Original Investigation

Effect of an Enhanced Medical Home on Serious Illness and Cost of Care Among High-Risk Children With Chronic Illness
A Randomized Clinical Trial

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IMPORTANCE Patient-centered medical homes have not been shown to reduce adverse outcomes or costs in adults or children with chronic illness.

OBJECTIVE To assess whether an enhanced medical home providing comprehensive care prevents serious illness (death, intensive care unit [ICU] admission, or hospital stay >7 days) and/or reduces costs among children with chronic illness.

DESIGN, SETTING, AND PARTICIPANTS Randomized clinical trial of high-risk children with chronic illness (≥3 emergency department visits, ≥2 hospitalizations, or ≥1 pediatric ICU admissions during previous year, and >50% estimated risk for hospitalization) treated at a high-risk clinic at the University of Texas, Houston, and randomized to comprehensive care (n = 105) or usual care (n = 96). Enrollment was between March 2011 and February 2013 (when predefined stopping rules for benefit were met) and outcome evaluations continued through August 31, 2013.

INTERVENTIONS Comprehensive care included treatment from primary care clinicians and specialists in the same clinic with multiple features to promote prompt effective care. Usual care was provided locally in private offices or faculty-supervised clinics without modification.

MAIN OUTCOMES AND MEASURES Primary outcome: children with a serious illness (death, ICU admission, or hospital stay >7 days), costs (health system perspective). Secondary outcomes: individual serious illnesses, medical services, Medicaid payments, and medical school revenues and costs.

RESULTS In an intent-to-treat analysis, comprehensive care decreased both the rate of children with a serious illness (10 per 100 child-years vs 22 for usual care; rate ratio [RR], 0.45 [95% CI, 0.28-0.73]), and total hospital and clinic costs ($16 523 vs $26 781 per child-year, respectively; cost ratio, 0.58 [95% CI, 0.38-0.88]). In analyses of net monetary benefit, the probability that comprehensive care was cost neutral or cost saving was 97%. Comprehensive care reduced (per 100 child-years) serious illnesses (16 vs 44 for usual care; RR, 0.33 [95% CI, 0.17-0.66]), emergency department visits (90 vs 190; RR, 0.48 [95% CI, 0.34-0.67]), hospitalizations (69 vs 131; RR, 0.51 [95% CI, 0.33-0.77]), pediatric ICU admissions (9 vs 26; RR, 0.35 [95% CI, 0.18-0.70]), and number of days in a hospital (276 vs 635; RR, 0.36 [95% CI, 0.19-0.67]). Medicaid payments were reduced by $6243 (95% CI, $1302-$11 678) per child-year. Medical school losses (costs minus revenues) increased by $6018 (95% CI, $5506-$6629) per child-year.

CONCLUSIONS AND RELEVANCE Among high-risk children with chronic illness, an enhanced medical home that provided comprehensive care to promote prompt effective care vs usual care reduced serious illnesses and costs. These findings from a single site of selected patients with a limited number of clinicians require study in larger, broader populations before conclusions about generalizability to other settings can be reached.

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Although the patient-centered or family-centered medical home is widely recommended,[1-5] its value in improving clinical outcomes or reducing health care costs remains to be demonstrated.[3,4] Medical homes are potentially the most cost-effective for high-risk patients,[6] particularly high-risk children with chronic illness whose care is often fragmented, costly, and ineffective[1,7-10] and who account for only 0.4%[11] of all children but approximately 40% of total pediatric hospital charges.[12] However, with the inadequate current payments for outpatient pediatric care[7] and the necessity to restrain health care spending, the payments required to develop and sustain such medical homes may not be forthcoming unless they are shown to improve outcomes with minimal or no increase in costs.

We conducted a randomized clinical trial to assess whether an enhanced medical home providing comprehensive care for high-risk children with chronic illness would reduce serious illnesses, medical costs, or both, from a health system perspective. To promote benefits and cost savings that may not be achievable in typical medical homes,[13] comprehensive care was provided by both primary care clinicians and specialists in the same clinic and included multiple features to promote prompt, effective care at all hours.

Methods

We included patients aged 18 years or younger with a chronic illness[14] who lived within a 1-hour commute of the University of Texas, Houston (UTH) and had high health care use (≥3 emergency department [ED] visits, ≥2 hospitalizations, or ≥1 pediatric intensive care unit [ICU] admissions during the prior year) and a high estimated risk of hospitalization during the coming year (>50% as judged by the clinic's medical director [R.A.M.] based on the patient's diagnosis and clinical course). Enrollment occurred between March 2011 and February 2013, with evaluation of outcomes continuing through August 31, 2013.

We excluded those patients with complex problems who received primary care by a specialist at all hours (eg, infants in the neonatal follow-up program and children with serious unrepaired congenital heart disease, a mitochondrial disorder, organ transplant, treatment with dialysis or central lines; or those with a do-not-resuscitate order). Study candidates were identified from review of faculty billings during the prior year and prospective screening of hospital admissions. Race/ethnicity was self-reported by parents in response to an open-ended question and was assessed to evaluate whether the benefits of comprehensive care relative to usual care were increased for Hispanics and blacks.

The study was approved by the UTH institutional review board as a randomized quality improvement study (eAppendix 1 in Supplement 1) that would increase access to care[15] and was expected to improve outcomes based partly on a prior trial of comprehensive care.[16] Verbal informed consent was allowed.[17-19] The trial protocol appears in Supplement 2.

Participants were stratified by maternal education (high school graduate or not) and estimated risk (high or very high) for hospitalization and then randomized to comprehensive care or usual care by opening sealed, opaque, sequentially numbered envelopes prepared using a variable block size by an investigator (J.E.T.) with no patient contact. If 2 siblings were found to be eligible, both were assigned to the same group.

Usual Care

Acute care was provided by a large number of pediatricians in private offices or in the UTH general pediatrics clinic staffed by residents supervised by general pediatrics faculty. This care was not modified by the study protocol. Same-day care was not always available. Chronic problems were also treated at UTH subspecialty clinics and at a twice-weekly clinic for children with special health care needs staffed by a pediatrician, social worker, and nutritionist who provided consultation and referrals but not primary care. As in other centers, calls from parents to the center at nights and on weekends were taken by on-call faculty or faculty-supervised pediatric residents unlikely to know the child well. Children were referred to the ED without discussion with the ED staff and had no automatic follow-up appointment.

Comprehensive Care

Comprehensive care was provided at the UTH High-Risk Children's Clinic as a medical home,[20] augmented by multiple measures to prevent serious illness. This care was based on an intervention assessed in a previous randomized trial,[16] and was shown to reduce life-threatening illness among high-risk infants after discharge from a neonatal ICU. The clinic was open 40 hours per week and staffed by the medical director (a pediatric pulmonologist with broad interests in children with chronic illness) and 2 pediatric nurse practitioners who provided primary care. The medical director committed 0.4 of time to attending the clinic, shared call with the nurse practitioners, and provided continuous backup. All parents had the cell phone number to directly reach 1 of the primary care clinicians at all hours. A Spanish-speaking clinician was always available, and each clinician could access clinic records from home.

The clinic was also staffed by a nutritionist and social worker. Children were scheduled as needed to see a dedicated pediatric gastroenterologist, neurologist, or allergist/immunologist, who each attended the clinic once monthly. A pediatric infectious disease specialist helped develop measures to reduce, promptly diagnose, and effectively treat infections (eAppendix 2 in Supplement 1). These subspecialists were promptly available by telephone for consultation at all hours.

Patients with acute illness presenting before 5:00 PM on a weekday were seen the same day. Families who called at night or on weekends were generally managed over the telephone with a clinic appointment when needed. Although some families with obvious emergencies went to the ED without first calling, only 3% of telephone calls resulted in a referral to the ED. When ED visits or hospitalizations were needed, the clinicians discussed the child's problems and treatment with the responsible physicians. A timely follow-up visit was arranged before discharge from the ED or

hospital. Brief parent satisfaction questionnaires were routinely completed after each clinic visit. We also obtained active input from the clinic-specific parent advisory board. To identify methods to enhance care, the nurse practitioners and medical director met weekly to discuss any parent complaints and scrutinize the care provided before all ED visits and hospitalizations.

Process Measures

The staff recorded all clinic visits and telephone calls for all patients given comprehensive care and asked the parents at each visit about any other services received. Every 3 months, patients receiving usual care were called to identify any ED visits, hospital admissions, and outpatient visits and were compensated with a $10 gift card for each query. Efforts to identify all outpatient visits for usual care patients were abandoned because we found that outpatient visits were not reliably identified by parental queries at 3-month intervals. However, Medicaid billings were used to compare outpatient services for patients in the 2 treatment groups who were insured at enrollment by Medicaid only. Parental ratings of outpatient care were obtained by research personnel uninvolved in the care. These personnel administered the Consumer Assessment of Healthcare Providers and Systems Child 12-Month Survey[21-24] in Spanish or English to each mother 12 months after enrollment (6-12 months after enrollment for patients enrolled during the last study year). We preselected 5 questions as being the most important toward optimizing patient outcomes.

Outcome Measures

The primary outcome was to assess whether an enhanced medical home providing comprehensive care reduces the number of children with serious illness (death, ICU admission, or hospital stay >7 days between enrollment and the trial's completion, which was 6 months after the last child was enrolled), reduces costs among such children, or both. As specified prior to starting the trial, the value of the program was considered to be established if the total number of children with a serious illness was reduced without increasing total clinic and hospital costs, these costs were reduced without increasing the total number of children with a serious illness, or both were reduced. The number needed to treat to prevent 1 child with serious illness was calculated by the inverse of the risk difference between treatment groups.

Secondary outcomes included individual components of serious illness, clinic and hospital costs, and total number of serious illnesses, ED visits, hospitalizations, ICU admissions, hospital days, and days in the ICU. To identify ED visits and hospitalizations, we reviewed the ED and hospital logs each weekday for Memorial Hermann Children's Hospital, a private institution that serves as the UTH primary teaching hospital. In addition, we assessed claims data from the 12 Memorial Hermann Health System hospitals and obtained billing records for all Medicaid patients from personnel at Texas Health and Human Services who were blinded to treatment group. Any discrepancies between these data and the data collected from parental queries were identified and resolved.

Economic Evaluations

A health care economist (E.B.C.A.) assessed total hospital and clinic costs from a health care system perspective. Hospital costs (including costs for patients admitted with an observational status) were estimated by multiplying charges by department-specific cost-to-charge ratios specified in the hospital's annual Medicare cost report. Clinic costs for comprehensive care were estimated using total expenditures to include costs for start-up, longer patient visits, extra services, and low patient-to-staff ratios not addressed by relative value units (eAppendix 3 in Supplement 1). We did not have access to pharmacy costs and did not attempt to compare costs for some services (eg, speech therapy and home services) that were largely not under our control and for which Medicaid coverage was rapidly changing and varied greatly between different health care payers.

Clinic costs for usual care were estimated as commonly done using relative value units.[25,26] Because some services are not captured using relative value units and because clinic visits were less fully identified with usual care than comprehensive care, the analyses provide a conservative estimate of the reduction in costs with comprehensive care. Costs were inflated to 2014 US dollars based on the consumer price index for medical services.[27] Cost differences between treatment groups were assessed using generalized linear models with log-link and γ distribution, adjusting for maternal education (receipt of high school diploma [yes or no]), estimated baseline hospitalization risk (high or very high), length of follow-up, and within-family correlation.

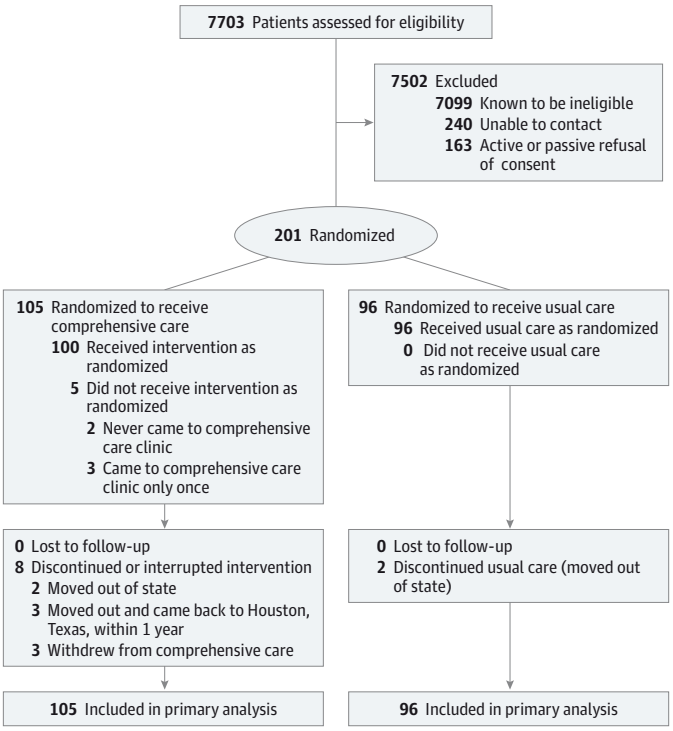
We also assessed the joint influence of comprehensive care on both cost and effectiveness using the net monetary benefit approach.[28] Using this approach, the treatment benefits are expressed in monetary terms based on the willingness to pay per unit of benefit. Even though the minimum willingness to pay to prevent 1 child from developing a serious illness is unknown, we assumed that the true value would certainly be greater than 0. Bootstrap regression analyses based on generalized linear models were used in assessing incremental net monetary benefit and the probability that comprehensive care reduced the number of children with serious illnesses, costs, or both.

We performed secondary analyses from the medical school perspective evaluating the effect of comprehensive care relative to usual care on the difference between expenses and total payments for clinic and hospital care (eAppendix 4 and eTable 1 in Supplement 1). Neither research costs nor grant support were included for the purpose of developing sustainable payment models. In addition, we assessed the effect of comprehensive care on Medicaid payments for hospital and clinic services for these high-risk children (eAppendix 5 and eTable 2 in Supplement 1).

Statistical Analyses and Stopping Rules

Intent-to-treat analyses were performed using Poisson regression models (for the proportion of children with an outcome) with robust standard errors[29] or negative binomial regression models (for total number of secondary outcomes) to estimate rate ratios (RRs). Models were adjusted for maternal educa-

Figure. Treatment Assignment for High-Risk Children With Chronic Illness



For the 7099 patients known to be ineligible for the study, the specific reason for ineligibility for each patient was not recorded. For the most part (an estimated 95%), the patients were either found not to have a chronic illness or they had a chronic illness but did not meet the criteria for high risk (ie, ≥3 emergency department visits, ≥2 hospitalizations, or ≥1 pediatric intensive care unit admission).

tion, length of follow-up, estimated baseline risk, and within-family correlation. All reported RRs were adjusted for these 4 variables. Interactions of treatment group with education, estimated hospitalization risk, and race/ethnicity were assessed for serious illness and total costs. A secondary analysis of serious illness included additional baseline variables of patient age, sex, race/ethnicity, prematurity (yes or no), and Medicaid insurance (yes or no).

A 2-sided $P < .05$ was considered statistically significant. Separate Bayesian analyses were performed to estimate the probability of reduced serious illnesses and the probability of reduced costs from comprehensive care. We used a neutral prior probability (risk ratio = 1.0 [95% credible interval, 0.5-2.0], which encompasses the largest likely effect size for major outcomes observed in randomized trials).

We planned to enroll 400 patients to identify a one-third reduction in total patients who developed serious illness (error of $\alpha = .05$; power level of 80%; and projected serious illness rate of 38% with usual care based on the first year of the trial). Under predefined stopping rules, enrollment would cease whenever Bayesian analyses performed annually from the end of the second year identified a 95% or greater probability that comprehensive care resulted in any of the following: reduced number of children with serious illness without an increase in costs, reduced costs without an increase in number of children with serious illness, or reduction in both. University or grant support would then be used to temporarily continue the program while pursuing a sustainable payment mechanism through Texas Health and Human Services.

All analyses were performed using Stata software version 11.2 (StataCorp) and R software version 3.1.1 (R Foundation for Statistical Computing).

Results

In accordance with the predefined stopping rules, enrollment was stopped at the end of year 2 by the principal investigator (J.E.T.) when the statistician (C.P.) reported that Bayesian analyses identified a 99% probability of reduced serious illness and 98% probability of decreased total clinic and hospital costs with comprehensive care. All patients were followed up for an additional 6 months until August 31, 2013, to assess treatment effects. A total of 183 child-years of comprehensive care and 172 child-years of usual care were provided. The median length of follow-up per child was 1.83 years (interquartile range, 1.41-2.29 years) for comprehensive care and 1.95 years (interquartile range, 1.43-2.31 years) for usual care.

We identified 364 eligible children; 163 (45%) did not consent to participate. Of the 201 children enrolled, 105 were randomized to comprehensive care and 96 to usual care (**Figure**). The groups were at similarly high baseline risk (**Table 1**), and 91% to 92% of patients in each group were insured by Medicaid.

Of the children randomized to usual care, 42% received their primary care from a private pediatrician and 58% from faculty-supervised residents; 27% also received care in the twice-weekly clinic for children with special health care needs. Of the children randomized to comprehensive care, 98% ini-

Table 1. Baseline Characteristics by Treatment Group

	Type of Care, No. (%)	
	Comprehensive (n = 105)	Usual (n = 96)
Age ranges		
0-12 mo	22 (21)	20 (21)
13 mo-2 y	29 (28)	27 (28)
3-5 y	22 (21)	22 (23)
6-11 y	26 (25)	18 (19)
12-18 y	6 (6)	9 (9)
Male sex	65 (62)	56 (58)
Race/ethnicity		
Black	45 (43)	33 (34)
White	10 (10)	11 (11)
Hispanic	50 (48)	52 (54)
No. of unique families[a]	96	95
Medicaid insurance	97 (92)	87 (91)
Risk stratum by maternal receipt of high school diploma		
High[b]		
Yes	67 (64)	63 (66)
No	25 (24)	20 (21)
Very high[b]		
Yes	11 (10)	12 (12)
No	2 (2)	1 (1)
Congenital disorders	38 (36)	31 (32)
Trisomy 21	5 (5)	4 (4)
Cystic fibrosis	4 (4)	4 (4)
Spina bifida	7 (7)	3 (3)
Other congenital[c]	6 (6)	7 (7)
Prematurity	41 (39)	42 (44)
Respiratory disorders	85 (81)	75 (78)
Asthma	43 (41)	31 (32)
Other chronic lung disorders[d]	46 (44)	49 (51)
Neurological disorders	40 (38)	36 (38)
Seizure disorder	19 (18)	22 (23)
Severe neurological impairment	25 (24)	13 (14)
Other neurological disorders[e]	7 (7)	6 (6)
Ventriculoperitoneal shunt	10 (10)	10 (10)
Gastrointestinal disorders	36 (34)	26 (27)
Dysphagia/swallowing disorders	33 (31)	24 (25)
Other gastrointestinal disorders	3 (3)	2 (2)
Colostomy	1 (1)	1 (1)
Disorders of other organs	26 (25)	22 (23)
Cardiac[f]	5 (5)	2 (2)
Musculoskeletal[g]	8 (8)	2 (2)
Immunological[h]	4 (4)	3 (3)
Ear, nose, and throat[i]	15 (14)	16 (17)
Treatments		
Mechanical ventilation	12 (11)	10 (10)
Gastrostomy tube	33 (31)	24 (25)

[a] With the exception of 1 family in comprehensive care that had 3 siblings enrolled, all other families with more than 1 child enrolled had 2 siblings.

[b] Estimated baseline risk of hospitalization during next year was based on the clinical judgment of the medical director.

[c] CHARGE syndrome (coloboma, heart defect, atresia choanal, retarded growth and development, genital hypoplasia, ear anomalies or deafness), VACTERL association (vertebral anomalies, anal atresia, cardiac defects, tracheoesophageal fistula and/or esophageal atresia, renal and radial anomalies, and limb defects), DiGeorge syndrome, acampomelic campomelic syndrome, Pierre Robin syndrome, Cornelia de Lange syndrome, or Prader-Willi syndrome.

[d] Bronchopulmonary dysplasia, cystic fibrosis, congenital lung disease, or chronic respiratory failure secondary to neurological impairment.

[e] Spinal muscular atrophy, progressive ataxia, or Gaucher disease.

[f] After repair, tetralogy of Fallot, ventricular septal defect, or atrial septal defect.

[g] Achondroplasia, significant limb deformity, arthrogryposis, clubfoot, severe scoliosis, or acampomelic compomelic dysplasia.

[h] Anaphylaxis, immunodeficiency related to CHARGE syndrome, or severe eczema.

[i] Tracheostomy or cleft palate.

tially attended our high-risk children's clinic; 88% attended the clinic throughout the trial (5% moved away from Houston, Texas). Comprehensive care patients had a mean of 12.9 telephone calls and 12.3 clinic visits per child-year. Medicaid patients (for whom total office visits could be tabulated in both groups) had a mean of 3.2 (95% CI, 0.9-5.4) more clinic visits per child-year with comprehensive care (12.1 visits) vs usual care (9.4 visits) (P = .007). Comprehensive care also was associated with systematically higher maternal ratings of care (**Table 2**).

Outcomes

Comprehensive care reduced the number and rate of children with a serious illness (10 per 100 child-years vs 22 treated with usual care) (RR, 0.45 [95% CI, 0.28-0.73]; number needed to treat = 4.5) while also decreasing the total hospital and clinic costs ($16 523 vs $26 781 per child-year, respectively) (cost ratio, 0.58 [95% CI, 0.38-0.88]; **Table 3** and **Table 4**). Using a conservative willingness-to-pay threshold of $0, the incremental net monetary benefit of comprehensive care relative to usual care was $10 734 (95% CI, $2069-$27 694) to prevent 1 child from developing a serious illness (Table 4). The probability that comprehensive care was cost neutral or cost saving at this threshold was 97%.

Comprehensive care did not significantly reduce deaths (2 vs 3 treated with usual care per 100 child-years; P = .40) but did substantially reduce the number and rate of children who had 1 or 2 components of serious illness (Table 3). Rates were also reduced with comprehensive care vs usual care for total serious illnesses (16 vs 44 per 100 child-years, respectively) (RR, 0.33; 95% CI, 0.17-0.66), ED visits (90 vs 190 per 100 child-years) (RR, 0.48; 95% CI, 0.34-0.67), hospitalizations (69 vs 131 per 100 child-years) (RR, 0.51; 95% CI, 0.33-0.77), number of days in the hospital (276 vs 635 per 100 child-years) (RR, 0.36; 95% CI, 0.19-0.67), ICU admissions (9 vs 26 per 100 child-years) (RR, 0.35; 95% CI, 0.18-0.70), and days in the ICU (28 vs

Table 2. Clinic Visits and Parental Ratings of Care by Treatment Group

Process Measures	No./Total (%)[a] Comprehensive Care	Usual Care	Difference (95% CI), %[a]	*P* Value
Clinic visits among patients with only Medicaid insurance at enrollment, mean (SD) per child-year	12.1 (9.0)[b]	9.4 (7.4)[c]	3.2 (0.9-5.4)[d]	.007[d,e]
Maternal responses to survey[f]				
Child always got appointment for care as soon as needed[g]	63/67 (94)	23/47 (49)	45 (30-60)	<.001
Clinician always listened carefully	82/84 (98)	61/83 (73)	24 (14-34)	<.001
Clinician always knew important information about child's medical history	77/82 (94)	57/83 (69)	25 (14-36)	<.001
Clinician always spent enough time with child	78/84 (93)	56/83 (67)	25 (14-37)	<.001
Clinician rating of 9 or 10[h]	78/84 (93)	49/83 (59)	34 (22-46)	<.001

[a] Unless otherwise indicated.

[b] There were 90 patients and 158 child-years.

[c] There were 79 patients and 144 child-years.

[d] Adjusted for maternal education, risk stratum, length of follow-up, and within-family correlation.

[e] Obtained from a negative binomial regression model adjusting for maternal education, estimated hospitalization risk, length of follow-up, and within-family correlation.

[f] The Consumer Assessment of Healthcare Providers and Systems Child 12-Month Survey[21-24] was administered to each mother 12 months after enrollment (6-12 months after enrollment for patients enrolled during the last study year). There were a total of 96 possible survey respondents in the comprehensive care group and 95 in the usual care group. The *P* values were obtained from the 2-sample *z* test for proportions.

[g] This question only applied if parents sought an appointment for their children.

[h] On a scale of 0 to 10 with 10 representing the best clinician possible.

Table 3. Outcome Measures by Treatment Group

Outcome Measure	Comprehensive Care (n = 105)[a] No.	Rate/100 Child-Years (95% CI)	Usual Care (n = 96)[b] No.	Rate/100 Child-Years (95% CI)	Rate Ratio (95% CI)[c]	*P* Value
Children with a serious illness[d]	18	10 (6-16)	38	22 (16-30)	0.45 (0.28-0.73)	.001[e]
Deaths	3	2 (0-5)	5	3 (1-7)	0.52 (0.12-2.19)	.37[e]
Children with a pediatric ICU admission	14	8 (4-13)	26	15 (10-22)	0.52 (0.29-0.93)	.03[e]
Children with a hospital stay >7 d	13	7 (4-12)	28	16 (11-23)	0.44 (0.26-0.77)	.004[e]
Children who died or received care at pediatric ICU	15	8 (5-13)	28	16 (11-23)	0.53 (0.31-0.92)	.03[e]
Serious illnesses	29	16 (11-23)	76	44 (35-55)	0.33 (0.17-0.66)	.001[f]
Emergency department visits	165	90 (77-105)	328	190 (170-212)	0.48 (0.34-0.67)	<.001[f]
Hospitalizations	127	69 (58-82)	226	131 (115-149)	0.51 (0.33-0.77)	.001[f]
Days in a hospital	506	276 (252-301)	1094	635 (598-673)	0.36 (0.19-0.67)	.001[f]
Pediatric ICU admissions	17	9 (5-15)	44	26 (19-34)	0.35 (0.18-0.70)	.003[f]
Days in a pediatric ICU	52	28 (21-37)	178	103 (89-120)	0.31 (0.11-0.87)	.03[f]

Abbreviation: ICU, intensive care unit.

[a] There were 183.5 child-years.

[b] There were 172.4 child-years.

[c] Adjusted for maternal education, risk stratum, length of follow-up, and within-family correlation.

[d] Serious illness was the primary outcome and included death, pediatric ICU admission, or hospitalization for longer than 7 days (composite outcome).

[e] Obtained from Poisson regression models adjusting for maternal education, estimated hospitalization risk, length of follow-up, and within-family correlation.

[f] Obtained from negative binomial regression models adjusting for maternal education, estimated hospitalization risk, length of follow-up, and within-family correlation.

103 per 100 child-years) (RR, 0.31; 95% CI, 0.11-0.87). The probability that comprehensive care improved each outcome except death ranged from 94% to 99%.

Secondary analysis of the proportion of children with a serious illness adjusting for additional baseline variables gave similar results (RR, 0.43; 95% CI, 0.27-0.69). In subgroup analyses comparing the rate of children with a serious illness, the RR for those with a high estimated baseline risk of hospitalization (RR, 0.33; 95% CI, 0.17-0.64) was lower than in the very-high-risk stratum (RR, 0.84; 95% CI, 0.44-1.61) (*P* = .049 for interaction). However, the number needed to treat to prevent 1 child from having a serious illness was low (4.4-6.5) in both strata. No other interactions were significant for serious illness or costs.

Secondary Economic Analyses

The total estimated costs per child-year were considerably lower with comprehensive care than with usual care due to a large reduction in hospital costs ($9810 vs $25 059, respectively) (cost ratio, 0.35; 95% CI, 0.21-0.58) that exceeded the increase in clinic costs ($6713 vs $1722) (cost ratio, 3.96 [95% CI, 3.12-5.01]; Table 4). Total Medicaid payments were sub-

Table 4. Estimated Costs per Child-Year

	Costs per Child-Year, Mean (95% CI), $[a]		Cost Ratio (95% CI)[d]	*P* Value[e]
	Comprehensive Care[b]	Usual Care[c]		
During trial				
Clinic	6713 (5616-8021)	1722 (1429-2075)	3.96 (3.12-5.01)	<.001
Hospital	9810 (7056-13 632)	25 059 (17 768-35 379)	0.35 (0.21-0.58)	<.001
Total	16 523 (12 526-21 789)	26 781 (20 061-35 787)	0.58 (0.38-0.88)	.01
During start-up period[f]				
Clinic	12 158 (9903-14 965)	2336 (1901-2865)	5.98 (4.82-7.42)	<.001
Hospital	11 413 (9195-23 257)	27 552 (21 324-52 296)	0.45 (0.26-0.78)	.005
Total	23 571 (16 066-34 639)	29 888 (20 390-43 730)	0.89 (0.57-1.38)	.59
After first year of enrollment				
Clinic	5124 (4101-6415)	1513 (1198-1913)	3.37 (2.42-4.69)	<.001
Hospital	9343 (6313-13 868)	24 213 (16 061-36 576)	0.34 (0.18-0.63)	.001
Total	14 467 (10 356-20 265)	25 726 (18 128-36 265)	0.54 (0.33-0.89)	.02

[a] The estimated costs per child-year were inflated to 2014 US dollars.[27] The incremental net monetary benefit was $10 734 ($2069-$27 694)[d] using a threshold willingness to pay of $0 US to prevent 1 child from developing a serious illness for comprehensive care vs usual care (*P* = .03). The results of the bootstrap regression analyses (using 1000 bootstrap replicates) were similar when we fitted a normal distribution or a γ distribution with a log link.

[b] There were 183.5 child-years.

[c] There were 172.4 child-years.

[d] Adjusted for maternal education, risk stratum, length of follow-up, and within-family correlation.

[e] Obtained from the generalized linear models with log-link and γ distribution adjusting for maternal education, estimated hospitalization risk, length of follow-up, and within-family correlation.

[f] Through first year of enrollment.

stantially less than the estimated health system costs both for comprehensive care ($9287 vs $16 523) and usual care ($15 529 vs $26 781). From a medical school perspective, comprehensive care resulted in a deficit (excess of costs over revenues) of $6018 (95% CI, $5506-$6629) per child-year ($4419 [95% CI, $4247-$4771] per child-year excluding start-up costs through the first year), a deficit of $4759 (95% CI, $3057-$6031) greater than with usual care (eAppendix 4 in Supplement 1). Medicaid payments were $6243 (95% CI, $1302-$11 678) lower per child-year with comprehensive care than usual care (eAppendix 5 in Supplement 1).

Discussion

In this randomized clinical trial, the triple aim of improved care, improved health, and lower costs was achieved in an enhanced medical home providing comprehensive care to high-risk children with chronic illness compared with usual care. Access to care and parent satisfaction were substantially increased, the number of high-risk children with a serious illness was decreased by 55%, and total clinic and hospital costs (assessed from a health system perspective) were reduced by $10 258 per child-year. Partly because we did not include savings to parents and because costs could be more completely assessed for comprehensive care than usual care, this analysis provides a conservative estimate of the total savings.

The failure to demonstrate improved outcomes or reduced costs in prior studies[4,30-35] recently prompted Schwenk[6] to reject the patient-centered medical home as a generic approach to health care delivery and to recommend evaluation of strategies to maximize their value for high-risk populations. The benefits and cost savings we identified with comprehensive care seem likely to be achievable only in high-risk populations treated in major academic centers with the subspecialists, resources, and clinician commitment to provide such care.

The limitations of the trial include an inability to obtain pharmacy costs. However, with the intervention's effort to avoid overuse and misuse of medications in providing evidence-based care, pharmacy costs may have been comparable or lower with comprehensive care than usual care. The most important limitation is the uncertain generalizability to other centers and clinicians. Our trial involved a single center, relatively few clinicians, and a patient population and methods of care that may differ from other centers. The consent rate for eligible children (55%) was reasonable for a challenging pediatric trial; however, parents who were relatively unhappy with their child's prior care may have been more inclined to consent, a factor likely to contribute to the large benefits and favorable parental ratings of comprehensive care.[36] Nevertheless, the findings are similar to those in a prior trial of comprehensive care at a different center in which 887 high-risk infants were randomized to comprehensive or usual care from the same clinicians after discharge from a neonatal ICU.[16] The RR for the outcome of life-threatening illness (death or pediatric ICU admission) was virtually identical in both trials (0.52-0.53). The higher absolute cost savings in our trial reflects both a higher-risk population and inflation.

Together, these 2 trials provide the strongest evidence supporting medical homes in any age group.[3,4,6] We believe these results reflect a combination of factors: (1) a high-risk population with high health care costs[30]; (2) experienced clinicians knowledgeable about each patient and available at all hours; (3) expert subspecialty care available at the same or at

a nearby clinic in the same facility; (4) the high priority given to minimizing unnecessary ED visits and hospitalizations and the intensive review of the care provided before every ED visit and hospitalization; (5) the identification each weekday of all children having ED visits and hospitalizations to ensure prompt follow-up and coordination of care[37]; and (6) a relatively low patient-to-staff ratio with patient-to-nurse practitioner ratio no greater than 50 in our trial and 75 in the prior trial (involving somewhat lower-risk patients) to allow for longer clinic visits, more telephone calls and e-mails, extensive quality improvement measures, and frequent nights and weekends on call. Whether similar benefits and cost savings can be achieved with higher patient-to-staff ratios remains to be demonstrated.

Analyses from a medical school perspective indicated that comprehensive care was associated with a deficit of $6018 of costs over revenues per child-year ($4419 per child-year excluding start-up costs through the first year). Financial effects would differ in different centers. However, limited payments reduce access to care in many locales, particularly for children with chronic illness[8] in largely Medicaid populations.[38] Inadequate payments appear to be the primary reason that few neonatal follow-up clinics provide comprehensive care to high-risk infants. Likewise medical schools or other institutions that have the expertise to provide comprehensive care to high-risk children with chronic illness are unlikely to implement programs that each year result in losses of thousands of dollars per child enrolled. In Texas, comprehensive care could be sustained without increasing Medicaid expenditures by providing the savings ($6243 per child year) as capitation, shared savings, or other support to the medical school.

Conclusions

Among high-risk children with chronic illness, an enhanced medical home that provided comprehensive care to promote prompt effective care vs usual care reduced serious illnesses and costs. These findings from a single site of selected patients with a limited number of clinicians require study in larger, broader populations before conclusions about generalizability to other settings can be reached.

ARTICLE INFORMATION

Author Contributions: Dr Mosquera had full access to all of the data in the study and takes responsibility for the integrity of the data and the accuracy of the data analysis.
Study concept and design: Mosquera, Avritscher, Pedroza, Evans, Pacheco, Clifton, Zupancic, Tyson.
Acquisition, analysis, or interpretation of data: Mosquera, Avritscher, Samuels, Harris, Pedroza, Navarro, Wootton, Moody, Franzini, Tyson.
Drafting of the manuscript: Mosquera, Avritscher, Samuels, Harris, Pedroza, Pacheco, Tyson.
Critical revision of the manuscript for important intellectual content: Mosquera, Avritscher, Samuels, Pedroza, Evans, Navarro, Wootton, Clifton, Moody, Franzini, Zupancic, Tyson.
Statistical analysis: Mosquera, Avritscher, Pedroza, Franzini, Tyson.
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Administrative, technical, or material support: Mosquera, Samuels, Navarro, Wootton, Pacheco, Tyson.
Study supervision: Mosquera, Navarro, Moody, Franzini, Tyson.

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REFERENCES

1. Burke R, Liptak GS; Council on Children with Disabilities. Providing a primary care medical home for children and youth with spina bifida. *Pediatrics*. 2011;128(6):e1645-e1657.

2. Cohen E, Jovcevska V, Kuo DZ, Mahant S. Hospital-based comprehensive care programs for children with special health care needs: a systematic review. *Arch Pediatr Adolesc Med*. 2011;165(6):554-561.

3. Homer CJ, Klatka K, Romm D, et al. A review of the evidence for the medical home for children with special health care needs. *Pediatrics*. 2008;122(4):e922-e937.

4. Jackson GL, Powers BJ, Chatterjee R, et al. Improving patient care: the patient centered medical home: a systematic review. *Ann Intern Med*. 2013;158(3):169-178.

5. Liptak GS, Murphy NA; Council on Children With Disabilities. Providing a primary care medical home for children and youth with cerebral palsy. *Pediatrics*. 2011;128(5):e1321-e1329.

6. Schwenk TL. The patient-centered medical home: one size does not fit all. *JAMA*. 2014;311(8):802-803.

7. Agrawal R, Shah P, Zebracki K, Sanabria K, Kohrman C, Kohrman AF. Barriers to care for children and youth with special health care needs: perceptions of Illinois pediatricians. *Clin Pediatr (Phila)*. 2012;51(1):39-45.

8. Bitsko RH, Visser SN, Schieve LA, Ross DS, Thurman DJ, Perou R. Unmet health care needs among CSHCN with neurologic conditions. *Pediatrics*. 2009;124(suppl 4):S343-S351.

9. Carnevale FA, Alexander E, Davis M, Rennick J, Troini R. Daily living with distress and enrichment: the moral experience of families with ventilator-assisted children at home. *Pediatrics*. 2006;117(1):e48-e60.

10. Greenberg JO, Barnett ML, Spinks MA, Dudley JC, Frolkis JP. The "medical neighborhood": integrating primary and specialty care for ambulatory patients. *JAMA Intern Med*. 2014;174(3):454-457.

11. Kuo DZ, Cohen E, Agrawal R, Berry JG, Casey PH. A national profile of caregiver challenges among more medically complex children with

special health care needs. *Arch Pediatr Adolesc Med*. 2011;165(11):1020-1026.

12. Simon TD, Berry J, Feudtner C, et al. Children with complex chronic conditions in inpatient hospital settings in the United States. *Pediatrics*. 2010;126(4):647-655.

13. Zickafoose JS, Clark SJ, Sakshaug JW, Chen LM, Hollingsworth JM. Readiness of primary care practices for medical home certification. *Pediatrics*. 2013;131(3):473-482.

14. Mokkink LB, van der Lee JH, Grootenhuis MA, Offringa M, Heymans HS; Dutch National Consensus Committee Chronic Diseases and Health Conditions in Childhood. Defining chronic diseases and health conditions in childhood (0-18 years of age): national consensus in the Netherlands. *Eur J Pediatr*. 2008;167(12):1441-1447.

15. Medical Home Initiatives for Children With Special Needs Project Advisory Committee; American Academy of Pediatrics. The medical home. *Pediatrics*. 2002;110(1 pt 1):184-186.

16. Broyles RS, Tyson JE, Heyne ET, et al. Comprehensive follow-up care and life-threatening illnesses among high-risk infants: a randomized controlled trial. *JAMA*. 2000;284(16):2070-2076.

17. Faden RR, Beauchamp TL, Kass NE. Informed consent, comparative effectiveness, and learning health care. *N Engl J Med*. 2014;370(8):766-768.

18. Faden RR, Kass NE, Goodman SN, Pronovost P, Tunis S, Beauchamp TL. An ethics framework for a learning health care system: a departure from traditional research ethics and clinical ethics. *Hastings Cent Rep*. 2013;(Spec No):S16-S27.

19. Kass NE, Faden RR, Goodman SN, Pronovost P, Tunis S, Beauchamp TL. The research-treatment distinction: a problematic approach for determining which activities should have ethical oversight. *Hastings Cent Rep*. 2013;(Spec No):S4-S15.

20. National Committee for Quality Assurance (NCQA). Patient-centered medical home recognition. http://www.ncqa.org/Programs/Recognition/PatientCenteredMedicalHomePCMH.aspx. Accessed March 13, 2014.

21. US Department of Health and Human Services; Agency for Healthcare Research and Quality. CAPHS: surveys and tools to advance patient-centered care. http://www.CAHPS.AHRQ.gov. Accessed October 12, 2013.

22. Carle AC, Weech-Maldonado R. Does the Consumer Assessment of Healthcare Providers and Systems Cultural Competence Survey provide equivalent measurement across English and Spanish versions? *Med Care*. 2012;50(9)(suppl 2):S37-S41.

23. Gallagher P, Ding L, Ham HP, Schor EL, Hays RD, Cleary PD. Development of a new patient-based measure of pediatric ambulatory care. *Pediatrics*. 2009;124(5):1348-1354.

24. Tom JO, Mangione-Smith R, Solomon C, Grossman DC. Integrated personal health record use: association with parent-reported care experiences. *Pediatrics*. 2012;130(1):e183-e190.

25. Gerstle RS, Molteni RA, Andreae MC, et al; Committee on Coding and Nomenclature. Application of the resource-based relative value scale system to pediatrics. *Pediatrics*. 2014;133(6):1158-1162.

26. Gold MR, Weinstein MC. *Cost Effectiveness in Health and Medicine*. New York, NY: Oxford University Press; 1996.

27. US Department of Labor; Bureau of Labor Statistics. Consumer price indexes. http://www.bls.gov/cpi/home.htm. Accessed February 21, 2014.

28. Stinnett AA, Mullahy J. Net health benefits: a new framework for the analysis of uncertainty in cost-effectiveness analysis. *Med Decis Making*. 1998;18(2)(suppl):S68-S80.

29. Zou G. A modified poisson regression approach to prospective studies with binary data. *Am J Epidemiol*. 2004;159(7):702-706.

30. Friedberg MW, Schneider EC, Rosenthal MB, Volpp KG, Werner RM. Association between participation in a multipayer medical home intervention and changes in quality, utilization, and costs of care. *JAMA*. 2014;311(8):815-825.

31. Jaén CRFR, Ferrer RL, Miller WL, et al. Patient outcomes at 26 months in the patient-centered medical home National Demonstration Project. *Ann Fam Med*. 2010;8(suppl 1):S57-S67, S92.

32. Hoffman A, Emanuel EJ. Reengineering US health care. *JAMA*. 2013;309(7):661-662.

33. Rosenthal MB, Friedberg MW, Singer SJ, Eastman D, Li Z, Schneider EC. Effect of a multipayer patient-centered medical home on health care utilization and quality: the Rhode Island chronic care sustainability initiative pilot program. *JAMA Intern Med*. 2013;173(20):1907-1913.

34. Werner RM, Duggan M, Duey K, Zhu J, Stuart EA. The patient-centered medical home: an evaluation of a single private payer demonstration in New Jersey. *Med Care*. 2013;51(6):487-493.

35. Paustian ML, Alexander JA, El Reda DK, Wise CG, Green LA, Fetters MD. Partial and incremental PCMH practice transformation: implications for quality and costs. *Health Serv Res*. 2014;49(1):52-74.

36. US Department of Health and Human Services; Agency for Healthcare Research and Quality. CAHPS database. https://www.cahpsdatabase.ahrq.gov/CAHPSIDB/Public/CG/CG_About.aspx. Accessed March 13, 2014.

37. Hesselink G, Schoonhoven L, Barach P, et al. Improving patient handovers from hospital to primary care: a systematic review. *Ann Intern Med*. 2012;157(6):417-428.

38. Berman S, Dolins J, Tang SF, Yudkowsky B. Factors that influence the willingness of private primary care pediatricians to accept more Medicaid patients. *Pediatrics*. 2002;110(2 pt 1):239-248.