
Cryptorchidism and Hypospadias

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Purpose: We quantified the burden of cryptorchidism and hypospadias in the United States by identifying trends in the use of health care resources and estimating the economic impact of the disease.

Materials and Methods: The analytical methods used to generate these results were described previously.

Results: Cryptorchidism is managed almost exclusively in the outpatient setting and insufficient data were available on inpatient health care use. Annual inpatient hospitalizations for hypospadias decreased by 75% between 1994 and 2000 from 2,669 (2.2/100,000 children) to 849 (0.6/100,000). Between 1992 and 2000 there were 611,647 physician office visits (96/100,000 per year) with undescended testis listed as the primary diagnosis. The rate of physician office visits for hypospadias by commercially insured boys younger than 3 years increased significantly from 429/100,000 in 1994 to 655/100,000 in 2002. The annualized rate of 18/100,000 in 1994 to 1996 remained relatively constant during these 3 years. Orchiopexy rates were highest in 0 to 2-year-old children, as generally recommended, but a substantial minority of these procedures was done in 3 to 10-year-old children. Geographic variation was noted with higher ambulatory surgery rates in the Northeast and Midwest than in the South and West. Data on commercially insured boys younger than 3 years revealed a 1.5-fold overall increase in the rate of hypospadias surgery from 321/100,000 in 1994 to 468/100,000 in 2002, reflecting the known increase in hypospadias incidence in the United States during the late 1990s.

Conclusions: Average cost per hospitalization for hypospadias exceeded \$5,389 with costs per case higher in children 3 years or older, although there were more cases in children younger than 3 years. The cost per case of hypospadias was higher in the Northeast and South than in the other regions. Data on cryptorchidism are too sparse to provide insights into its downstream economic costs.

Key Words: testis, cryptorchidism, hypospadias, health services research, health care costs

Cryptorchidism, which is the most common male genital anomaly identified at birth, affects 3% of full-term male newborns, an incidence that has not increased in the last few decades. In comparison the incidence of hypospadias appears to be increasing. Hypospadias has historically been identified in 0.3% of newborn boys¹ but recent evidence suggests that the worldwide incidence may now be 0.8%² of white and 0.4%³⁻⁶ of nonwhite newborn males. Paulozzi et al reported data from 2 surveillance systems in the United States that demonstrated almost a doubling of the rate of hypospadias between 1968 and 1993 with an overall annual rate of increase of 1.4%.³ When analyzed by race, the overall yearly rate of increase was 2.9% in the white and 5.7% in the nonwhite population.³ Additionally, the rate of severe hypospadias increased 3 to 5-fold.³ Gallentine et al reported a contemporary incidence of hypospadias of 0.7% of 99,219 live male births (table 1).⁷ Although no statistically significant racial difference in the incidence of hypospadias was noted, there was a trend toward a greater incidence in the white population.⁷ The observed association between hypospadias and genetic abnor-

malities of sex chromosome number, androgen receptor and testosterone biosynthetic pathways provides evidence that the urethral defect occurs as a consequence of failed induction of the androgen sensitive urethral plate.^{2,8,9} With this in mind some argue that many cases of idiopathic hypospadias may be caused by antiandrogenic substances in the environment, thus, explaining the observed increasing incidence of hypospadias in industrialized Western countries.^{3,5-14} More recent data indicate that the rate of increase may now be reaching a plateau.¹⁵

Ethnic/racial differences in maternal serum testosterone during pregnancy may be also be responsible for hypospadias, as suggested by the inverse correlation between the risk of hypospadias, which is greatest in white boys, and testosterone levels, which are lowest in pregnant white women. Additional support for racial differences in androgenic exposure during early gestation comes from a similar analysis of serum testosterone in Hispanic women, whose infants have an incidence of hypospadias between that of white and that of black American women. Other risk factors for hypospadias are advanced maternal age,² paternal risk factors such as a history of undescended testes and varicoceles,¹⁶ prematurity,^{16,17} low birth weight¹⁷ and monozygotic twin gestation.¹⁸

To quantify the burden of cryptorchidism and hypospadias on the American public the National Institute of Diabetes and Digestive and Kidney Diseases commissioned the Urological Diseases in America project. Its central goal was

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Race/Ethnicity	No. Male Live Births/No. Hypospadias (%)
White	68,444/520 (0.8)
Black	18,984/120 (0.6)
Asian	1,761/9 (0.5)
North American native	175/1 (0.6)
Unknown	9,846/59 (0.6)
Totals	99,210/709 (0.7)

to use existing population based health care claims data to provide a snapshot of the epidemiology, practice patterns and economic burden associated with urological disorders. Data on pediatric urological conditions was extraordinarily limited. This report describes several key findings of this effort and highlights the need for more comprehensive data collection in pediatric urology.

MATERIALS AND METHODS

The analytical methods used to generate these results were described previously (see Appendix).^{19,20}

RESULTS

Trends in Health Care Use

Inpatient. Since cryptorchidism is managed almost exclusively in the outpatient setting, insufficient data were available on inpatient health care use to allow such use to be analyzed. According to the HCUP data set annual inpatient hospitalizations for hypospadias decreased by 75% between 1994 and 2000 from 2,669 (2.2/100,000 children) to 849 (0.6/100,000) (table 2). This decreasing trend was noted across all racial/ethnic groups on which data were collected. Age adjusted rates of inpatient hospitalization did not meet the standard for reliability for black and Hispanic Americans, which limited the analysis of use trends in recent years for these 2 groups.

Additional data from HCUP KID in 1997 and 2000 demonstrated that the majority of inpatient hospitalizations for hypospadias occurred in children younger than 3 years (table 3). In fact, inpatient hospitalizations were 10 times more likely to occur in these children than in those 3 to 10 years old. This is consistent with the common surgical practice of performing correction in younger children, ideally within year 1 of life. Despite the trend toward referring infants younger than 1 year for hypospadias repair some children who were hospitalized for hypospadias in 1997 were as old as 17 years. Many hospital admissions of older children may have been to treat complications of earlier hypospadias repairs or they may have been late referrals of children who should have undergone correction earlier. The proportion of visits for children hospitalized in 2000 for hypospadias repair who were older than 3 years increased to 28% (391 of 1,385), which may reflect an overall trend toward performing more hypospadias procedures in infant as outpatient surgery. Interestingly there was a small trend toward an increase in the rate of inpatient stays for hypospadias repair in children 11 to 17 years old from 0.6/100,000 in 1997 to 0.8/100,000 in 2000. Although this was not statistically significant, it raises the question of whether more complications are being managed in older children. Data

	1994		1996		1998		2000	
	Count	Rate (95% CI)	Count	Rate (95% CI)	Count	Rate (95% CI)	Count	Rate (95% CI)
Totals	2,669	2.2 (1.4-3.0)	1,955	1.5 (0.8-2.2)	1,600	1.2 (0.6-1.9)	849	0.6 (0.4-0.9)
Age (yrs):								
Younger than 18	2,558	7.3 (4.6-10)	1,846	5.1 (2.7-7.4)	1,472	4.0 (1.8-6.2)	686	1.8 (1.0-2.7)
18 or Older	*	*	*	*	129	*	*	*
Race/ethnicity:								
White	1,402	1.5 (0.8-2.3)	1,064	1.1 (0.5-1.8)	821	*	371	0.4 (0.2-0.5)
Black	424	2.9 (1.6-4.1)	308	2.0 (0.8-3.2)	197	*	*	*
Hispanic	231	1.8 (0.8-2.7)	245	*	*	*	*	*
Region:								
Midwest	516	1.8 (0.8-2.7)	164	*	*	*	*	*
Northeast	684	*	667	*	913	*	177	*
South	958	*	711	*	207	0.4 (0.2-0.7)	187	*
West	511	*	413	*	324	*	367	*
Urban MSA†	2,602	2.8 (1.8-3.9)	1,898	1.9 (1.0-2.8)	1,572	1.6 (0.7-2.4)	817	0.8 (0.4-1.1)

Rate per 100,000 based on 1994, 1996, 1998 and 2000 population estimates from CPS, CPS Utilities, Unicon Research Corp. for relevant demographic categories of adult male civilian noninstitutionalized population in the United States, age adjusted rate adjusted to the United States Census derived age distribution of the year under analysis and individuals of other races, and with missing or unavailable race and ethnicity, and missing MSA included in the total (counts may not sum to total due to rounding) (source: HCUP Nationwide Inpatient Sample, 1994, 1996, 1998 and 2000).

* Value does not meet reliability or precision standard.

† Rural MSA value does not meet reliability or precision standard.

TABLE 3. Inpatient hospital stays for hypospadias as primary diagnosis in 1997 and 2000

	1997				2000		
	Count	Rate (95% CI)	Age Adjusted Rate	% All Hospitalizations	Count	Rate (95% CI)	% All Hospitalizations
Totals	1,889	5.2 (3.6–6.7)	5.1	0.06	1,385	3.7 (2.5–5.0)	0.04
Age (yrs):							
Younger than 3	1,421	24 (16–31)		0.06	993	17 (11–22)	0.04
3–10	385	2.3 (1.6–3.1)		0.10	277	1.6 (0.9–2.4)	0.09
11–17	82	0.6 (0.3–0.9)		0.02	114	0.8 (0.4–1.1)	0.03
Race/ethnicity:							
White	954	4.0 (2.7–5.4)	4.1	0.07	643	2.8 (1.8–3.7)	0.04
Black	169	3.0 (1.5–4.5)	3.1	0.04	132	2.3 (1.4–3.3)	0.03
Hispanic	274	*	4.2	0.07	200	*	0.04
Region:							
Midwest	149	1.7 (0.9–2.6)	1.6	0.02	140	*	0.02
Northeast	706	10 (5.6–15)	11.0	0.11	463	7.0 (3.5–10)	0.08
South	388	*	3.1	0.03	282	2.2 (1.0–3.5)	0.02
West	646	*	7.2	0.08	499	*	0.06
MSA:							
Rural	44	*	*	0.01	25	*	0.01
Urban	1,845	6.6 (4.6–8.5)	6.3	0.07	1,357	4.7 (3.1–6.2)	0.05

Rate per 100,000 based on 1997 population estimates from CPS, CPS Utilities, Unicon Research Corp. for relevant demographic categories of male civilian noninstitutional population younger than 18 years in the United States, age adjusted rate adjusted to 2000 United States Census and individuals of other races, and with missing race and ethnicity, and missing MSA included in the total (counts may not sum to total due to rounding and Race/ethnicity breakdown not included because of large percent of missing values in 1997) (source: HCUP KID, 1997 and 2000).

* Value does not meet reliability or precision standard.

on inpatient hospital stays for children insured commercially or through Medicaid were insufficient to generate reliable estimates of use by insurer type.

Outpatient. Children are customarily seen in the outpatient setting for cryptorchidism and hypospadias evaluation, and they subsequently undergo ambulatory surgery. We focused on visits for which cryptorchidism or hypospadias was the primary diagnosis.

Physician office visits. According to National Ambulatory Medical Care Survey data between 1992 and 2000 there were 611,647 physician office visits (96/100,000 yearly) for undescended testis listed as the primary diagnosis (table 4). The overwhelming majority of the patients were younger than 18 years.

Tables 5 and 6 list CHCPE data on outpatient care of children with commercial and Medicaid insurance, respectively. In commercially and Medicaid insured boys the most common site of care was the physician office (tables 5 and 6). The rate of physician office visits for hypospadias by commercially insured boys younger than 3 years in-

creased significantly from 429/100,000 in 1994 to 655/100,000 in 2002. Data from Medicaid for 1994 and 2002 were less reliable. However, they suggested stable use of physician office visits at 160/100,000 population throughout the mid to late 1990s, decreasing to 118/100,000 in subsequent years.

Ambulatory surgery. Data from the National Survey of Ambulatory Surgery indicated an annualized rate of 18 cases per 100,000 population in 1994 to 1996 (table 7). The rate remained relatively constant during these 3 years. Although orchiopexy rates are highest in 0 to 2-year-old children, as recommended, a substantial minority of these procedures was done in 3 to 10-year-old children. Geographic variation was noted with higher ambulatory surgery rates in the Northeast and Midwest than in the South and West.

Since hypospadias is a disease that must be treated surgically in most cases, it is not surprising that almost as many visits occurred at ambulatory surgery centers as in the office setting. Data on commercially insured boys younger

TABLE 4. Physician office visits for undescended testis as primary diagnosis and outpatient hospital visits for hypospadias as any diagnosis

	Count	Rate (95% CI)	Av Annualized Rate/Yr	Age Adjusted Rate
Physician office visits:*				
Totals	611,647	480 (288–671)	96	476
Age younger than 18 yrs	534,144	1,492 (838–2,146)	298	
MSA	583,235	599 (351–847)	120	589
Total outpt hospital visits†	69,457	54 (30–77)	14	54

* Rate per 100,000 based on 1992, 1994, 1996, 1998 and 2000 population estimates from CPS, CPS Utilities, Unicon Research Corp. for relevant demographic categories of male civilian noninstitutionalized population in the United States, age adjusted rate adjusted to the United States Census derived age distribution of the mid point of years, individuals of missing or unavailable race and ethnicity, and with missing MSA included in the total, and age 18 years or older, race/ethnicity and nonMSA values did not meet reliability or precision data (counts may not sum to total due to rounding) (source: National Ambulatory Medical Care Survey, 1992, 1994, 1996, 1998 and 2000).

† Rate per 100,000 based on 1994, 1996, 1998 and 2000 population estimates from CPS, CPS Utilities, Unicon Research Corp. for relevant demographic categories of male civilian noninstitutionalized population in the United States, age adjusted rate adjusted to the United States Census derived age distribution of the mid point of years, and individuals of missing or unavailable race and ethnicity, and with missing MSA included in the total (counts may not sum to total due to rounding) (source: National Hospital Ambulatory Medical Care Survey, 1994, 1996, 1998 and 2000).

TABLE 5. Visits for hypospadias as primary diagnosis for children with commercial health insurance

	1994		1996		1998		2000		2002	
	Count	Rate	Count	Rate	Count	Rate	Count	Rate	Count	Rate
Physician office	124	24	215	27	329	27	387	28	335	28
Age (yrs):										
Younger than 3	88	429	143	483	224	497	280	583	252	655
3-10	19	*	43	41	70	45	73	44	57	43
11-17	7	*	14	*	13	*	13	*	15	*
Region:										
Midwest	84	27	123	28	147	25	168	25	173	27
Northeast	11	*	17	*	41	41	20	*	11	*
Southeast	14	*	56	26	112	24	173	32	141	31
West	15	*	19	*	29	*	26	*	10	*
Emergency room	0	*	1	*	1	*	4	*	1	*
Inpt	3	*	6	*	2	*	7	*	6	*
Hospital outpt	1	*	15	*	19	*	18	*	21	*
Ambulatory surgery	86	17	127	16	187	15	262	19	256	22
Age (yrs):										
Younger than 3	66	321	82	277	134	297	189	387	180	468
3-10	10	*	33	31	36	23	48	29	50	38
11-17	4	*	5	*	8	*	11	*	12	*
Region:										
Midwest	55	18	71	16	89	15	126	19	123	19
Northeast	14	*	13	*	18	*	11	*	9	*
Southeast	9	*	28	*	63	13	106	19	112	24
West	8	*	15	*	17	*	19	*	12	*

Value does not meet reliability or precision standard.

* Rate per 100,000 based on member months of enrollment in calendar years for males in the same demographic stratum (source: CHCPE, 1994, 1996, 1998, 2000 and 2002).

than 3 years revealed a 1.5-fold overall increase in the rate of hypospadias surgery from 321/100,000 in 1994 to 468/100,000 in 2002, reflecting the known increase in hypospadias incidence in the United States during the late 1990s (tables 5 and 6). Similar estimates could not be obtained for Medicaid insured boys. Data from the National Survey of Ambulatory Surgery showed that during 1994, 1995 and 1996 more than 39,000 visits to ambulatory surgery centers were associated with hypospadias repair, including 67% in infants and the remainder in 3 to 10-year-old children (table 7). Age adjusted rates of visits were highest in the Northeast and Midwest, that is an average of 1.5 to 2.2 times the rates in the West and South, respectively.

Hospital outpatient and emergency room care. Data from the National Hospital Ambulatory Medical Care Sur-

vey indicated an average annualized rate of hospital outpatient visits for hypospadias of 14/100,000 children (table 4). CHCPE data on visits by children insured commercially or through Medicaid for whom hypospadias was listed as a primary diagnosis were inconclusive since most children with hypospadias are evaluated on a nonemergent basis (tables 5 and 6).

Economic Impact

The average cost per hospitalization for hypospadias exceeded \$5,389 with costs per case higher in children 3 years or older, although there were more cases in children younger than 3 years (table 8). The cost per case of hypospadias was higher in the Northeast and the South than in the other regions. A total of 1,385 cases of hypospadias were observed

TABLE 6. Visits for hypospadias as primary diagnosis for children with Medicaid health insurance

	1994		1996		1998		2000		2002
	Count	Rate	Count	Rate	Count	Rate	Count	Rate	Count*
Physician office	19	†	52	161	36	163	32	118	21
Age (yrs):									
Younger than 3	15	†	37	537	29	†	21	†	15
3-10	4	†	12	†	5	†	10	†	4
11-17	0	†	3	†	2	†	1	†	2
Region:									
Midwest	18	†	18	†	0	†	0	†	0
Northeast	0	†	21	†	19	†	16	†	14
Southeast	0	†	0	†	0	†	0	†	0
West	1	†	13	†	17	†	16	†	7
Emergency room	0	†	0	†	0	†	0	†	1
Inpt	1	†	1	†	1	†	0	†	0
Hospital outpt	0	0	0	0	0	0	0	0	6
Ambulatory surgery	10	†	27	†	21	†	21	†	18

Rate per 100,000 based on member months of enrollment in calendar years for males in the same demographic stratum (source: CHCPE, 1994, 1996, 1998, 2000 and 2002).

* Rate does not meet reliability or precision standard.

† Value does not meet reliability or precision standard.

TABLE 7. Ambulatory surgery visits for undescended testis as primary diagnosis and hypospadias as any diagnosis

	1994-1996				1994			1995			1996		
	Count	Rate (95% CI)	Av Annualized Rate/Yr	Age Adjusted Rate	Count	Rate (95% CI)	Age Adjusted Rate	Count	Rate (95% CI)	Age Adjusted Rate	Count	Rate (95% CI)	Age Adjusted Rate
Totals	69,731	55 (47-63)	18	55	24,247	20 (15-25)	20	18,781	15 (11-18)	15	26,703	21 (16-26)	21
	<i>Undescended testis</i>												
Age (yrs):													
0-2	25,907	422 (329-515)	141		8,622	141 (89-192)		8,581	139 (78-201)		8,704	142 (96-189)	
3-10	31,853	196 (153-239)	65		11,842	74 (45-104)		6,668	41 (28-54)		13,343	81 (53-109)	
11-17	5,782	43 (26-60)	14		*	*		*	*		*	*	
Region:													
Northeast	17,172	69 (46-92)	23	72	7,325	*	31	4,500	8	19	5,347	22 (13-30)	22
Midwest	17,806	60 (44-76)	20	59	5,910	20 (10-30)	20	5,899	20 (10-29)	20	5,997	20 (12-28)	20
South	21,593	50 (37-62)	17	49	5,974	14 (9.4-20)	15	4,656	11 (6.9-14)	11	10,963	25 (14-35)	25
West	13,160	46 (31-61)	15	45	5,038	18 (8.6-28)	17	3,726	13 (5.5-21)	12	4,396	15 (6.7-24)	15
	<i>Hypospadias</i>												
Totals	39,631	31 (25-38)	10.3	31	16,171	13 (8.2-18)	13	10,465	8.2 (5.0-11)	8.2	12,995	10 (7.2-13)	10
Age (yrs):													
0-2	26,381	430 (333-527)	143.3		9,032	147 (84-211)		7,700	125 (75-176)		9,649	158 (104-211)	
3-10	7,296	45 (22-68)	15.0		*	*		*	*		*	*	
Region:													
Northeast	9,706	39 (19-59)	13.0	40									
Midwest	13,480	45 (28-62)	15.0	45									
South	8,345	19 (13-25)	6.3	19									
West	8,100	28 (16-40)	9.3	27									

Rate per 100,000 based on 1994, 1995 and 1996 population estimates from CPS, CPS Utilities, Unicon Research Corp. for relevant demographic categories of male civilian noninstitutionalized population in the United States, grouped years age adjusted to the United States Census derived age distribution of the mid point of years and individual years age adjusted to the United States Census derived age distribution of the year under analysis (counts may not sum to totals due to rounding) (source: National Survey of Ambulatory Surgery, 1994, 1995 and 1996).

Value for ages 18 years or older for undescended testis, and 11 to 17 and 18 years or older for hypospadias do not meet reliability or precision standard.

* Value does not meet reliability or precision standard.

TABLE 8. Mean inpatient cost per child admitted with hypospadias as primary diagnosis in 1999 to 2001

	Count	Mean Cost (95% CI)* (\$)
Totals	765	5,389 (5,170–5,609)
Age (yrs):		
0–2	551	5,194 (4,925–5,463)
3–10	154	5,858 (5,414–6,303)
11–17	47	5,716 (5,136–6,296)
18–29	13	6,914 (4,546–9,282)
Race/ethnicity:		
White	453	5,366 (5,090–5,642)
Black	83	5,802 (4,925–6,679)
Asian	25	4,871 (3,889–5,852)
Hispanic	92	5,416 (4,822–6,011)
Missing	31	6,420 (5,425–7,415)
Other	81	4,832 (4,138–5,526)
North American native	0	
Region:		
Midwest	126	5,330 (4,802–5,858)
Northeast	155	5,834 (5,309–6,360)
South	233	5,582 (5,168–5,996)
West	251	4,966 (4,610–5,321)

* Calculated using adjusted ratio of costs to charges, including variable and fixed cost among participating children's hospitals (source: National Association of Children's Hospitals and Related Institutions, 1999 to 2001).

in 2000 in KID data, incurring an estimated \$8 million in national inpatient expenditures (table 3).

DISCUSSION

Cryptorchidism and hypospadias are managed almost exclusively in the outpatient setting. According to the HCUP data set annual inpatient hospitalizations for hypospadias decreased by 75% between 1994 and 2000, illustrating the trend toward outpatient treatment. This decreasing trend was noted across all racial/ethnic groups. The rate of physician office visits for hypospadias by commercially insured boys younger than 3 years increased significantly from 429/100,000 in 1994 to 655/100,000 in 2002. According to National Ambulatory Medical Care Survey data between 1992 and 2000 the rate of physician office visits for undescended testis was constant at 96/100,000 population. Orchiopexy rates from the National Survey of Ambulatory Surgery indicate an annualized rate of 18 cases per 100,000 population in 1994 to 1996 and the rate remained relatively constant during these 3 years. Data on commercially insured boys younger than 3 years revealed a 1.5-fold overall increase in the rate of hypospadias surgery from 321/100,000 in 1994 to 468/100,000 in 2002, reflecting the known increase in hypospadias incidence in the United States during the late 1990s.

The economic impact of cryptorchidism could not be assessed due to limited data. The average cost per hospitalization for hypospadias exceeded \$5,389. The cost per case of hypospadias was higher in the Northeast and South than in the other regions. HCUP KID data on 2000 indicate an estimated \$8 million in national inpatient expenditures for hypospadias treatment (1,385 cases).

APPENDIX

Updates are available on line (www.uda.niddk.nih.gov).²⁰

Abbreviations and Acronyms

CHCPE	=	Center for Health Care Policy and Evaluation
CPS	=	Current Population Survey
HCUP	=	Health Care Cost and Utilization Project
KID	=	Kids' Inpatient Database
MSA	=	metropolitan statistical area

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