The Cost of Congenital Heart Disease in Children and Adults

A Model for Multicenter Assessment of Price and Practice Variation

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Objective: To assess the cost of congenital heart disease (CHD) and to assess whether practice pattern or price was more responsible for variation.

Research Design and Setting: Data were collected from Charleston, NC; Columbus, Ohio; Detroit, Mich; Houston, Tex; Los Angeles, Calif; and New York, NY. The CHD was first classified as to physiologic characteristics and severity. For each type of CHD, the number of clinic visits, hospitalizations, and years of medication use were estimated.

Results: On the basis of actual charges, the "prices" were calculated as follows, in 1992 dollars: for patients from birth to 21 years: benign disease (19% of patients), $3940; acyanotic disease (45%), $49 730; cyanotic disease (36%), $102 084; and average for all CHD categories, $59 877; for patients 22 to 40 years of age (of whom 24% had resolved defects or were dead): benign disease (19%), $3470; acyanotic disease (52%), $12 981; cyanotic disease (29%), $39 187; and average for all CHD, $18 773. The cost for the group from birth to 21 years varied from $47 500 to $73 600, accounting for 55% by practice (number of echocardiograms and cardiac catheterizations) and 45% by price, although mortality was similar.

Conclusions: The treatment of CHD is comparatively inexpensive, especially in adult survivors. The variation in both practice and price bears further study, with comparisons to determine the most cost-effective strategies for treating these patients.

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THE POPULATION of children and adults with chronic disease has increased markedly in the last decade because of improved survival.[1-3] It has been estimated that 6% of children have a chronic illness (approximately 3.6 million in the United States).[4] If this population were concentrated in one city, it would be the fourth largest city in the United States. As this group grows, concerns are appropriately directed at the cost of their care for the following reasons: (1) to budget appropriately for current and future programs to pay for their health care, (2) to use these statistics as a baseline for assessing future cost-benefit ratios for certain forms of treatment, and (3) to assess the investment by the health care system in these children so that ways are found to optimize that investment when the children become adults.

A major focus of cost analysis is to attempt to reduce present and future costs by addressing the variation in pricing as well as the variation in the practice of medicine. For example, if two institutions have similar outcomes but one institution performs twice as many tests, this should provide an objective basis for determination of an appropriate level of testing to be reimbursed.

Therefore, in this study, we sought to assess both the cost and the variation in pricing and practice for a chronic disease among six geographically representative centers in the United States. Congenital heart disease was chosen as a

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PATIENTS AND METHODS

CLASSIFICATION OF CONGENITAL HEART DISEASE

Congenital heart disease was categorized by functional criteria rather than by single anatomic diagnosis. Six categories for the ages of birth to 21 years were used: (1) benign (no anticipated need for cardiac catheterization or surgery) (eg, small ventricular septal defect); (2) acyanotic congenital heart disease with infant surgery, which required the patient to undergo surgery within the first year of life (eg, complete atrioventricular septal defect); (3) acyanotic with later surgery (beyond 1 year of age) (eg, simple coarctation of the aorta); (4) acyanotic with intervention, in which a catheter was used to treat congenital heart disease (eg, valvular pulmonary stenosis); (5) cyanotic, without the Fontan operation (eg, tetralogy of Fallot); and (6) cyanotic, with the Fontan operation (eg, tricuspid atresia, with a procedure to direct systemic venous inflow to the lungs).

Two age groups were examined: birth to 21 years (corresponding to the duration of the majority of state funding programs for children) and 22 to 40 years. The upper age limit of 40 years was chosen to estimate the onset of superimposed diseases, such as coronary artery disease. For the ages of 22 to 40 years, the following five categories were used: (1) benign; (2) postoperative, acyanotic; (3) postoperative, cyanotic (tetralogy of Fallot); (4) postoperative, cyanotic (Mustard's operation [atrial repair for transposition] or Fontan operation); and (5) pulmonary vascular obstructive disease.

SUBCATEGORIES

For each category, subcategories were developed to reflect (1) the timing with respect to intervention and (2) the severity of the patient's course as would be described by the numbers of procedures, hospitalizations, and clinic visits. For example, for the age group birth to 21 years, the cyanotic, non–Fontan operation category (eg, tetralogy of Fallot) had five subcategories. The first subclass incorporated all patients for the first year of life, with the assumption that a typical patient would have a neonatal hospital stay, an operation (either palliation or correction), and a number of clinic visits. However, it was then assumed that after the first year, patients would have different clinical courses depending on their specific lesion and the results

of surgery. Physicians assigned typical clinical courses to these subcategories (eg, "good") for the patients with good results. The lists of categories and subcategories are shown in **Table 1** and **Table 2**.

CATEGORIZED SERVICES

The clinical course of each patient was described objectively by seven services: (1) A routine clinic visit was defined as a physician visit, with an electrocardiogram and a chest roentgenogram. (2) A complex clinic visit was defined as a routine clinic visit plus either an echocardiogram or a 24-hour electrocardiogram (Holter). (3) A medical hospitalization was defined as admission for either diagnostic or medical treatment. (4) A surgical hospitalization was defined as hospitalization for cardiac surgery and its recovery. (5) Interventional hospitalization was one in which the patient underwent cardiac catheterization with an interventional procedure. (6) Pacemaker hospitalization was defined as the implantation or changing of a pacemaker generator and/or pacemaker leads. (7) The final category was the number of years the patient was taking cardiac medication.

SERVICES AND CHARGES FROM SIX CENTERS

Six centers were chosen for their geographic and population variability: Charleston, SC, Columbus, Ohio, Detroit, Mich, Houston, Tex, Los Angeles, Calif, and New York, NY.

The following data were obtained from each center: (1) The investigators were asked to estimate, on the basis of their personal experience in that institution, the percentage of patients who fell into each subcategory. Part of each subcategory assignment also included a mortality for that subclass over the period covered (either birth to 21 years or 22 to 40 years). (2) Each investigator then estimated the number of clinic visits, hospitalizations, and years of medication use for each of the subcategories. For example, if an infant with a small ventricular septal defect was seen twice in the first year of life (one visit with an echocardiogram and one without) and then was followed up every 2 years to the age of 21 years, with every other visit (every fourth year) including an echocardiogram, the total would be six routine clinic visits and six complex clinic visits. These data had to be estimated (rather than obtained through chart review) for two reasons: such detailed data were not available on patients for the last 21 years and the practice of pediatric cardiology has changed markedly during the last 21 years. The inves-

chronic disease that has a relatively high prevalence (almost 1% of all births),[5] that has broadly similar approaches to treatment (ie, the majority of cyanotic infants will require some form of surgery), and that has excellent survival to adulthood (85% to 90%).[1-3] This model could be applicable to other forms of chronic disease in both children and adults as an approach to characterizing the various clinical courses in an objective form and to costing of each outcome.

RESULTS

DISEASE CATEGORIES FOR BIRTH TO 21 YEARS OF AGE

The disease categories, their estimated relative frequencies, and mortality are shown in Table 1. The largest group was the acyanotic category (45%), followed by cyanotic (36%)

tigators were asked to project how they would handle a child in each subclass on the basis of current medical knowledge. (3) Investigators provided data from each of their local institutions on mean charges (physician fee and hospital charge) for each of the types of clinic visit and hospitalization, in 1992 dollars. The medication prices were calculated for a 1-year supply of digoxin and furosemide (liquid or tablet formulation appropriate to the age of the patient).

After the data on practice pattern were collected, the pooled data were presented to each of the investigators in a modified Delphi approach for possible change in the estimates.

STATISTICAL METHODS

Pooled Data

For each institution, a weighted average (by the percentage of patients in each subcategory) was calculated for each of the number of clinic visits, hospitalizations, and years of medication. For example, in the disease category cyanotic, non–Fontan operation, the number of services provided in the first year was added to the weighted average of the four subcategories from 1 to 21 years of age for a total number of services provided between birth and 21 years of age. The input data for that calculation are shown in **Table 3**.

For the age category birth to 21 years, data for each of the six disease categories were averaged among the six institutions. Similar calculations were made for the five disease categories between 22 and 40 years of age. The output data for these calculations are shown in **Table 4**.

For each disease category, the average number (among the six institutions) of each of the services (eg, routine clinic visit) was multiplied by the average charges to obtain total charges by class (**Table 5**).

With the pooled data from the six institutions, the percentage of patients estimated in each of the age groups in the three major types of congenital heart disease (benign, acyanotic, and cyanotic) and the charges estimated for each of the three types were calculated (**Table 6**).

Estimation of Variation in Price and Practice Among the Six Centers

The field of accounting has long dealt with variance in cost resulting from price and volume variances. Our approach was to generalize from the three standard cost accounting relationships described below to accommodate multiple

modalities (volumes), prices, and sites.[6] The purposes of this analysis were to identify (1) the dollar amount of variation in treatment costs of congenital heart disease across six cities, (2) the causes (sources) of variation, and (3) the proportion of total variation in costs that is attributable to each of the sources. Three definitional relationships formed the basis for this approach.

1. Price variance=(price for a service at a center−mean price for that service for six centers)×frequency for that service at that center.

2. Practice pattern variance=(frequency of a service at a center−mean frequency for that service for six centers) ×mean price for that service.

3. Cost variance=price variance+practice pattern variance.

Since an overall course of treatment is composed of multiple medical services, there will also be a "treatment pattern" (a combination of choice and frequency of medical services, ie, a composite of practice patterns) for each city and correspondingly a variation in treatment patterns and in total cost of treatment. The cost of treatment in each city is determined by combining prices of individual medical services in each city with the treatment pattern for a given condition for that city.

If the treatment of congenital heart disease involved only one medical service, then the variation in treatment cost across different cities would result from the variation in the price of that service and the variation in frequency of prescribing that service (variation in practice pattern for that service). The analysis of variation in treatment costs could then be handled by simply applying the three definitional relationships given above. However, since the treatment of congenital heart disease involves more than one medical service, our approach to analyzing cost variation must be generalized to deal with multiple services and multiple prices, and the substantial offsetting effects of variations in practice patterns and prices on variations in total treatment costs.

Expected Lifetime

Expected lifetime was defined as the age by which 50% of subjects would be expected to die. The following estimates of cumulative mortality were used for the normal US population: 1% by age 21 years, 5% by age 40 years, and 50% by age 75 years. The expected lifetime for individuals with congenital heart disease was calculated by adding the normal US population data to the excess associated with congenital heart disease.

and benign heart disease (19%). The overall mortality for the group was 9%, thus giving an estimated 91% survival to age 21 years for patients with congenital heart disease.

DISEASE CATEGORIES
FOR 22 TO 40 YEARS OF AGE

The disease categories and their relative frequencies are shown in Table 2. Also shown is the statistic indicating

patients who were unavailable for follow-up or dead. For the patients with benign disease, this implies unavailability for follow-up, whereas for those with acyanotic or cyanotic congenital heart disease, it implies death before the age of 21 years. Because of the higher death rate in cyanotic patients and the large number of patients with benign heart disease assumed to be unavailable for follow-up, the percentage of acyanotic patients over the age of 21 years is greater than the percentage less than 21 years.

Mortality from 21 to 40 years of age was estimated at 2% for acyanotic patients, 5% for cyanotic patients without the Fontan operation, and 10% for those undergoing the Mustard or Fontan operation.

The expected lifetimes were calculated to be 64.9 years for acyanotic patients, 49.0 years for cyanotic patients, and 57.3 years for all patients.

VOLUME OF SERVICES FOR CLINICAL PRACTICE

The volume of services for birth to 21 years of age is shown in Table 4. The patients who had the Fontan operation used the most services, followed by other cyanotic patients and then acyanotic patients who underwent surgery in infancy. These were the three most intensive diagnoses, followed by the other three in decreasing order: acyanotic with later surgery, acyanotic with intervention, and benign. An example of the wide range of services provided depending on diagnosis is indicated by total number of clinic visits for the period from birth to 21 years: 34.6 in the cyanotic, Fontan operation group; 26.6 in the cyanotic, non–Fontan operation group; 22.1 in the acyanotic, infant surgery group; 14.8 in the acyanotic, later surgery group; 11.9 in the acyanotic, intervention group; and 8.3 in the benign group.

CHARGES

Mean charges are shown in Table 4. The charges for a complex clinic visit (including an echocardiogram or 24-hour Holter monitoring) were 4.1 times the cost of a routine clinic visit. The charges for surgical hospitalization were 3.25 times the charges for an interventional hospitalization.

Total Charges by Category

The total charges from birth to 21 years of age are shown in Table 5. Surgical hospitalization accounted for the largest amount of the charges; the two next highest charges were complex clinic visits and medication.

Charges by Lesion

The overall charges estimated by type of lesion are shown in Table 6. Of the charges that occurred between birth and 40 years of age, 74% occurred in the first 21 years of life. Of the overall charges, patients with cyanotic congenital heart disease accounted for 67% (although they only composed 36% of the patients), patients with acyanotic congenital heart disease accounted for 30% (although they were approximately 50% of the patient population), and patients with benign congenital heart disease accounted for 3% of the charges (approximately 14% of the patients). Thus, charges for patients with congenital heart disease averaged $59 877 between birth and 21 years of age and $21 274 between 22 and 40 years of age, for a total of $81 151 per patient.

Table 1. Disease Categories, Birth to 21 Years

Category	% of Patients*	% Mortality
Benign†	19±3	0
Acyanotic, infant surgery	19±3	12
Acyanotic, later surgery	14±3	4
Acyanotic, with intervention	12±5	2
Cyanotic, non–Fontan operation	24±3	11
Cyanotic, Fontan operation	12±3	25

*Mean± SD.
†No intervention or medical treatment required.

Table 2. Disease Categories, Ages 22 to 40 Years

Category	% of Patients*	% No Longer Seen or Dead by Age 21 y
Benign	12±3	53
Postoperative, acyanotic	52±2	13
Postoperative, tetralogy of Fallot	13±2	...
Postoperative, Mustard operation/Fontan operation	20±4	25
Eisenmenger complex	3±1	...

*Mean± SD.

Table 3. Use of Services in the Cyanotic, Non–Fontan Operation Group, Birth to 21 Years

Subcategory, Age and Results	% of Age Group	Clinic Visit*			Hospitalization*			Years Taking Medication
		Routine	Complex	Pacemaker	Medical	Surgical	Pacemaker	
Birth-1 y	100	2.3	3.3	0	1.1	1.1	1.1	1.0
1-21 y, good	26	5.2	7.0	0	0	0	0	0.2
1-21 y, moderate	43	7.8	13.7	0	0.5	0.5	0.5	13.0
1-21 y, poor	18	5.8	25.8	0	1.6	1.6	1.6	19.0
1-21 y, pacemaker	5	1.7	10.0	24.5	0.5	0.7	0.7	10.0

*Average number of services used during the period stated (eg, between birth and 1 year of age, the average patient had 2.3 routine clinic visits).

Table 4. Volume and Cost of Services by Disease Category, Birth to 21 Years

	Clinic Visit			Hospitalization				
	Routine	Complex	Pacemaker	Medical	Surgical	Intervention	Pacemaker	Medication
Cost, $	164	681	950	5632	35 857	11 042	18 250	650
Disease category, No. of services								
Benign	4.4	3.9	0	0.4	0	0	0	0
Acyanotic, infant surgery	9.0	11.0	1.2	2.0	1.2	0	0.2	8.6
Acyanotic, later surgery	4.9	9.2	0.7	0.1	1.0	0	0.1	0.4
Acyanotic, intervention	3.7	8.2	0	0	0	1.1	0	0
Cyanotic, non–Fontan operation	8.6	16.6	1.4	2.3	1.6	0	0.2	0.5
Cyanotic, Fontan operation	9.4	24.0	1.2	1.9	2.2	0	0.2	13.8

Table 5. Total Charges by Disease Category, Birth to 21 Years

	Charge, $								
	Clinic Visit			Hospitalization					
Category	Routine	Complex	Pacemaker	Medical	Surgical	Intervention	Pacemaker	Medication	Total
Benign	721	2656	0	563	0	0	0	0	3940
Acyanotic, infant surgery	1474	8104	1140	11 262	41 953	0	2920	5590	72 443
Acyanotic, later surgery	803	6265	703	450	35 857	0	1825	260	46 163
Acyanotic, intervention	606	5584	0	0	0	11 925	0	0	18 115
Cyanotic, non–Fontan operation	1409	11 305	1330	12 951	56 295	0	3468	6175	92 933
Cyanotic, Fontan operation	1540	16 344	1140	10 699	78 527	0	2920	8970	120 140

Interinstitutional Variation

The interinstitutional variations between birth and 21 years of age are shown in **Table 7** and **Table 8**. Overall, the minimum charge for an average patient was $47 515 and the maximum was $73 606, a 55% overall variation. This $26 091 variation between the lowest and the highest was accounted for both by variations in practice pattern (volume), which accounted for 55% of the variation, and by price, which accounted for 45% of the variation. The greatest variation in practice was in interventional hospitalizations, medical hospitalizations, and complex clinic visits (echocardiograms); the least variation was in surgical hospitalization. In price, the greatest variation was in medical hospitalization and the least was in the cost of medicine per year.

Mortality and Charges

In Table 8 the mortality from birth to 21 years of age for each institution is shown along with the charges for that institution. There was essentially no relationship between the mortality and the charges. Since the charges were calculated and averaged only for live patients, no

Table 6. Charges by Lesion

	Birth-21 y		22-40 y		Total, $
	%	$	%	$	
Benign	19	3940	12	3470	7410
Acyanotic	45	49 730	52	12 981	62 711
Cyanotic	36	102 084	36	39 187	141 271
Mean	...	59 877	...	21 274	81 151

relationship should be expected between mortality and charges. Therefore, these data can be used as a crude estimate of the benefit-cost ratio.

COMMENT

COST

In this study, we estimated that 91% of patients with congenital heart disease survive to adulthood. A figure of 90% survival to adulthood has been confirmed in other studies.[1-3] According to the data on natural survival patterns in congenital heart disease, the expected lifetime for patients

	Institution					
	A	B	C	D	E	F
Routine clinic visit						
No. of services	6.2	5.5	10.1	7.4	7.0	6.4
Charge per service, $	225	150	233	190	175	210
Total, $	1395	825	2353	1406	1225	1344
Complex clinic visit						
No. of services	16.9	8	9.5	12.9	16	16.3
Charge per service, $	625	1000	643	700	425	642
Total, $	10 563	8000	6109	9630	6800	10 465
Medical hospitalization						
No. of services	1.28	0.55	1.28	1.0	1.23	1.75
Charge per service, $	6350	3500	6350	7000	4500	6090
Total, $	8128	1925	8128	7000	5535	10 658
Surgical hospitalization						
No. of services	1.01	0.94	1.01	0.88	0.96	1.18
Charge per service, $	33 571	39 203	33 571	45 000	26 956	36 844
Total, $	33 907	36 851	33 907	39 600	25 878	43 476
Intervention hospitalization						
No. of services	0.13	0.13	0.10	0.13	0.20	0.05
Charge per service, $	12 500	9000	10 750	10 000	12 500	11 500
Total, $	1625	1170	1075	1300	2500	575

Table 8. Institutional Variation in Mortality and Charges, Birth to 40 Years

	Institution					
	A	B	C	D	E	F
Mortality, %	8	8	5	4	8	7
Charges, $	63 283	57 071	59 187	62 346	47 515	73 606
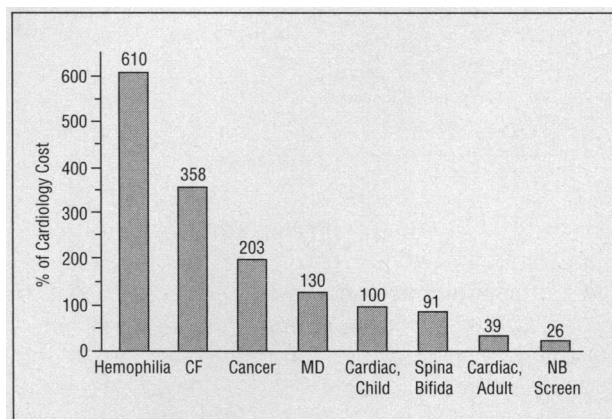
Comparative costs of pediatric diseases. CF indicates cystic fibrosis; MD, muscular dystrophy; and NB, newborn. Data from the Texas Chronically Ill and Disabled Children's Program.[9]

with acyanotic heart disease is a minimum of 19.5 years (because of possible underdetection), and that for unoperated-on cyanotic heart disease is 1.4 years.[7] According to the estimates from this study, with optimal medical and surgical treatment, the expected lifetime for patients with acyanotic heart disease is 64.9 years, and for those with cyanotic heart disease, 46.0 years. These numbers can then be compared with data for patients with untreated heart disease. With the use of the crude measure of years of improved mortality per dollar spent, the cost-benefit ratio for all acyanotic congenital heart disease (including benign) is $46 914/45 years=$1029 per life year, and that for cyanotic congenital heart disease is $141 271/44.6 years=$3168 per life year. For comparison, the data published by Kuppermann et al[8] were adjusted to 1991 constant dollars. Our data for congenital heart disease compare extremely favorably with those of Kuppermann et al (adjusted for 1991 constant dollars): neonatal intensive care (1000- to 1500-g infants), $7040 per life year; neonatal intensive care (500- to 999-g infants), $49 664; and coronary artery bypass surgery for three-vessel disease, $92 016.

COST-BENEFIT COMPARISON WITH OTHER DISEASES

The results of the present study indicate that charges for congenital heart disease averaged $2851 per year between birth and 21 years of age and $1120 per year between the ages of 22 and 40 years. In an attempt to validate these numbers, we compared them with actual payment data from the Texas Chronically Ill and Disabled Children's (CIDC) Program for 1991,[9] in which annual expenditures per patient aged 0 to 21 years were $1854. The CIDC Program pays approximately 70% of charges, thus allowing for calculated charges of $2649. This agrees closely with our estimate of

$2851 for charges. In the **Figure**, the CIDC Program expenditures for children aged 0 to 21 years with heart disease are graphed and compared with expenditures made by the CIDC Program for other chronic diseases in childhood as well as newborn screening. The estimate of the cost per patient per year for adult congenital heart disease on the basis of our study is also included. Therefore, compared with the common childhood diseases, congenital heart disease is among the least expensive. For example, hemophilia costs $11 313 per year (6.1 times the cost of congenital heart disease), and cystic fibrosis costs $6642 per year (3.6 times the cost of congenital heart disease). One weakness of the present study is that the data are based on estimates of practice rather than on actual data. It is not possible to obtain the actual data, since records of each test ordered at each visit for 40 years are not available. Therefore, our method provides the best possible estimates available at present.

VARIATION IN PRACTICE AND PRICE

This study demonstrated a 55% variation among six centers in the charges for congenital heart disease between birth and 21 years of age. This study also demonstrated that this variation in charges was not related to outcome as measured by mortality. It is well recognized that there are other important outcomes, including morbidity and patient satisfaction, but these were not assessed in the present study. Since no benefit of spending more on the patient was demonstrated with the use of mortality as the only outcome, future studies should assess other outcomes. This study demonstrated that approximately half of the variation was in practice pattern. As has been well described by Fowler et al[10] and Wennberg,[11] the variation in practice pattern is not necessarily based on formal estimation of outcome.

On the basis of this study, it appears that the greatest variation was in the use of complex clinic visits and cardiac catheterization. It therefore appears appropriate to examine prospectively the necessity for follow-up echocardiography and follow-up cardiac catheterization after the initial diagnosis in patients with congenital heart disease. Since there was a 55% variation in overall charges, and half of that was accounted for by practice pattern, it should be possible to approach approximately a 30% reduction (0.55×0.50) in overall charges for congenital heart disease by having a practice pattern more like that of hospital B in Table 7 and less like that of hospital E.

Similarly, it may be possible to reduce overall charges by decreasing the variation in prices. This would only hold true if, as demonstrated, values for outcomes were the same at the lowest resource utilization. The currently allowed Medicare variation among the cities represented in this study was 9%.[12] On this basis alone, it is likely that the price variability should be significantly less in the future than that reflected in the present study.

A MODEL FOR CHRONIC DISEASE

This study has demonstrated that a common chronic disease of childhood can be classified into a number of types and, within each type, subclassified by severity of course. By means of a multicenter approach, practice variation can be assessed and related to outcome. The degree to which practice variation does not relate to outcome provides the opportunity to reduce the variation and produce objective guidelines with utilization of less services than currently provided. It is possible that, when presented with this type of objective data, physicians will respond in a positive way to make the practice of medicine more appropriate and to reduce the cost of medical care.

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