floor charges were most directly correlated with LOS, which significantly increased the longer the patient remained on mechanical ventilation postoperatively. Institutional findings complemented those found in a much larger national database. Further research is necessary to explore whether device selection significantly alters clinical outcomes and costs, and how to best minimize LOS in the intensive care unit.

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DISCLOSURE

The authors have no financial interest to declare in relation to the content of this article.

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