



# Geographical Distribution of Access to Healthcare in Patients Diagnosed with Hypospadias

## *Distribución geográfica del acceso a la asistencia sanitaria en pacientes con diagnóstico de hipospadias*

Nicolas Fernandez<sup>1</sup> Jessica Santander<sup>2</sup> Marcela Pérez-Sosa<sup>3</sup> Alejandro Agudelo<sup>4</sup>  
Daniel Sánchez<sup>4</sup> Nevianni Vélez<sup>4</sup> Ignacio Zarante<sup>5</sup> Andres Gutierrez<sup>6</sup>

<sup>1</sup> Division of Urology, Seattle Children's Hospital, University of Washington, Seattle, Washington, United States

<sup>2</sup> Department of Urology, Fundación Santa Fe de Bogota, Universidad de los Andes, Bogotá, Colombia

<sup>3</sup> School of Medicine, Universidad Nacional de Colombia, Bogotá, Colombia

<sup>4</sup> School of Medicine, Universidad de los Andes, Bogotá, Colombia

<sup>5</sup> Instituto de Genética Humana, Pontificia Universidad Javeriana, Bogotá, Colombia

<sup>6</sup> Division of Urology, Hospital Universitario San Ignacio, Pontificia Universidad Javeriana, Bogotá, Colombia

Address for correspondence Nicolás Fernández, MD, PhD, Division of Urology, Seattle Childrens Hospital, 4800 Sandpoint Way NE. 98105, Seattle, Washington, United States  
(e-mail: nicolas.fernandez@seattlechildrens.org).

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### Abstract

**Objective** Hypospadias is a congenital disease of unknown etiology involving multiple epigenetic, genetic, and endocrinological factors. It is a highly incapacitating condition. Its surgical management is one of the most frequent surgical procedures done by pediatric urologists. Furthermore, the geographical distribution and health-care access is limited in Colombia. The Colombian Ministry of Health has consolidated a nationwide registry called Integrated Social Protection Information System (SISPRO, in the Spanish acronym) to collect comprehensive information on the use and frequency of resources associated with health care in Colombia. The aim of the present study was to analyze the number of cases reported between 2014 and 2018 and the geographical distribution of access to healthcare of patients with hypospadias in Colombia.

**Methods** An observational, retrospective study of hypospadias in Colombia, 2014–2018, was performed using data extracted from the Individual Health Records System (RIPS) in SISPRO. Satscan, version 9.6 was used to perform a distribution analysis of the georeferenced population using a Poisson model. To visualize the results, the software projected the result onto a Google Earth map.

**Results** Between January 2014 and December 2018, a total of 8,990 cases of hypospadias were evaluated in Colombia. The geographical distribution in the national territory has areas with high evaluation rates. On average, the departments in which the majority of cases were evaluated during the study period were Bogotá, D.C.,

### Keywords

- ▶ hypospadias
- ▶ spatial analysis
- ▶ prevalence

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Antioquia, and Valle del Cauca (2,196, 1,818 and 1,151 cases, respectively). The statistical analysis of the space exploration (► **Fig. 1**) identified the area with the highest concentration of cases (red) and the areas in which the lowest number of patients was evaluated (blue). The geographical distribution showed increasing trends in areas near the center of the country, especially in the cities of Bogotá, Cali, Ibagué, and Pereira. **Conclusion** There is a greater concentration of cases evaluated in the center of the country, where the cities with better access to subspecialized medical care are located. This highlights inequalities in health services and the opportunity for surgical care among regions of the country. If we consider that the prevalence rates of hypospadias remain stable, ~ 87% of the patients with hypospadias will not be evaluated by a subspecialist.

## Resumen

**Objetivo** Hipospadias es una enfermedad congénita de etiología desconocida que involucra múltiples factores epigenéticos, genéticos y endocrinológicos. Es una condición sumamente incapacitante. Su manejo quirúrgico es uno de los procedimientos quirúrgicos más frecuentes realizados por urólogos pediátricos. Además, la distribución geográfica y el acceso a la atención médica son limitados en Colombia. El Ministerio de Salud de Colombia ha consolidado un registro a nivel nacional denominado Sistema Integrado de Información de Protección Social (SISPRO) para recopilar información integral sobre el uso y frecuencia de los recursos asociados a la atención de la salud en Colombia. El objetivo del presente estudio fue analizar el número de casos notificados entre 2014 y 2018 y la distribución geográfica del acceso a la atención médica por los pacientes con hipospadias en Colombia.

**Métodos** Se realizó un estudio observacional y retrospectivo de hipospadias en Colombia, 2014-2018, utilizando datos extraídos del Sistema de Registros Sanitarios Individuales (RIPS) en SISPRO. Se usó Satscan, versión 9.6 para realizar un análisis de distribución de la población georreferenciada usando un modelo de Poisson. Para visualizar los resultados, el *software* proyectó el resultado en un mapa de Google Earth.

**Resultados** Entre enero de 2014 y diciembre de 2018, se evaluaron un total de 8.990 casos de hipospadias en Colombia. La distribución geográfica en el territorio nacional tiene áreas con mayor concentración de la atención de pacientes con hipospadias, al igual que áreas sin atención de esta condición. En promedio, los departamentos donde se evaluaron la mayoría de los casos durante el período de estudio fueron Bogotá, D.C., Antioquia, y Valle del Cauca (439.2, 363.6, y 230.2, respectivamente). El análisis estadístico de la exploración espacial (► **Figura 1**) identificó el área con la mayor concentración de casos (rojo) y las áreas donde se evaluó el menor número de pacientes (azul). La distribución geográfica mostró tendencias crecientes en áreas cercanas al centro del país, especialmente en las ciudades de Bogotá, Cali, Ibagué y Pereira.

**Conclusiones** Existe una mayor concentración de casos evaluados en el centro del país, donde se encuentran las ciudades con un mejor acceso a atención médica subspecializada. Esto pone de relieve las desigualdades en el acceso a los servicios de salud y la oportunidad de atención quirúrgica entre las regiones del país. Si consideramos que las tasas de prevalencia de hipospadias permanecen estables, aproximadamente el 87% de los pacientes con hipospadias no serán evaluados por un subspecialista.

## Palabras clave

- hipospadias
- análisis espacial
- prevalencia

## Introduction

Hypospadias is a congenital malformation caused by an abnormal closure of the urethral folds during penile development, giving rise to a urethral opening on the ventral surface of the penis.<sup>1-3</sup> It is the second most common congenital malformation, after cryptorchidism.<sup>1</sup> Its prevalence has been described with results such as 20.9 per 10,000 live births worldwide, between 10.6 and 34.7 per 10,000 live births in the European continent, 34.2 per 10,000 live births in North America, and a range between 1.5 and 8.6 per 10,000 live births for Latin American countries.<sup>2,4</sup> In Colombia, the prevalence of hypospadias has been described to be of 11.3 per 10,000 live births.<sup>3,5</sup> It is a multifactorial disease, as it has been associated with disruptions in hormonal homeostasis during embryogenesis, multiple environmental factors, and genetic predispositions.<sup>6-8</sup> The anatomical abnormalities of hypospadias have several long-term consequences on genitourinary function that carry a high Burden of Disease (BoD).<sup>9</sup> Early surgical intervention has been proven to be necessary for a favorable quality of life and reduction in disability.<sup>10,11</sup> Hypospadias is a disease with serious and long-lasting psychosocial repercussions.<sup>12,13</sup> In mid- and low-income countries, access to specialized care is limited. Understanding the geographical distribution of pediatric urology services is needed to improve access and allocate public health resources to improve areas with lack of coverage.

The Colombian Ministry of Health has developed the Integrated Social Protection Information System (SISPRO, in the Spanish acronym). One of the components of SISPRO is the Individual Health Record System (Registro Individual de Prestación de Servicios [RIPS, in the Spanish acronym]). The purpose of the RIPS is to centralize health information, facilitating the knowledge of the health or disease status of the population across the regions by time period, insurance regimen, gender and age, among other characteristics. The RIPS is publicly available and is reported on a monthly basis by healthcare providers, health maintenance organizations, and healthcare professionals.

The aim of the present study is to describe the geographical distribution of access to healthcare of patients with hypospadias in Colombia.

## Methods

### Data Collection

For the present observational, retrospective study, data were collected from the RIPS from 2014 to 2018, according to the International Statistical Classification of Diseases and Health Problems 10th revision (ICD-10) related to hypospadias (Q540, Q541, Q542, Q543, Q548, Q549). These diagnoses were scrutinized with two diagnostic filters: confirmed new and confirmed repeated cases, excluding the type of diagnosis, diagnostic impression, and unspecified. To quantify the number of patients with this diagnosis, the function “people attended” was used, which includes each person only once, even if that person has been attended more than once during the previous 5 years.

Analyzed data included all registered male births during the study period in each geographic area, according to vital statistics of births and deaths reported by the National Administrative Department of Statistics (DANE, in the Spanish acronym). For geographical analysis, the geolocation data (latitude and longitude) from each department was extracted from the geoportal of the National Administrative Department of Statistics, using their capital cities as a geographical reference.

To determine the availability of a specialist, we identified all of the members of the Colombian Urological Society. We considered pediatric urologists those who have a dedication of > 70% of their time to seeing urology pediatric patients.

### Statistical Analysis

The information was recorded in Microsoft Excel for MacOS (Microsoft Corp., Redmond, WA, USA). Satscan v9.6 for MacOS (Satscan) was used to analyze the database, where the georeferenced population distribution values were obtained according to the geographic coordinates. Using a Poisson model, a geographical distribution of hypospadias in Colombia was made. To visualize the results obtained, the software projected the outcome in a Google Earth map.

## Results

Between January 2014 and December 2018, a total of 8,990 cases (“people attended”) of hypospadias were assessed in the Colombian territory. ► **Table 1** shows the number of cases seen per year, in each department. The geographical distribution in the national territory has areas with high evaluation rates. On average, the departments where most cases were evaluated during the study period were Bogotá, D.C., Antioquia and Valle del Cauca (2,196, 1,818 and 1,151 cases, respectively).

Spatial scan statistical analysis identified a total of three clusters, as shown in ► **Figure 1**. The area with the highest concentration of cases is shown in red. The geographic distribution showed increasing trends in areas close to the center of the country, especially in the municipalities of Bogotá, Cali, Ibagué, and Pereira, which agrees with the area where more specialists are available. Meanwhile, clusters in blue shows those municipalities below the average value of cases of hypospadias assessed for the years between 2014 and 2018. The analysis detected areas in the north region of the country, including Barranquilla, Cartagena, Sincelejo, and Montería, where the number of children evaluated was lower compared with the center of the country. The same occurs for the area delimited by the second cluster, which encompasses the Amazon and Orinoquia regions.

## Discussion

### Main Findings

The present study provides data about the geographical distribution of the cases of hypospadias seen in Colombia from 2014 to 2018. These results show that there is a greater concentration of evaluated cases in the center of the country, where major cities are located with possible better access to a

**Table 1** Number of people attended in the Colombian territory between 2014 and 2018

	Hypospadias cases assessment*					TOTAL
	2014	2015	2016	2017	2018	
DEPARTMENT						
Amazonas	1	3	0	1	1	6
Antioquia	326	334	289	382	487	1818
Arauca	9	6	3	6	10	34
Archipiélago de San Andrés, Providencia y Santa Catalina	3	2	1	0	1	7
Atlántico	232	227	139	173	194	965
Bogotá, D.C.	478	416	379	472	451	2196
Bolívar	149	125	76	109	117	576
Boyacá	53	50	35	38	36	212
Caldas	54	49	52	28	62	245
Caquetá	17	10	15	15	25	82
Casanare	10	17	13	13	12	65
Cauca	31	38	44	25	32	170
Cesar	118	141	134	159	125	677
Chocó	12	20	17	28	21	98
Córdoba	94	93	88	81	87	443
Cundinamarca	144	150	150	157	182	783
Guaviare	2	2	2	0	1	7
Huila	55	43	39	49	62	248
La Guajira	33	31	26	46	56	192
Magdalena	89	78	47	65	85	364
Meta	36	21	22	21	27	127
Nariño	47	54	42	59	57	259
Norte de Santander	54	52	34	54	59	253
Putumayo	5	6	7	10	6	34
Quindio	29	20	13	21	26	109
Risaralda	90	75	37	54	67	323
Santander	128	144	122	109	156	659
Sucre	88	68	38	45	56	295
Tolima	54	44	36	32	48	214
Valle del Cauca	233	239	193	225	261	1151
Vaupés	3	2	0	0	0	5
Vichada	2	1	0	0	0	3

\*Reported as n.

subspecialized medical care. This highlights inequities in access to health services and surgical care opportunity among the different regions of the country. To our knowledge, there are 11 full-time pediatric urologists in Bogotá, 2 in Bucaramanga, and 1 in Cali. This explains why the care of hypospadias patients is concentrated in the center of Colombia, making the access to children from other regions difficult. The concentration of resources is inequitable. Specifically for hypospadias, there should be a national system that controls referrals and, probably, a system in

which specialized teams go to remote areas and treat these patients may be a cost-effective system. The variety of congenital anomalies in pediatric urology added to the prevalence of these conditions worldwide and the lack of access to trained specialists highlights the critical need to consider pediatric urology as a global surgical health priority.<sup>14</sup>

Taking into account the DANE vital statistics for 2017, congenital abnormalities were among the main causes of mortality in children < 1 year old, causing 23.4% of the



**Fig. 1** Spatial-temporal analysis. Identified hypospadias clusters with increasing trends in cases assessed (red) and clusters with decreasing trends (blue).

deaths in our country.<sup>15</sup> The “Ten-Year Public Health Plan 2012–2021” (Plan Decenal de Salud Pública) includes the surveillance of birth defects as a fundamental element to guide actions aimed at reducing the morbidity and mortality of these conditions, preventing the consequences that represent a high cost for the family and the state.<sup>16</sup> Although the notification of these birth defects has increased in recent years, there are still limitations regarding the registration of cases, mainly because data are based on births that occur in hospitals, without taking into account the percentage of births that occur in rural areas.

### Contrast with Literature

During the last decade, the prevalence of hypospadias has remained constant in Colombia (6.8 per 10,000 newborns).<sup>3</sup> International guidelines have suggested that the ideal age for hypospadias correction surgery is between 6 and 18 months old.<sup>17</sup> The average age at the moment of surgery for hypospadias patients is 7.9 years old in Colombia. Its main reasons are lack of awareness of how to refer patients, delays on administrative

processes during referral, and limited manpower of subspecialized pediatric urologists within the national territory.<sup>18</sup> It is necessary to continue monitoring these patients to identify complications that may require reintervention in a timely manner as well as re-evaluating the psychological repercussions associated with this condition and long-term results of surgical interventions.<sup>19</sup> In a health system with limited resources such as ours, these patients are not prioritized because this is not considered a life-threatening condition.<sup>20</sup> Nonetheless, prompt surgical treatment prevents a secondary disability. Delaying it may increase the BoD.<sup>21,22</sup> There are also indirect implications. At our system, having a patient with a congenital anomaly creates an economic burden to the family of the patients, since one of the guardians needs to stay at home and care for the child, reducing familial income and creating more limitations to healthcare access.<sup>18</sup> A study conducted by Poenaru et al<sup>23</sup> collected surgical records of 13 different congenital anomalies from a high-income setting (Canada) and a low-income country (Kenya). The objective was to calculate DALYs (Disability Adjusted Life Years) caused by delayed

treatment, taking into account the weights of the disability as well as the ideal and real age of surgery. The results showed that delayed surgery were longer in Kenya for most of the procedures included in the analysis. The greatest delays were mainly in children with intestinal and urological conditions. Regarding hypospadias repair, African children had a greater delay in treatment when compared with Canadian children (50.0 months versus 17.2 months,  $p < 0.0001$ ), which translated into higher accumulated DALYS. Similar findings were reported by other groups, like that of Yousef et al.,<sup>20</sup> that reported surgical delays of 2.1 years for the treatment of the most common congenital anomalies, being the longest for hypospadias and cryptorchidism. These conditions are associated with significant disability weights and, therefore, are responsible for more than 75,000 DALYS. According to our results, cases presented in regions far from the center of the country may not be treated in a timely manner, which increases the burden of the disease and DALYS. Timely surgery avoids years of potential morbidity and reduces the risk of lifelong disability. DALYS increase with each year of treatment delay, and even surgical results may be worse as the age of the children increases for some congenital anomalies.<sup>24</sup>

According to the Global Burden of Disease Study, ~ 11% of the global BoD is attributable to surgical conditions, including congenital malformations.<sup>25</sup> Birth defects are a global health problem with limited access to surgical services, mainly in low- and middle-income countries (LMICs).<sup>26</sup> Congenital abnormalities of the urinary tract represent a large proportion of these conditions. In Colombia, the incidence of genitourinary tract anomalies has been estimated as 43 per 100,000 live births.<sup>27</sup> Based on this information, hypospadias is the most common congenital anomaly in the Colombian territory, and the lack of a well-distributed health system will pose a burden to the system if this and other congenital anomalies are not treated appropriately. The long-term implications should be taken into account, and efforts to improve subspecialized medical access need to be implemented. Although universal healthcare coverage is a highlight of the Colombian system, conditions like hypospadias need to be referred to and managed at highly specialized centers. A recent study conducted by Hanna et al.<sup>28</sup> found that 7.1 million people (~ 15.1% of the population) in Colombia do not have timely healthcare access. The most affected populations are found in the Amazon and Orinoquia regions, which supports our results. If we consider that the prevalence rates of hypospadias remain stable, if not increasing, over time, there is hypothetically the possibility that roughly 87% of the patients with hypospadias will never be assessed by a subspecialist if the distribution of patient care is reflected based on our results.

### Limitations

Among the limitations of the present study, it is important to mention the difficulty related to data reporting by the RIPS, such as the inadequate register of the cases. Additionally, we did not consider pediatric surgeons who also treat these patients. Despite this limitations, the present study offers an overview of the distribution of access to health services for patients with a pathology that can be disabling.

## Conclusion

There is a greater concentration of cases evaluated in the center of the country, where the cities with better access to subspecialized medical care are located. This highlights inequalities in health services and the opportunity for surgical care among regions of the country. If we consider that the prevalence rates of hypospadias remain stable, ~ 87% of patients with hypospadias will not be evaluated by a subspecialist.

### Consideraciones Éticas

Se preservarán con exactitud los datos de los resultados obtenidos, en concordancia con los principios reconocidos científicamente. Los investigadores del estudio deben garantizar la protección de los datos recolectados. Se seguirá el artículo 1.6 de la declaración de Helsinki que determina: "Siempre debe respetarse el derecho del participante en la investigación de proteger su integridad y debe adoptarse todo clase de precauciones para resguardar la privacidad del individuo y para reducir al mínimo los efectos de la investigación sobre su integridad física y mental y sobre su personalidad".

Este estudio no conlleva ningún tipo de riesgo dado que no se realizarán intervenciones sobre el paciente. Para el desarrollo del estudio se tendrán en cuenta las consideraciones éticas descritas en el capítulo I, artículo 11 de la "investigación sin riesgo", en los que no se realiza ninguna intervención, contenido en la resolución n° 008430 de 1993 del Ministerio de Salud y la Ley 84 de 1989.

The present study complies with the principles established in the Declaration of Helsinki. The authors declare that there are no conflicts of interest. This study is classified as a research study without risk, according to resolution 8430 of 1993, since it is part of the studies that use retrospective documentary research techniques and methods and those in which no intervention or intentional modification of the variables is carried out.

### Conflict of Interests

The authors have no conflict of interests to declare.

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