# The Global Spread of Autism Spectrum Disorder: The Case of Costa Rica

By

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# LIST OF ABBREVIATIONS

| | |
|---|---|
| ADOS | Autism Diagnostic Observation Schedule |
| ADI-R | Autism Diagnostic Interview-Revised |
| ADHD | Attention deficit hyperactivity disorder |
| BIC | Bayesian Information Criterion |
| CATIE | Tropical Agricultural Research and Higher Education Center |
| CENSINAI | Centers for Education, Nutrition, and Integrated Infant Health |
| DSM | Diagnostic and Statistical Manual of Mental Disorders |
| EBAIS | Basic Teams for Integrated Healthcare |
| HNN | Hospital Nacional de Niños |
| INEC | Costa Rican National Institute of Statistics and Census |
| IRR | Incident rate ratio |
| LLR | Log likelihood ratio |
| M-CHAT | Modified Checklist for Autism in Toddlers |
| RR | Relative risk |
| SES | Socioeconomic status |
| VBAS | Vineland Adaptive Behavior Scale |

## ABSTRACT

This dissertation considers the diagnostic category of autism as it emerges in a new national setting, Costa Rica. The data for this dissertation include interviews with physicians and staff at the National Children's Hospital (HNN) in San Jose, Costa Rica; a complete medical file review of all children who were assessed for autism from 2010-2013; 58 semi-structured interviews with parents of diagnosed children; and ethnographic data from the HNN and from a small city far from the hospital where there was recently a disproportionately high number of cases identified. The four chapters characterize the diffusion of childhood autism and address the more global patterns of autism diagnosis and diffusion. I address how the category is adopted and the first cases are diagnosed (Chapter 1), how cases are distributed across space and time (Chapter 2), what factors lead parents to be concerned for their children and then pursue professional help (Chapter 3), and what patient and clinical factors and characteristics lead cases to be referred to the diagnosing hospital (Chapter 4).

The findings show that a genetic study of autism was largely responsible for the early adoption of the category, as cases were initially recruited from a founder population surrounding San Jose. Interestingly, geographic clusters of cases soon appeared well outside of the early area of recruitment. Contrary to findings from the U.S., these clusters do not result from information

diffusion through parental social networks. Moreover, findings indicate that parents of all income and education groups are good at detecting early developmental anomalies. Diagnostic variation in Costa Rica appears to be driven by referral patterns between the public and private healthcare sectors. Specifically, parents with higher incomes are more likely to utilize the private sector, and compared to physicians in the public sector, private physicians have more face-time with patients and their families, leading to improved identification of developmental delays. The dissertation concludes by considering the role of sociology in an era of psychiatry that is open to the understanding of disease etiology and treatment in social organizational as well as biological terms. In future work, cross-disciplinary and collaborative work may be fruitful.

# INTRODUCTION

Medical sociology has long been critical of psychiatric authority and its increasing use of diagnostic classification to catalogue deviant behavior (e.g., Zola 1972; Horwitz 1982) – what Illich (1976) called the "medicalization of life." The criticism is certainly not without justification, as psychiatry has often appeared to have aims that are anything but helpful to the patient. For example, Benjamin Rush, the founder of U.S. psychiatry and a signer of the Declaration of Independence, coined the term "anarchia" to label political dissenters with a special form of insanity (Brown 1990). Rush's treatments for these supposed deviants involved cages, straps, and immobilization boards that evoke "The Machine" in the 1987 movie *The Princess Bride*.

Psychiatry's reach intensified after World War II (Szasz 1963) with the development of psychoactive drugs (Guimon 1989), beginning what Conrad (1975) referred to as the pharmaceutical revolution. At the same time, a process of institutionalization and a population of traumatized veterans with behavioral problems made precise categorization more important (Brown 1990). The Diagnostic and Statistical Manual of Mental Disorders (DSM), which was first published by the American Psychiatric Association in 1952 (Grob 1991), included 106

unique mental disorders, some of which were controversial even among physicians (e.g., Szasz 1960).

Sociologists (e.g., Belknap 1956) and anthropologists (e.g., Caudill 1958) criticized what was later hauntingly portrayed by Ken Kesey in his novel *One Flew Over the Cuckoo's Nest* (1962), where the contemptuous Randle McMurphy fakes insanity to avoid jail time for a battery charge, instead landing himself into a mental hospital. With persistent behavioral problems and frequent outbursts that infuriate the hospital staff, McMurphy receives increasingly invasive treatments that are meant to calm and control his behavior.

Fictitious or not, the patient McMurphy would have been an interesting case study for the "moral career" of the mental patient, which Erving Goffman (1959, 1961) coined to discuss how the moment of arrival at the hospital was in no way inevitable; rather, the labeling process for the mental patient, perhaps especially during the "prepatient phase," involves a series of unfortunate "contingencies" that lead to the eventual loss of freedom at the hospital. Goffman's work challenged psychiatry's insistence that mental illness existed within the individual, suggesting instead that the very idea of "insanity" allows medical professionals to decide what treatments are best for a patient, even at the patient's insistence of his own sanity or resistance to the professional courses of action (Szasz 1970). The extreme case is epitomized when the patient McMurphy, an obstinate but otherwise patently "normal" individual, is subjected to a lobotomy.

Even psychiatrists were concerned about the possibility of patients being incorrectly labeled, institutionalized, and treated. In a famous set of experiments, Rosenhan (1973) showed that one could gain admission into a psychiatric hospital and receive a diagnosis of schizophrenia simply by claiming to hear voices, even in the absence of other symptoms. Other research showed that the DSM was unreliable and clinicians were inconsistent with their assignment of

labels to patients (e.g., Spitzer and Fleiss 1974). Psychiatry was in a state of crisis, and the dominant psychoanalytic model was under attack (Wilson 1993).

One outcome was that psychiatrists began to debate how to improve the reliability of diagnostic procedures (e.g., Spitzer, Endicott, and Robins 1978), which culminated in the DSM revision in 1980 (Mayes and Horwitz 2005). For the first time, the writers moved away from psychoanalysis by aligning with a biomedical model of mental illness; specific diagnostic criteria were developed explicitly to improve the reliability of diagnosis (Wilson 1993). While some labels eventually fell out of favor and were removed,[1] subsequent revisions eventually increased the number of disorders by 2.5-fold. These developments were carefully followed by medical sociologists who were interested in the social factors of medicalization.[2]

**THREE EPIDEMICS**

The 1980 DSM revisions are at least partly to blame for three false "epidemics," including childhood bipolar disorders, attention deficit hyperactivity disorder (ADHD), and autism spectrum disorder[3] (Wykes and Callard 2010). Perhaps with the exception of autism, the patterns have been exacerbated by the marketing of drugs to treat the conditions (Frances 2013; Moynihan et al. 2002). The story of these false epidemics most certainly starts with the subjective character of the conditions, as they each rely purely on clinical judgment. Furthermore, each condition is marked by a long history of physicians, psychologists, and sociologists debating its essence.

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[1] For example, in the DSM-I, homosexuality was classified as a "sociopathic personality disturbance" (Conrad and Schneider 1980).

[2] Studies are available for mental illness (Scull 1975), alcoholism (Schneider 1978), schizophrenia (Mechanic 1972), and even for categories that were never included in the DSM but were "discovered" as forms of deviance, such as child abuse (Pfohl 1977). For more recent examples, see Jutel (2009, 2010).

[3] Throughout this dissertation, the term "autism" is used in lieu of autism spectrum disorder. This choice reflects the orientation and understanding of the interviewees.

For bipolar disorder symptoms, which are most generically thought to be the combination of depression and mania, discussions about their relationship and diagnosis go as far back as the Ancient Greeks (Marneros 2009). Like many other conditions, further advancements were thought to occur around the middle of the 19th century, in this case when a French psychiatrist wrote about the condition of "circular madness," or a movement among mania, melancholia, and typical behavior (Glovinsky 2002). In the early 20th century, the symptoms were an indication of "circular insanity," which was lumped into the broad category of manic-depressive insanity (Angst and Sellaro 2000), and some psychiatrists and psychoanalysts suggested that the condition occurred in children, albeit rarely (Renk et al. 2014).

It was not until 1966 that bipolar disorders really took shape in like fashion as today's diagnoses, as two seminal works argued for the separation of "unipolar" depression disorders, or what became to be known as major depression (Marneros and Angst 2000), and by the 1970s, discussions frequently included the possibility that the bipolar types were present and detectable in children (Glovinsky 2002). Finally, with the DSM-III in 1980, bipolar disorder was formally used to replace "manic depressive disorder," and the label was formally included as a category for children (Weller, Weller, and Fristad 1995).

Childhood bipolar disorder continues to be controversial among psychiatrists, in part because it was shown that the U.S. and U.K. assigned very different diagnoses for the same symptoms, where the U.S. was more likely to use the label bipolar disorder (Dubicka et al. 2008). In response, and even at the expense of validity, psychiatrists attempted to improve the reliability of the diagnosis (Horwitz and Wakefield 2007) by including sub-categories of bipolar disorder in 1987 (DSM-III-R, or "Revised" edition) and in 1994 (DSM-IV). Partially as a result, the rates of diagnosis began to increase: in the U.S. between 1994-1995 and 2002-2003, the rate

of bipolar disorders among children increased by 40-fold, from 25 to 1,003 per 100,000 (Moreno et al. 2007). Relatedly, since its inclusion in the DSM in 1980, drug therapy treatment for bipolar disorders, an example of increasing pharmaceuticalization (Abraham 2010), has increased by 50-fold (Healy 2006). Some authors have blamed this trend on the "disease-mongering" of the pharmaceutical industry (Moynihan and Cassels 2005; Moynihan et al. 2002; see also Barker 2011), which purportedly convinced many consumers that they are sick and in need of drugs.

The story of ADHD is not dissimilar to that of bipolar disorders in that an array of social factors, including a societal affection for impulse-control, as well as the preponderance for eventual drug treatments, were influential in the development of an epidemic (Lakoff 2000), false or otherwise. Bennett (2007) describes how as early as 1902, children with what is now known as ADHD were described as having "defective moral control." By 1937, amphetamines were somewhat successfully used to control these deviant behaviors, but it was not until 1957 that the behaviors were officially labeled "hyperkinetic impulse disorder." In 1966, this new disorder and several other sets of behaviors were classified under the umbrella term "minimal brain dysfunction" (Conrad 1975). Attention deficit disorder did not formally arrive until the DSM-III in 1980, and ADHD arrived with the DSM-III-R in 1987.

The ADHD category and drug therapies for the associated behaviors became more widely used in the 1990s, when as many as 1.5 million children were prescribed Ritalin (Lakoff 2000). In the same vein as the DSM critics in the 1950s and 1960s, many authors have expressed skepticism about the validity of the diagnosis as a neurodevelopmental syndrome, suggesting instead that it is simply a "cultural construct" (Timimi and Taylor 2003). Others, even going back to the 1970s, suggested that the hyperactive child was a "myth" that was propagated by the medical establishment as a form of social control (Schrag and Divoky 1975).

Physicians have responded to criticism of the ADHD diagnosis by arguing that the sociology of medicine is teaching "college students the horrendous social drama of the 'medicalization of deviancy'—the ongoing relabeling of behavioral difference (a normal temperamental variant) as medical disorder in order to place the patient in the grip of a malevolent medical industrial complex" (Accardo and Blondis 2001:7). For Accardo and Blondis (2001), the critiques are especially troubling because drug treatments have been shown to be markedly more effective than behavioral interventions for treating ADHD, and contrary to popular myths that parents are flocking to their doctors for drugs, parents *and their physicians* have been shown to be reluctant to rely on drugs (MTA Cooperative Group 1999). The Ritalin scare (see DeGrandpre 2000) is arguably a function of a preoccupation, common as well within medical sociology, with the supposed perils of science (Gross and Levitt 1998).

For autism diagnosis, which has also seen a dramatic increase in use, the behaviors we have come to associate with the condition were described long before the term was used (e.g., Melville's 1853 character "Bartleby"). The psychiatrist Leo Kanner (1943) was the first to use the word autism to describe the peculiar behaviors; his patient Donald, at 30 months of age, had been able to recite all of the presidents and vice presidents, but was "unable to carry on an ordinary conversation" (Kanner 1973a:93). The parents of the other patients like Donald thought their children were living in another world (Kanner 1973b), a metaphor that continues to be used to describe autism patients (see Hacking 2009).

In the 1950s and 1960s, autism was also one of three "epidemics" that psychiatrists, including Kanner, expressed concern about (mental deficiency, childhood schizophrenia, and autism; see Eyal et al. 2010:127-129). The concern was that the labels were commonly

misdiagnosed, in part because of the "understandable excitement about trying to solve the riddles" of the newly popularized conditions (Kanner 1971:19). Kanner (1973b:413) wrote:

> Almost overnight, the country seemed to be populated by a multitude of autistic children, and somehow this trend became noticeable overseas as well. Mentally defective children who displayed bizarre behavior were promptly labeled autistic and, in accordance with preconceived notions, both parents were urged to undergo protracted psychotherapy in addition to treatment directed toward the defective child's own supposedly underlying emotional problem.

To Kanner's relief, the autism label (and childhood schizophrenia) fell out of favor as the "brain-injured child" became the newest preoccupation among psychiatrists (Eyal et al. 2010). However, by the 1970s, networks of parents and advocates began to influence the clinical understanding of autism (Eyal 2013; Eyal et al. 2010), with some authors arguing that autism represented a "continuum" of behaviors (Wing and Gould 1979). Eventually, the continuum was thought to include extreme instances of normal behavior (e.g., Happe 1999), and by the DSM-IV, the "autism spectrum" was born (Wing 1997).

At the same time, in Europe and the U.S., deinstitutionalization was taking place. For children, this process was associated with increased "surveillance," where children who would have previously been wards of the state were now cared for at home; this meant that there was an increased interest in labeling and treating children with behavioral problems, and the process made room for "the proliferation of socially innovative therapies" (Eyal et al. 2010:64) that "blurred the boundaries between mental retardation and mental illness" (190). Autism became a unique label that was neither permanent like mental retardation nor stigmatized like mental illness. One result was that a process of diagnostic substitution—primarily from mental retardation (Shattuck 2006) and developmental language disorder (Bishop et al. 2008)—began to lead to new case reports, and in 1991, autism was included as a special education category. This

led to even more cases, and when major media outlets noted the changes in incidence, they reported an "epidemic" (Gernsbacher et al. 2005).

In 1998, not long after autism had become a household name, Wakefield published his infamous article—now retracted—blaming autism on childhood vaccines (Baker 2008). A flood of research dollars and media attention followed, and some states even adopted insurance incentives for early diagnosis; this led one National Institute of Mental Health clinician to say, "I'll call a kid a zebra if it will get him the educational services I think he needs" (Grinker 2007:131), making it clear that clinicians would willingly misdiagnose autism if they thought the resultant therapies would improve the child's development. Allegedly aware of this, scholars have argued that clinicians *and parents* are driving the continually increasing prevalence of autism (e.g., Liu, King, and Bearman 2010).

## SOCIAL CONSTRUCTIONISM

Unsurprisingly given the recent controversial history of psychiatry and the DSM, and often explicitly going back to Michel Foucault (1966) and Thomas Kuhn (1962), medical sociologists have long focused on the social and political contexts in which medicalization has occurred (e.g., Figlio 1982, 1978; Cooter 1982; Jewson 1974; Scott 1990). At times, these studies effectively bracket any biological factors in disease and illness. Many have focused on a social constructionist view of diagnosis, where in the extreme, the very rationality implied in "discovering" new diseases is placed essentially in a social context (e.g., Nicholson and McLaughlin 1987). As Sedgwick (1982:30) put it, "There are no illnesses or diseases in nature."

In the 1980s, medical sociologists were actively debating the value of social constructionism, with more positivist writers arguing that the critics of medicine had exaggerated the amount of social control that medicine exerts (Bury 1986), and that extreme relativism even

prevents the possibility of critical engagement with science and medicine (Jarvie 1983). In the debate between disease discovery (i.e., via medicine) and disease invention (i.e., social constructionism), King (1987) argued, using the history of "transexualism" as an example, that the invention argument is somewhat implausible and ignores the biological import of the condition (for which there is mounting evidence). In the middle of the two poles but in defense of social constructionism, Brown (1990:404) argued that psychiatry was unable to explain "why certain diagnostic categories appear and disappear over time," and that examples of changing categories "are essentially sociopolitical phenomena which are not comprehensible within the medical framework of diagnosis."

Potentially as a response to the increasing popularity of biomedical understandings of illness and disease (Busfield 2000), social constructionism has persisted in medical sociology, particularly within science and technology studies (e.g., Rees 2011). The result is that sociologists of diagnosis continue to voice or imply criticism for medical practice, and there is a tendency to belabor social pathways and mechanisms at the expense of biology, as if any mention of the latter is off limits to sociologists. In a recent special issue of *Social Science & Medicine* devoted to the sociology of diagnosis, Annemarie Jutel (2011b; see also 2011a) focuses on the social consequences of diagnosis, at times eschewing the role of biological mechanisms involved in an individual's condition. When a condition is discussed, such as obesity among an indigenous group, she argues that it "emerges from a context of post-colonization and globalization in which commercial and colonizing factors present the social group with more physical affronts than their waistlines or endocrine systems can handle. ... Big Food and Big Farm lobbies all converge to produce increased disease frequencies amongst Indigenous peoples"; she concludes that "we can see the limitation of conceiving of these diseases in the

simple pathophysiological terms" (Jutel 2011b:798), yet she also avoids discussing or considering any such pathophysiological aspects of obesity.

In other articles, Davenport (2011:873) explains how medical residents develop a series of "narrative templates" based on "'facts' about the patient and his or her complaint," and these templates are used as a form of "storytelling" that categorizes the patient's problem. Again eschewing the biological, Brown, Lyson, and Jenkins (2011:939) argue that "social diagnosis" refers to how diagnosis is connected to "a set of political, economic, cultural and social conditions or factors."

These arguments may very well be accurate, but they also avoid considering features unique to the actual conditions, including individual symptoms or problem behaviors that are relevant to patients and their families. And by bracketing the medical condition itself, these arguments are somewhat unhelpful for a diagnosing physician encountering the patient, who in many cases, has arrived at the clinic seeking medical solutions to legitimate problems.

The single-minded pursuit of social aspects of diagnosis can constitute a form of "biophobia" (Freese, Li, and Wade 2003:233), where sociologists tend to avoid discussions of the relevance of biology, thereby perpetuating the view that biology and sociology are "inevitably opposed." One way to avoid such an approach is to consider social aspects of diagnosis while also engaging with the health sciences, taking seriously biological variation that may have sociological consequences. For example, Conley and Bennet (2000) considered low-birth-weight infants – no doubt a category, especially for borderline cases, that has some social roots. The authors found that socioeconomic status (SES) does not predict low birth weights among infants, but even after controlling for SES, low birth weight is associated with lower educational attainment throughout the life course. This suggests that the mechanism driving the

diminished educational outcomes may be beyond the reach of a strictly sociological approach, possibly involving a relationship among nutrition, physiology, and early childhood development. For a diagnosis such as autism—where the subjectivity of the diagnosis means that clinicians continue to struggle for diagnostic reliability (see Lord and Jones 2012)—serious consideration must be given to behaviors and symptoms that may very well be biologically based.

## SOCIOLOGY OF DIAGNOSIS VIS-À-VIS EPIDEMICS

The aforementioned disease "epidemics" suggest an interesting topic of study that could consider biological, social, and even environmental aspects of illness and disease. To date, accounts of these epidemics have been in retrospect, providing only post-hoc explanations for how the categories emerged and then proceeded to spread throughout a population. And again, medical sociologists have tended to focus on the social dimensions of the consequent spread, rarely considering dimensions beyond the social[4] that could easily account for the spread of new cases.[5]

What is needed is an empirical setting in which a new category emerges and a potential epidemic begins. Such a setting would allow for the consideration of how the disease category is adopted, whether and how the symptomology was present in the population before the category emerged, how patients begin to consider the new disease, how old symptomology is translated into a new disorder, and the consequences of identifying cases in the population both on subsequent cases and on how the category evolves – all questions that have been considered in Jutel's (2009) call for a new sociology of diagnosis. Conrad and Schneider's (1980) five-stage model for how deviant behaviors are medicalized is a starting point, where behaviors are thought

[4] In autism research, Peter Bearman is an exception, as he frequently collaborates with health scientists and epidemiologists (see Cheslack-Postava et al. 2011; King et al. 2009).

[5] Of course, a likely reason for the over-emphasis on the social is the difficulty of obtaining relevant biological or environmental data.

to be (1) defined as deviant; (2) discovered within the medical community; (3) claimed to be appropriate for a unique label; (4) designated as an improved designation compared to old ones; and (5) institutionalized. No study has considered this process in a setting from the beginning, including how a medical category is adopted in the first place and before an epidemic emerges.

The present study does this. It focuses on how, in a country where the diagnosis of autism had not pre-existed, clinicians began to use the category, and then how it diffuses throughout the population over time. The study considers the following broad questions:

(1) How is the diagnostic label of autism adopted in a new setting?

(2) What social, biological, and environmental factors are relevant when clinicians and parents identify cases, and how are these cases distributed across space and time?

(3) How do diagnosed cases influence the identification of additional cases?

Several international studies of autism are relevant to this work: in China, there is evidence that parents know more about the category than physicians (Zhang et al. 2011), suggesting that "networks of expertise" (Eyal 2013) operate through select channels. Also in China, parents of children who are later diagnosed experience long delays in receiving a diagnosis, both because there are few services for autism, and because cultural factors lead parents to attribute language delays to high intelligence (Sun et al. 2013). In India, physicians in some areas are reluctant to diagnose autism for fear that parents will not accept the diagnosis (Daley 2004), suggesting that expertise alone does not determine which clinicians use the category. For parents, early problems in their children—especially when the behaviors emerged in public settings—were influential in how the parents developed their own networks of expertise with extended family and physicians (Desai et al. 2012).

In Korea, South Africa, and elsewhere, autism is underdiagnosed and services are mostly unavailable (Grinker et al. 2013), but the only prevalence study using a population sample (and U.S. diagnostic criteria), in a small city in Korea, has suggested that the prevalence of autism (1 in 38 children) is actually much higher than was previously thought (Kim et al. 2011). In the U.S., the implication is that even the latest Centers for Disease Control rate of 1 in 68 is low (Centers for Disease Control 2014).

No research has suggested that the distribution of autism symptomology varies across populations. In a range of settings, including Colombia, India, Jamaica, Jordan, and Mexico, the symptoms of primary concern to parents, as well as biological features[6] of cases of autism, have been shown to be similar (Samms-Vaughan 2014). But rates of diagnosis are consistently variable: autism is more likely to be diagnosed in children from high SES families in urban areas (e.g., in Denmark, see Larsson et al. 2005; Sweden, Sivberg 2003; the U.K., Taylor, Jick, and MacLaughlin 2013; Baird et al. 2006; the U.S., Durkin et al. 2010; King and Bearman 2011; and elsewhere, Grinker 2007). Thus, lower income families in rural areas have lesser chances of receiving a diagnosis and the important services that have become associated with the label (Liptak et al. 2008).

## THE DISSERTATION: AN OUTLINE

The data for this dissertation include interviews with physicians and staff at the Hospital Nacional de Niños (HNN); a complete medical file review of all children who were assessed for autism at the HNN from 2010-2013, including medical histories, referral pathways, and address data; 58 semi-structured interviews with parents of children who were receiving follow-up at the HNN or who received a diagnosis in 2013; and ethnographic data involving observations and

[6] In her review, Samms-Vaughan (2014) did not specify what was meant by “biologically-based factors.”

interviews at the HNN and in local referring clinics, a special education school, and a small city two hours from the hospital where there is a disproportionately high number of cases of autism.

This dissertation includes four chapters that characterize the global diffusion of childhood autism in Costa Rica, including how the category is adopted and the first cases are diagnosed (Chapter 1), how cases are distributed across space and time (Chapter 2), what factors lead parents to be concerned for their children and then pursue professional help (Chapter 3), and what patient and clinical factors and characteristics lead cases to be referred to the diagnosing hospital (Chapter 4). The dissertation concludes by considering the implications of this work for developing a sociology of diagnosis in psychiatry and medicine.

The first aim of the dissertation is to describe how the category of autism is adopted in a new setting and how a potential epidemic emerges there. While Eyal et al. (2010:70) briefly considers how autism, as a "truly global phenomenon," involves "the spread of a network of expertise," little is known about how expertise specifically leads to the adoption of the category. Are physicians trained to use diagnostic instruments in the U.S.? Does the pharmaceutical industry lead the charge (e.g., Lakoff 2005) of identifying autism? How do networks of expertise operate (e.g., Eyal et al. 2010), and where does knowledge about the category come from? How do these networks lead to the first cases of autism? Chapter 1 considers these questions in Costa Rica, where all that is known about the adoption of the category is that a genetic study of autism (McInnes et al. 2005) was conducted in the capital, San Jose, and a public information campaign was conducted to recruit cases in the early- to mid-2000s. Thus, the first case of autism may have been diagnosed for the genetic study, although it is unclear how additional cases were recruited outside of the genetic study.

Chapter 2 focuses on spatial patterns of cases of autism to determine whether the aggregate patterns of cases are similar as those found in the California studies (e.g., Mazumdar et al. 2010). The chapter combines Costa Rica census data with data on all diagnoses of autism in Costa Rica from 2010-2013, which is between five and eight years after the information campaign stopped explicitly recruiting cases. The chapter follows several U.S. studies that have identified geographic clusters (e.g., Mazumdar et al. 2010; Hoffman et al. 2012) and is the first inquiry to identify geographic clusters outside of the U.S. setting. Several studies suggested that the primary cause of the clustering phenomenon is related to parental action (e.g., Liu, King, and Bearman 2010), where some parents but not others seek medical care for their symptomatic children. Other studies have suggested that clustering is caused by environmental factors such as toxicants from air pollution (Becerra et al. 2013) or coal power plant emissions (Lewandowski et al. 2009). However, data from the U.S. come from hundreds or thousands of clinics that are variably distributed across space (e.g., Mazumdar et al. 2013), and these clinics engage in highly uneven diagnostic practices (Bresnahan, Li, and Susser 2009; Charman et al. 2009). Costa Rica provides a less noisy setting because there is only one diagnosing clinic for young children and the diagnosis is relatively new. Chapter 2 begins at the country scale and moves to smaller scales, concluding at the neighborhood level, to identify spatial patterns of autism diagnosis that are similar to those found in the U.S. The primary aim of Chapter 2 is to assess the "risk" of diagnosis (e.g., Larsson et al. 2005) across space and time, thereby allowing for the possible detection of (a) information spread through social networks and (b) environmental toxicants that could cause autism clusters.

Chapter 3 relies on interview data from the 58 in-person interviews with parents of diagnosed children and ethnographic data from an area with a geographic cluster of cases to test

an increasingly familiar hypothesis that is meant to explain the variable detection of cases in the U.S. What is referred to as the *cul-du-sac effect* (e.g., Liu, King, and Bearman 2010) is taken for granted as a primary mode of the social spread of autism diagnosis. The idea is that parents who acquire information about autism from other parents or families have an increased likelihood of identifying and developing concern for symptoms and reporting the concerns to clinicians (Weintraub 2011). A problem with this theory is that investigators have not directly spoken to parents and the data remain anecdotal (e.g., Grinker 2007:159; for a similar hypothesis for ADHD, see Conrad 2005). Additionally, geographic clusters of cases can be explained by a number of factors, including the variable distribution of diagnosing clinics, variation among clinicians in propensities to diagnose borderline cases, and variation in insurance coverage.

The aim of Chapter 3 is to describe how cases in my sample of families are related and whether they recognize symptoms and decide to seek help (Mechanic 1961, 1975). In the spirit of Goffman's (1961) work on mental patients, the approach is to bracket concern for individuals with similar symptoms who never arrive at the hospital for a possible diagnosis. Notably, the diagnosis is not predestined or even entirely predictable based on certain behaviors or symptoms. Rather, a series of contingencies explains patients' pathways to the clinic, where various events and encounters with concerned friends, family members, and clinicians are essential to the eventual diagnosis (and in accounting for it after the fact). Thus, Chapter 3 focuses on how patients move through formal and informal referral networks before eventually arriving at the diagnosing clinic (Friedson 1960, 1961; Horwitz 1977; for a review, see Pescosolido, McLeod, and Avison 2007), considering both lay and professional social networks among parents of diagnosed cases. The sample allows for the consideration of two low severity[7] geographic

[7] Low severity cases are the very ones whose parents should be the most likely to require information to know to pursue help (see Liu, King, and Bearman 2010).

clusters from Chapter 2 that are not explained by SES or education, including an apparent sudden increase in prevalence in 2013 in a small city nearly two hours from the diagnosing clinic.

Chapter 4 moves to the professional networks of referral that appear to explain much of the clustering phenomenon. Specifically, focusing on the interviewed sample, multiple regression is used to predict two factors: the age at the time of symptom recognition, and the time between symptom recognition and arrival at the diagnosing clinic (i.e., referral time). For the former, older mothers and families of children with an older sibling are associated with an earlier age of first symptom recognition, and neither maternal age nor older siblings are associated with symptom severity. For the referral time, parents and patient characteristics do not predict the arrival at the clinic, providing further evidence that parental action is not the primary explanation for the aggregate patterns. Chapter 4 then focuses on the low severity cases in the interviewed sample, where it seems that more cases than expected are being referred from the private sector. The chapter argues that low severity cases are more likely to be identified in private clinics and schools (where there are lower teacher-to-student ratios) than in public clinics, suggesting that differential patterns of scrutiny can lend to the explanation of aggregate patterns in diagnosis. In public clinics, even though physicians are equally well trained as those in private clinics, appointment times are short, and it seems apparent that parental concerns may not be articulated such that, in turn, referrals are less likely.

This dissertation concludes by considering how these findings implicate the development of a sociology of diagnosis in a medically informed era of psychiatry. For autism specifically, Costa Rica may be similar to other low- and medium-income countries that are considering the new diagnosis, where there are many undiagnosed cases that would qualify for a diagnosis if only they were assessed, and the primary determinant of such an outcome is simply the arrival at

a diagnosing clinic. For parents in Costa Rica, knowledge about the disorder may be analogous to knowledge levels in the 1970s or 1980s in the U.S., whereas for clinicians, the standards of diagnostic precision are in line with those currently in the U.S. and Western Europe. What can a setting such as this reveal about the spread of diagnostic categories? What does this mean for the development of increased diagnostic rates? For the sociology of diagnosis, the answers may suggest caution in the tendency to belabor the importance of social constructionism or the role of medical authority (e.g., Jutel 2011b). Instead, sociologists can focus on the rather mundane features of diagnostic processes; for autism, this means focusing on how parents and clinicians come to regard developmental anomalies in children as a medical disorder and specifically apply autism as the relevant diagnostic category.

It is possible that parents and clinicians simply do not associate classical autism symptoms with autism, such that those symptoms may not be in their "symptom repertoires" (Shorter 1986, 1987) but only later become associated with autism as they learn more about the disorder. While this may be the case, the present research indicates that identifying those classical autism symptoms may be of less importance than was previously thought (e.g., Liu, King, and Bearman 2010): consistent with epidemiological studies in different locations (Samms-Vaughan 2014), this dissertation shows that parents and clinicians initially are concerned with generic, non-autism-specific symptoms, such as language delay. An autism expert likely would classify these generic symptoms as related to autism, but the primary orientation is to provide these children with medical and therapeutic treatments to remediate the problems and place them on a better developmental track. However, once clinicians have learned about the autism diagnosis, in Costa Rica the cultural reliance on professional authority to solve medical puzzles (e.g., Low 1988) places these clinicians in a pivotal place. Their role in the

diffusion of autism seems to be more crucial than “networks of expertise,” which have been shown to be a main mechanism in the U.S. (Eyal et al. 2010)

This is not an argument on behalf of social constructionism or the medicalization of deviance. Rather, it is to recognize that, as psychiatry has changed dramatically since the 1960s and 1970s, biomedical approaches to various psychiatric conditions have made considerable progress. Medical sociologists should retain an awareness and even emphasis on social pathways but without ignoring or even bracketing biological or environmental phenomena. This dissertation concludes by discussing the role of the medical sociologist in the era of biomedicine.

# CHAPTER 1

## The Emergence of Autism in Costa Rica

To date, research has only speculated about how "epidemics" materialize, with only one research team (Eyal et al. 2010) considering the diffusion of the autism diagnosis specifically, although only in the U.S. The aim of this chapter is to describe how the category of autism was adopted in a new setting, Costa Rica, beginning with the first diagnosed case and including the early spread of the label. Costa Rica may be representative of many countries worldwide in which the diagnostic category has only recently been or will soon be introduced into a population that is not well informed about the label, thereby making the population an unlikely source of the early spread. Thus, this chapter considers how physicians initially begin to consider the label. To use language from Eyal et al. (2010), how do "networks of expertise" (Eyal et al. 2010) operate in the Costa Rican setting, and how much are those networks tied to existing networks in the U.S.?

### NETWORKS OF EXPERTISE

Eyal et al. (2010) detail how networks of expertise influenced the diagnostic category of autism. The idea is that the recent "epidemic" of autism was not possible until persons with mental retardation were deinstitutionalized, such that a "new institutional matrix" of parents and

professional other than psychiatrists worked as advocates for these children. By the 1980s in the U.S., children who were diagnosed with mental retardation, which is now known as intellectual disability, were increasingly assigned a label of autism – a process referred to as diagnostic substitution (Shattuck 2006). Thus, this story of networks of expertise in the U.S. eventually involved changes in how the diagnostic category of autism was conceptualized, and these changes, including deinstitutionalization itself, may not have been possible without psychiatry's crisis involving the role of psychoanalysis in treatment.

Recall that it was in the late 1960s that the autism authority Bruno Bettelheim blamed autism on "refrigerator mothers," and in his now-scandalous extended description of autism, he even suggested that having autism was akin to being a prisoner in a Nazi concentration camp (Bettelheim 1967). In his book, he claimed that several patients with autism had been cured by his psychoanalytic methods – claims that were later refuted.[8] But importantly, Bettelheim was not the only one to blame mothers for various conditions in children; the field of psychiatry had been doing so since at least the 1940s (Ornitz and Ritvo 1968), as had the originator of the diagnosis, Kanner (1943). In our own *American Journal of Sociology*, Bettelheim (1959:455) argued that children with autism from middle-class parents must have experienced "extreme emotional deprivation" that could be compared to "the traumatic experiences" of children reportedly reared by wolves.

Several prominent figures displayed strong resistance to blaming parents for their children's problems, perhaps especially including Bernard Rimland, a psychologist, researcher, and father of a child with autism. Rimland (1964a,b) agreed with Kanner that autism was a rare condition and argued in adamant defense of mothers who were being blamed by the likes of

[8] Notably, after Bettelheim committed suicide in 1990, some of his former patients argued that instead of being "cured" of autism, they had had unrelated behavioral problems that led to being treated by Bettelheim, who, it turned out, did not even have a psychology degree (Severson, Aune, and Jodlowski 2008).

Bettelheim and mainstream psychiatry. Citing Kanner's work, he noted that out of 42 early cases that received psychotherapy, 29 did not improve, and among the 13 cases that did improve, psychotherapy was either not used or was not "good" psychotherapy. Even before Rimland, authors were arguing that the tendency to blame parents rather than genes or biology was simply a product of psychiatry's insistence that its methods were effective (e.g., Lewis 1954; see Feinstein 2010).

Rimland is central to Eyal et al.'s (2010) history of networks of expertise because his resistance to mother-blaming led to a concerted effort, involving an alliance with Kanner, to destigmatize autism and to stop blaming parents for the condition. The new aim was to treat the disorder with a range of therapies *other than* psychoanalysis, and the new question was whether children were responsive to the therapies.

At the same time as these alliances were being formed, a process of widespread deinstitutionalization was taking place in the U.S. (a similar process was already underway in much of Northern Europe), which in the first place involved an increased "surveillance of childhood" (Eyal et al. 2010:64). Instead of becoming entities of the state, children with a range of behavioral problems remained at home. In a widely influential book, Wolfensberger et al. (1972) emphasized that individuals previously deemed to be mentally deficient should instead be integrated into mainstream society. This "normalization" movement created space for therapies and interventions that would transform problem children into functional, normal children, and that broad aim created room for new forms of expertise involving parents of diagnosed children who had seen success in their efforts to integrate and normalize their own children. In short, state psychiatrists were no longer the experts on these children; their parents and therapists were.

These new experts began to have a considerable influence on mainstream psychiatry. In the 1980s, the early diagnostic checklists for autism suggested that the diagnosis involved a spectrum of behaviors that could even involve opposites: one child could be completely nonfunctional and nonverbal while another could display savant abilities; one could be over-sensitive and another under-sensitive; one could be quiet and detached and another hyperactive (Eyal et al. 2010).

In addition to the broadening of the category in psychiatry, the alliance of parents even influenced popular portrayals of autism in Hollywood, including the 1988 movie *Rain Man*. Minor celebrities were made of individuals with autism and Asperger's, including Temple Grandin, the Colorado State University professor who transformed the design of slaughterhouses by claiming to have—as a result of her autism—an uncanny ability to empathize with cattle. Her case showed that typical, classical autism symptoms during childhood could evolve into a fully functional and productive set of behaviors in adulthood. Nobody argued (least of all Grandin) that the condition had disappeared; instead, Grandin became a prototype of how autism was an alternative way of experiencing the world, a new identity and a new way of being.

The key to this spectrum of behaviors—and the key difference from mental retardation[9]—was that it was treatable. Early detection was imperative because the children *could be* included in mainstream schools; with the help of therapies, they *could be* typical children. The alternative way of seeing the world, now called autism, only required new sets of skills, new "behavioral 'tools for living'" (Eyal et al. 2010:268), to function normally in the world. Of course, the networks of expertise, now including parents, were motivated to continue conceptualizing autism as a treatable condition, keeping it in a space between mental illness and

[9] Note that the mental retardation label was only recently changed to "intellectual disability" with the DSM-5, but the diagnostic criteria for the new label continue to overlap with the autism label.

mental retardation where, unlike what these diagnoses entailed, there could be hope for improvement. An unintended outcome of the associated diagnostic reclassifications was a rapid increase in the number of included cases.

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The role of networks of expertise in the expansion of the autism label in the U.S. is one part of an emerging story of the medicalization and globalization of autism. Eyal et al. (2010) emphasize the importance of the historical context in these networks: if not for a critical movement against mainstream psychiatry and psychoanalysis, culminating in a massive deinstitutionalization in which former and would-be patients of psychiatric hospitals began to be cared for at home, these networks of expertise may not have emerged or had such an impact on the autism label. Absent these historical forces or the networks themselves, we can only speculate about how autism would have changed over time. Nonetheless, we can consider how autism continues to emerge and evolve in other settings.

The category of autism now appears in countries in all six continents—making it "a truly global phenomenon" (Eyal et al. 2010:70)—but no research has considered how the label first appears in a setting and then begins to spread as clinicians increasingly apply it to patients. This chapter considers the case of Costa Rica, where all that is known about the emergence of the category is that a genetic study of autism (McInnes et al. 2005) was conducted in the capital, San Jose, and cases were recruited in the early- to mid-2000s. Thus, the first case of autism may have been diagnosed for the genetic study, although it is unclear how additional cases were recruited outside of the influence of the genetic study. What did the setting and its patients look like before autism? How was the first case of autism diagnosed, and under whose influence? How do networks of expertise operate in relation to the emergence of the category? Are the experts of

autism tied to networks of expertise in the U.S. and Europe? Where does knowledge about the category come from, and what is the motivation to begin and then continue diagnosing cases?

**METHODS**

Before proceeding with answers to these questions, it is worth noting how this project materialized and detailing the methods that are used throughout the dissertation. This is of special importance in understanding the emergence of the autism label in Costa Rica because it tells us something that is perhaps uniquely Latin American, or at least unique to small low-to-medium-income countries, in terms of how professional networks of expertise may function. Specifically, professional networks are tight, relatively closed networks in which most people have strong connections, primarily because there are relatively few people with advanced degrees, and they are more likely than not to live in the centralized metropolitan areas in and around San Jose. These small, tight networks also mean that certain characters are deeply important in the story or autism, including the local "institutional sponsor" of the present study, Dr. Patricia Jimenez Gonzalez.

* * * * *

The original aim of the present chapter was to understand how social networks of parents related to the diagnosis. Did parents influence other parents to obtain a diagnosis for their children? Was information about autism necessary for parents to pursue a diagnosis? How does that information spread, and what are the consequences? Essentially, the project aim was to learn how autism spreads through a setting in which it was not previously in use.

My first suspicion that Costa Rica would be an appropriate study site for the spread of autism was after reading a genetic study of autism in the journal *BMC Psychiatry* (McInnes et al. 2005). The study was led by L. Alison McInnes, the research director at the Neurobehavioral

Genetics Laboratory at the Mount Sinai School of Medicine in New York, and included two researchers from the University of Illinois at Chicago. One of those researchers, Elina Manghi, originally from Argentina but working as a clinical psychologist at a bilingual autism clinic, recruited Jimenez to identify and recruit local cases suspected of autism. At the time, Jimenez was the director of the developmental unit at the HNN and became the lead clinician on the study after completing Dr. Catherine Lord's[10] certification course on the Autism Diagnostic Observation Schedule (ADOS), the gold standard diagnostic instrument for autism.

The 2005 article suggested that Costa Rica would be an ideal setting for a study on the diffusion of autism, especially including the possible effects of information spread on the detection of new cases. Costa Rica appeared to provide many advantages for such a study: (1) the setting was small,[11] so it was conceivable that population data would be available; (2) in Illinois, Manghi had developed a model for training clinicians and special education teachers to identify high functioning cases of autism, and she had taught Jimenez this training model, who was thereby spreading information about autism to important actors; (3) Jimenez had connections with the Autism Parents Association in Costa Rica and had worked to educate its members; and (4) autism was being formally diagnosed with a standardized instrument, the ADOS (McInnes et al. 2005). So it seemed that autism had arrived in Costa Rica, and Jimenez was leading the way to recruit cases for the genetic study.

* * * * *

Born in Costa Rica in 1955, Jimenez received her medical degree from the University of Costa Rica in 1980. She completed her residency at the HNN from 1981-1983, specializing in

[10] Catherine Lord is a leading expert on the autism diagnosis and also works at Mount Sinai.

[11] Two alternatives, Mexico and Brazil, also regularly diagnosed autism, but a systematic study of the diagnosis would have been difficult given their size.

pediatrics. During that time, she was interested in alleviating problems associated with dehydration from diarrhea – still a leading cause of death among children and infants in rural communities. From 1984-1985, she worked as an assistant pediatric specialist in the emergency department, and then from 1985-1986, she completed a second residency in developmental pediatrics at the Boston Children's Hospital. Back in Costa Rica, she worked part-time in the neurology unit and taught a course on early stimulation until 1990, becoming head of the developmental unit starting in 1991. At that time, she advised two medical students studying attention deficit disorder, and she often saw cases in the clinic.

In 2000, she temporarily returned to New England to complete a Master's in Education from Framingham State College. By that time, the autism label had only recently gained global prominence following mention of an epidemic in the 1990s and the blaming of vaccines in 1998. As a pediatrician following U.S. standards of practice (as do all Costa Rican physicians), these developments were likely on her radar. After all, she had an impressive resume with pediatric experience at top hospitals in Costa Rica and the U.S.

* * * * *

When I finally met Jimenez, I explained that I was in Costa Rica working on my Spanish and considering a study on autism for my dissertation. I told her that I hoped to focus on Costa Rica because I was developing a social network study, and I hoped to focus on a setting where there were a small number of cases, which was different from a recent study in the U.S. (Liu, King, and Bearman 2010) where they had discovered "clusters" of cases of autism.

Her immediate concerns about a study were that she did not want more parents learning about autism and potentially seeking a diagnosis. She mentioned a six-month to one-year waiting period for children that needed to be assessed, and she was concerned that parents learning about

autism would want something in return; the HNN, though, had nothing to offer. She suggested posting something on the hospital website. However, I called her attention to the fact that the HNN website was barely functioning, and I doubted that patients relied on it at all. I suggested that this strategy might miss parents with no internet, and she agreed. I asked her how many cases she had, and she showed me her computer database with 114 from the last two years. She said she currently had far too many cases coming in, as she wanted to retire soon. Our discussion turned to personal themes, and we ended the meeting by agreeing that I would send a short proposal by email.[12]

I wrote a proposal to interview parents of children with autism, initially intending to sample the entire roster of parents in the previous two years. I wrote an interview schedule that focused on incoming and outgoing social network connections among these parents: who they knew with autism before their child was diagnosed, who (if anyone) influenced them to notice symptoms, who referred them to the HNN, and finally, who they subsequently influenced to seek medical care for autism symptoms.

The sampling design for the interviews became the predominant concern as I communicated with Jimenez. One of the initial stumbling blocks was obtaining an accurate list of all patients who were diagnosed over a known period of time, as the hospital database was difficult to work with and often failed to indicate the diagnosis. However, Jimenez maintained Word documents of all patients who had undergone an ADOS assessment since 2009, so we compiled a list of these children. Next, Jimenez assisted me with the process of obtaining bioethics approval through CENDEISSS, the Center for Strategic Development and Information on Health and Social Security. For subject recruitment, Jimenez offered to have her secretary call

[12] My previous attempts to contact her had failed because I had sent emails to an address listed with the HNN domain, which she did not use.

parents, tell them briefly about the study, and ask them to come to the hospital for an interview. She had an empty office that I could use for the interviews.

After submitting a proposal to CENDEISSS and an English version to the health sciences internal review board at the University of Wisconsin (the approval process took half of 2012 and the first months of 2013), we attempted some pilot interviews. It became immediately clear that many of the phone numbers were no longer in service, and many of the parents lived very far from the hospital. There was no way to achieve a reasonable response rate with this recruitment method, so the proposal was amended. The eventual strategy was to interview the parents of all new cases of autism and all cases with autism that visited the developmental unit for follow-up.

From one to four times per week starting in 2013, Jimenez would call me when she learned of a possible case, and I would arrive at the hospital for the interview. At the end of each appointment, Jimenez would inform the parents about the study and ask if they were willing to speak with me in an adjacent office. All parents were enthusiastic about the study, as they were interested in learning more about autism and were happy to share their experiences obtaining a diagnosis. All parents agreed to participate and signed consent forms after hearing further details about the aims of the study.

Interviews were conducted with the assistance of a research assistant, Pedro Solis, a Master's student in psychology from the University of Costa Rica. Interviews were conducted in Spanish, and Solis was available to assist with interpretation when needed. Both Solis and I took detailed notes throughout each interview and compared and combined our notes immediately after the interview was completed.

Because of the network design, we also obtained approval from CENDEISSS to review the medical files of all families from 2010-2013 to crosscheck these names with those provided

by the interviewees. For these cases, we also obtained approval to record address data at the level of district (to search for diagnostic clusters) and several variables for symptom severity. Importantly, the symptom severity variables would allow us to compare the symptoms of the interviewees' children—some of whom were diagnosed prior to 2010—with those of the population of diagnosed children from 2010-2013. Because the interviewees were not randomly sampled, symptom severity would be one way to check for potential sampling biases.[13]

For the CENDEISSS bioethics approval, Jimenez became the study's official institutional sponsor, and the agreement was that she would be the second author on all publications from the study. Now as a co-author, she took me under her wing to learn about how the developmental unit worked, and I interviewed her regularly about the genetic study, the early cases of autism, and how information was spread to other clinicians. I also became a regular fixture at the HNN, often eating lunch with her and other physicians there, and I attended several conferences at the medical school where she gave presentations about autism. At the hospital, I also interviewed the developmental unit staff, psychologists, and speech therapists, and I observed ADOS assessments for two children.

During this time, I continued to live in Turrialba, a town of approximately 30,000 people in the mountains east of San Jose. I eventually spoke with psychologists, special education teachers, and clinicians in the Turrialba area, and I talked about autism whenever possible.[14] Interestingly, Turrialba is located directly within a spatial cluster of cases that involved a rapid increase in diagnosis in 2013, the same year that I lived full-time at CATIE.

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[13] The most plausible bias would occur if severe cases attended more follow-up appointments than milder cases, which would make the interview sample more severe, on average, than the population of cases. This turned out not to be the case.

[14] Because I was clearly an outsider, I was typically asked some version of "what are you doing here?" when I spoke with people in the community. I would answer, "I am a sociologist, and I study autism," which almost always led to a discussion about autism (academics and clinicians were also interested in why a sociologist would study autism).

Finally, in addition to interview data with Jimenez, the present chapter involves a complete review of all genetic studies that were conducted on human populations in Costa Rica leading up to the 2005 autism study. Including the first study in 1992, there were 18 such studies, and they focused on conditions including deafness, Louis-Bar Syndrome, bipolar disorder, Tourette's Syndrome, Bartter's Syndrome, and schizophrenia (for a review, see Mathews et al. 2004). A list was compiled of all 108 authors involved in the 19 studies (including the autism study), and their affiliations were tracked using information from the articles or, when possible, biographical information available online. Then, the software Visone (Brandes and Wagner 2004) was used to create a two-mode network of connections among the study authors, where each of the studies is shown as the center of 19 subnetworks (see Figure 1). This set of connections will be an important part of the story of the first case of autism in Costa Rica: The network diagram in Figure 1 shows the central figures in the genetic studies in Costa Rica, and it shows how the autism study is connected to a network of authors in Costa Rica and in the U.S. In short, Figure 1 displays the networks of expertise that eventually precipitated the diffusion of the autism label in Costa Rica.

**Figure 1** Author networks in the 19 genetic studies focusing on human populations in the Central Valley of Costa Rica, 1992-2005 (articles in white).


## THE FIRST CASE

Prior to the informal diagnosis of "patient zero" in 1999, Jimenez had occasionally noted in the hospital case files when a child exhibited symptoms that were suggestive of autism, but she was unwilling to formally diagnose autism because she had not received training to do so. The 1999 case was especially indicative of autism (in addition to persistent developmental delays, the child began making stereotypical movements with his head before 12 months of age), so when she returned to Boston for her Master's, she delivered a video of the child to one of her colleagues from her residency there. This colleague confirmed a diagnosis of autism.

Furthermore, the colleague had connections with Mount Sinai, and news made its way to McInnes that the head of the developmental unit at the HNN was interested in autism.

Some background on McInnes is essential to this story: McInnes originally graduated from Stanford but completed her medical degree in 1991 at Columbia and then returned to the West Coast in 1995 to the University of California at San Francisco for her residency in psychiatry. She remained in San Francisco for six years, consulting with a pharmaceutical company (Millennium Pharmaceuticals) and working as an adjunct professor at UC-SF. In 2001, she returned to New York for a faculty position at Mount Sinai. In the puzzle of autism in Costa Rica, this background may be the most important piece.

Back at UC-SF in the 1990s, McInnes became affiliated with the Institute for Human Genetics, and a team of researchers there was interested in Costa Rica because of a "founder" population there. Founder populations are populations that were founded by a relatively small number of individuals and then grew over many generations with little mixing with other populations. Because these populations tend to be somewhat genetically homogenous, they provide one way to overcome extreme genotypic and phenotypic variability that is especially prevalent in humans, especially when multiple genetic pathways are suspected to contribute to a rare condition of interest. After ascertaining precisely when a population was established, often with the help of a genealogist, one can estimate the effect of heritability – especially for conditions with higher-than-average prevalences in the founder populations.

Founder populations are themselves rare: in the Western hemisphere, there is a population of Mennonites in the U.S., which has been used to study Hirschsprung's disease (Puffenberger et al. 1994), a population in Antioquia State in northwest Colombia, a population in the Oriente region of southeast Guatemala, and the population in the Central Valley of Costa

Rica (see Mathews et al. 2004).[15] The founder population in Costa Rica includes much of the population surrounding San Jose and can be traced back to fewer than 90 families who migrated from Spain to Guatemala to Costa Rica beginning in 1561. This original population mixed with the indigenous populations and then remained relatively isolated, geographically and politically, for the following four to five centuries (Escamilla et al. 1996).

The use of genetic work on human populations is relatively new. It was only in approximately 1980 that methodological advances allowed researchers to conduct "genetic mapping" studies in humans (Altshuler, Daly, and Lander 2008), and in 1983, genetic mapping was used to localize a specific disease (Huntington's disease; Gusella et al. 1983); by the late 1980s, there were approximately 100 disorders mapped to specific genes, and there was increasing interest in localizing new disorders and conditions (Altshuler, Daly, and Lander 2008), with many studies starting to exploit founder populations for their work.

For Costa Rica specifically, there were several relevant research groups involved in the early efforts to localize disorders in the founder population there. Locally, there was the Centro de Biologia Molecular y Celular with the University of Costa Rica, which published an early genetic paper on deafness (Leon et al. 1992); there was the department of psychiatry at UCLA,[16] which linked up with an immunologist at the HNN on a study of Louis-Bar syndrome (Uhrhammer et al. 1995); there was the Department of Medicine at the University of Alabama in Birmingham, which published a study on Bartter's syndrome (Kurtz et al. 1997); and there was the group in San Francisco that included McInnes.

[15] There is also a founder population in Finland that has been used to study autism (Auranen et al. 2000, 2003).

[16] The UCLA department also had connections with the UC-SF department, and there is substantial movement among faculty between the two departments. However, none of the early studies included authors from both institutions.

By the time McInnes arrived at UC-SF in 1995, researchers there were already using Costa Rica's founder population to study bipolar disorder (Escamilla et al. 1996; Freimer et al. 1996a,b) and Tourette's (Mathews et al. 1996). They had partnered with the Centro de Biologia Molecular y Celular, and one of those authors, Eduardo Fournier—a historian and genealogist—also worked at the HNN. Thus, McInnes had her hands in the early genetic studies in Costa Rica by 1995, and when she moved to Mount Sinai in 2001, she became a member of a team with extensive experience and interest in autism, and the team had connections with the University of Illinois autism clinic that conducted outreach to Spanish-speaking families.

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McInnes learned of autism being diagnosed in Costa Rica shortly after she moved to Mount Sinai, and she led the efforts to coordinate between the team in New York, the clinic in Illinois, and the HNN. First, she contacted Jimenez and encouraged her to receive formal training on using the ADOS. Jimenez agreed, receiving training in California and additional training from Catherine Lord at Mount Sinai. Soon thereafter, Jimenez began using the ADOS to diagnose her own patients at the HNN.[17]

While Jimenez coordinated the bioethics approval for the study, McInnes obtained a research project grant from the National Institute of Neurological Disorders and Stroke. At the same time, Manghi began teaching Jimenez the training model she had developed in Illinois.

[17] Jimenez and two other physicians also diagnose autism at their private practices, but the number of patients who pay for a full assessment is small, and most of those patients receive a formal diagnosis through the public system as well, as a diagnosis (not necessarily of autism) must be obtained from the HNN to be considered for special education or financial support from the Costa Rican government. For Jimenez, the majority of patients who are suspected of autism arrive by way of the public system. For the other two private practices, the diagnoses of autism are informal and primarily affect children older than those included in the present chapter, although being seen by one of those two private physicians is a relatively common pathway to the HNN for a subsequent ADOS assessment.

Then, Jimenez began searching specifically for individuals who had developmental delay (including language delay) and significant disruptive behaviors in school or at home.[18]

For the genetic study, the team would need cases with a range of autism severity; however, the early cases that Jimenez identified were more severe than what predominates in cases outside of Costa Rica, so the team developed a strategy to recruit low severity cases with normal or near normal IQ (see McInnes et al. 2005). The plan was to offer training sessions to pediatricians, psychiatrists, and special education teachers so that these professionals could assist in recruiting difficult-to-identify cases of low severity autism. Jimenez discussed the project with the Education Minister, and she began promoting early symptom identification of autism at the medical school. She also spoke on television and made regular radio appearances in 2002 and 2003, and she visited the special education schools in the San Jose metropolitan area to discuss autism with the teachers, psychologists, and administrators.

By the end of 2003, autism was being consistently diagnosed at the HNN, and it seemed there was no turning back. Now there were parents and clinicians and teachers who had all heard of the disorder, and soon enough, there was a waiting list to undergo an ADOS assessment at the HNN, the only public clinic that offered the assessment.[19] Jimenez eventually compiled a list of 245 patients with symptoms of autism, and most of these cases were located within the study area where the founder population was known to live. If birth records for the cases could verify that six out of eight great-grandparents were born in the Central Valley, the cases were included

[18] The team suggested identifying cases with these two criteria, as more than one study had suggested a link between mild autism cases and symptoms of attention deficit hyperactivity disorder (Keen and Ward 2004; Sturm and Fernell 2004).

[19] It is unclear whether the waiting list was generated internally, where cases already at the hospital were selected to undergo an ADOS assessment, or externally, where new cases from the outside were seeking or being referred for an assessment. Evidence compiled during and after the interviews suggest that much later, it was the former: the waiting list was internally supplied; from 2003-2005, though, cases may have been referred directly as a result of the recruitment efforts.

in the genetic study. The team eventually identified and invited 76 cases for participation, and 70 cases were enrolled in the study.[20] Before the team used up its limited funding, Jimenez had reviewed 35 initial cases.[21]

**AUTHOR NETWORK CONNECTIONS**

Another way to trace the diagnosis of autism in Costa Rica is to consider the genetic studies that preceded that of the 2005 autism study. Figure 1 reveals several patterns in the network connections of the genetic study authors. First, seven of the autism study's nine authors were involved in no other studies, and they are far from the main cluster of studies and authors. The autism study is connected to the main cluster via the historian, Fournier (30, node in red with a green border shown in the insert), and McInnes (60 in light blue), whose node is directly next to Fournier's. Fournier is the only author affiliated with the HNN within the main cluster.

The autism study is near the 1992 study, the first genetic study in Costa Rica, which involved a small team at the Centro de Biologia Molecular y Celular; both of the 2005 and 1992 studies are somewhat far from the main cluster of 10 studies, and they are near to one another because of node 50 (near Fournier and McInnes). Node 50 represents an author on the 1992 study (from the Centro de Biologia Molecular y Celular) who was involved in 11 studies, nine of which Fournier was also involved in. Finally, of the 19 studies, four are entirely unconnected to the main cluster, including both the UCLA and University of Alabama studies.

[20] First, Jimenez administered the Autism Diagnostic Interview-Revised (ADI-R) to the parents of children who were suspected of autism, which is one diagnostic tool but based on parental reportage, and then the ADOS was administered to the children by Jimenez or a trained psychologist in a private office at the HNN. The ADOS was videotaped and independently scored by at least two team members who had undergone ADOS training.

[21] Of those 35 cases, 24 displayed language delay, 12 had first- or second-degree relatives with language delay or learning problems, and 21 had involved obstetric complications or difficulties with labor. Thirty-one of 35 were male, and the average age at diagnosis was just under 7 years of age. Additional data on these children and their families were limited.

Fournier and McInnes also provide links to a genetic study in 2001 on bipolar disorder (McInnes et al. 2001), and the study included eight authors from Millennium Pharmaceuticals, where McInnes had provided consulting work while she was at UC-SF. Contrary to the situation reported in Lakoff's (2005a, 2005b) account of how international pharmaceutical companies influenced the category of bipolar disorder in Argentina, considering all genetic studies in Costa Rica, the pharmaceutical involvement was very limited. In fact, it was McInnes that initiated their involvement, but it was limited to only one study, and their influence on the other genetic studies in Costa Rica appears to be minimal.

There are other differences between Argentina and Costa Rica. By the time these studies appeared in Costa Rica, the diagnoses were well established, and many of the physicians in Costa Rica had been trained in the U.S. and tended to be well informed on U.S. diagnostic criteria. This was not the design of pharmaceutical or corporate interests; rather, physicians in Costa Rica go out of their way to be exposed to the most up-to-date information in their fields, and the medical schools and hospitals in the U.S. continue to be the best places to be trained. Thus, the label of autism did not change when the genetic study appeared; instead, a process of diagnostic substitution was established, where cases of mental retardation or attention deficit disorder were diagnosed with autism instead.

## CONCLUSION

So was autism exported to Costa Rica by clinicians in the U.S., or was it imported by clinicians in Costa Rica? Both accounts are partially accurate. What is clear from Figure 1 and the biographies of many of the genetic authors is that starting with either Jimenez or the autism study fails to provide a complete picture. The process began long before the autism study, potentially with the Centro de Biologia Molecular y Celular, which is only one of two human

genetic departments at the University of Costa Rica. Costa Rica's emphasis on medical genetics traces back to the 1970s, but the emphasis on tracking particular diseases only traces back to 1991 (Saborio 1992). The genetic researchers, which include physicians, microbiologists, statisticians, and cytogeneticists—all of whom were following U.S. practices—are in some ways at the root of the eventual inclusion of autism as a diagnostic label.

The genetic researchers' influence may not have been possible had it not been for Costa Rica long following the U.S. model of psychiatry, which is similar to what Lakoff (2005b) describes in Argentina. This means that when diagnostic changes occur in the U.S., they are adopted in Costa Rica as a matter of course. I recall seeing Jimenez's excitement when the new DSM-5 arrived in the mail at the hospital: she always saw it as her role to diagnose children as correctly as possible, and she was always up-to-date on advances in autism research (i.e., the now defunct link between autism and vaccines). Indeed, she was unwilling to diagnose autism until she received training on the gold standard diagnostic instrument.

In other locations, such as China, the dominant model of medicine can look very different from that of the U.S., and there is arguably less sharing of information and expertise between physicians and hospitals. Lee (2000, 1996) even documented how patients with anorexia nervosa actually experienced the disorder differently after China began to adopt a Western understanding of anorexia (e.g., involving a "fat phobia"). Arthur Kleinman reported a similar phenomenon with patients with depression in China (for a review of "cultural psychopathology" research, see Lopez and Guarnaccia 2000). For Costa Rica, because it had long followed the U.S. model of psychiatry, patient experiences were not drastically different after the advent of the genetic studies, although this theme will resurface in Chapter 3 when the interview data are presented.

The U.S. psychiatric model aside, autism would not have arrived in Costa Rica when it did had it not been for Jimenez, and for that matter, it may have been delayed had McInnes not initiated the genetic study and encouraged Jimenez to complete the ADOS training. However, there is little doubt, given the influence of U.S. psychiatry, that others would have eventually diagnosed autism there. Thus, it is safe to say that the use of autism was a consequence of following the U.S. psychiatric model, of being informed by best practices in genetics and psychiatry, and in being at the forefront of studies on humans populations.

**AUTISM AFTER 2005**

Interestingly, the genetic study on autism came to a halt soon after the 2005 publication, as the study did not receive additional funding. Although plans to continue the genetic analyses are said to be on hold,[22] Jimenez continues to consider her regular patients for later inclusion in the study. However, explicit efforts to include low severity cases for the study are no longer pursued. Jimenez continues to regularly educate physicians at the medical school (she had been involved as an educator there before she began speaking about autism specifically), and she is well known in the small medical community as being the autism expert in the country.

The HNN does not provide support for physicians in the developmental unit to diagnose autism, but Jimenez is able to commit one day every week to conducting ADOS assessments. Given that there are many weeks when this is not possible, Jimenez assesses 30-40 children per year, and several of those cases are not diagnosed with autism. Thus, after the initial 245 cases, there has likely been an additional 300-500 cases (35 cases over 10-12 years), making the entire

[22] The genetic study will likely never be pursued, as an important team member in the U.S. recently passed away.

population of children with autism in Costa Rica well below 1,000 cases.[23] Prior to the present study's efforts, little was known about this population or their parents.

[23] The HNN only began using the category of autism in the computer records after 2007, and then only on a limited basis for several years. Thus, it is impossible to precisely estimate the prevalence of autism in Costa Rica.

# CHAPTER 2

## The diffusion of autism in Costa Rica

In the U.S., children with autism have been found to live in spatial clusters (Mazumdar et al. 2010; Hoffman et al. 2012). From a public health perspective, these clusters mean that children in certain neighborhoods—often overrepresented by Latinos, African Americans, and low SES families (Durkin et al. 2010)—are less likely than others to receive important services that can improve life outcomes (Liptak et al. 2008). Identifying the causes of the clusters will inform public health officials on how to remediate these inequities and inform us on "contextual drivers" of the increasing prevalence of autism (Mazumdar et al. 2013).

Theories about the causes of the autism clustering are diverse. For example, sociologists have found strong positive correlations between the diagnosis and both SES and parental education (Van Meter et al. 2010; Hoffman et al. 2012), and others have found anecdotal evidence of a social network effect on the diagnosis (Liu, King, and Bearman 2010). In California, autism prevalence in low SES neighborhoods nearly caught up to the rate in high SES neighborhoods, and this happened as information about autism was becoming widespread (King and Bearman 2011). In other studies, biologists have identified associations between autism clusters and congenital malformations of the reproductive system, which can be caused by

exposure to toxicants during pregnancy (Rzhetsky et al. 2014). Others have linked the spatial patterns to exposure to heavy metals (Windham et al. 2006), air pollution from traffic (Becerra et al. 2013), and mercury from coal-fired power plants (Palmer et al. 2006; Palmer et al. 2009).

Determining the cause of the clustering is difficult in the U.S. setting for several reasons. Specifically, there is substantial variation in access to healthcare (Kirby and Kaneda 2006; Wang and Luo 2005; Williams 2012). This has been documented for autism specifically (see Liptak et al. 2008; Fountain and Bearman 2011). There is also variation in the distribution of diagnosing clinics (e.g., Mazumdar et al. 2013), and within the clinics, there are variable diagnostic practices (Bresnahan, Li, and Susser 2009; Charman et al. 2009) that are partly affected by insurance incentives (Grinker 2007). All of these factors would likely create spurious correlations with neighborhood and demographic characteristics (e.g., Kirby, Taliaferro, and Zuvekas 2006).

Another difficulty in the U.S. setting, particularly for hypotheses suggesting that information diffusion through parental social networks leads some parents and not others to seek a diagnosis for their children (e.g., Weintraub 2011; Keyes et al. 2012), is that information about autism is readily available from many sources (Eyal et al. 2010). While there is evidence that parents rely on other parents for information *after* their children are diagnosed (Mackintosh, Myers, and Goin-Kochel 2005), it is unclear whether these parents make contact *before* the diagnosis. Other sources of information about autism, such as resources on the internet, would not directly lead to spatial clustering.

The objective of the present chapter is to identify and explain spatial patterns of autism diagnosis in young children—a group that has seen dramatic increases in prevalence in the U.S. (Keyes et al. 2012)—in a setting in which the diagnosis has not been widely publicized, there is no variation in healthcare access or diagnostic practices, and there are no insurance incentives for

autism (see McInnes et al. 2005). These factors allow us to effectively control for several potentially misleading, cluster-producing variables.

In our study site, Costa Rica, the diagnosis of autism was first used in 1999, and fewer than 1000 children have been diagnosed since then. Information about autism is not easily available on Costa Rican websites, and there is very limited information about autism in media outlets. In fact, according to our unpublished surveys, most parents have not heard of the diagnosis, and many clinicians are not aware of autism symptoms. However, children in Costa Rica have universal healthcare that is relatively accessible and equitably distributed (Rosero-Bixby 2004; Savedoff 2009), and for diagnostic services, all children under six years old are referred to one centralized clinic, the HNN. Thus, any clustering in Costa Rica will implicate factors other than variation in healthcare access or diagnostic services.

The first such factor is an information effect. While people tend to know little about autism, an information effect is still likely in Costa Rica, as the period of focus in this chapter is five years after a small-scale, targeted information campaign was employed to locate cases for a genetic study on autism (McInnes et al. 2005). The study targeted the founder population that is limited to the Central Valley of Costa Rica, of which the capital, San Jose, is on the eastern edge. The campaign targeted clinicians in close proximity to the HNN by organizing seminars about the diagnosis, and teachers in special education schools were also contacted to identify potential cases. Clustering around these clinics could indicate an information effect, but clustering outside the Central Valley, where the special education schools were not contacted by the campaign, would likely indicate a different clustering mechanism; any clustering outside the Central Valley would likely not be a result of information spread from the campaign. Alternatively, if the information campaign caused clustering, then there should be an increased risk of autism near

the HNN and potentially near daycare centers, preschools, and special education schools where teachers have learned to identify symptoms.

Also related to an information effect, because early symptom recognition is difficult, information spread would lead to earlier diagnosis and the identification of less severe symptoms (Liu, King, and Bearman 2010; King and Bearman 2011). If information spread through professional or parental networks is the cause of clustering, then children living inside a geographic cluster will have received an earlier diagnosis and will exhibit less severe symptoms than children living outside the cluster.

The second factor that could be implicated in the case of clustering is an environmental mechanism that led to more cases in exposed areas. Importantly, there are no coal-fired power plants in Costa Rica, which are significant emitters of mercury and have been linked to spatial patterns of autism diagnosis (Palmer et al. 2009). In terms of previously identified environmental factors, the most plausible mechanism for an environmental agent in Costa Rica is air pollution (e.g., Becerra et al. 2013), and because of the mountains that surround the densely populated Central Valley, the highest levels of air pollution are spatially concentrated near the HNN (Barrientos 2010). Therefore, any clustering patterns that exclude the HNN (and west of the hospital) are likely indicative of non-environmental factors. Clustering patterns that include the HNN could still be explained by travelling distance or urbanicity, which has been associated with the diagnosis (Mandell, Novak, and Zubritsky 2005).

The final factor that could be implicated in the case of clustering is population density. If population density were to explain all the variation in diagnosis, there would be no evidence of social or environmental causes of the clustering.

This chapter begins on an exploratory note by searching for spatial clusters of autism diagnosis among children. Then, we use census data and hospital records of the diagnosed children to identify and rule out potential causes of the clustering patterns, first by predicting cases at the smallest census scale and then by focusing on smaller scales.

## MATERIALS AND METHODS

This project was approved by the bioethics committee at the HNN. The data for the study originate from two sources: (1) district-level census data from the Costa Rican National Institute of Statistics and Census (Instituto Nacional de Estadística y Censos, INEC); and (2) four years of hospital records (2010-2013) of children who were assessed with the ADOS at the HNN. The hospital records included date of birth, sex, age at the time of first words, household district at the time of diagnosis, date of diagnosis, Modified Checklist for Autism in Toddlers (M-CHAT) and Vineland Adaptive Behavior Scale (VBAS) scores for autism screening, developmental age, and diagnostic outcome.

We excluded children who were older than five years at the time of screening, as a subgroup of older children is referred to a separate hospital. Also, because the diagnosis and severity of autism are determined at the time of assessment, we chose not to exclude cases based on the assessment outcome. Thus, cases were defined as all individuals who were screened for autism at the HNN before the age of six and who were later assessed using the ADOS between 2010 and 2013. Using the M-CHAT and VBAS, the screening process was completed in the developmental unit by a nurse who was trained by the primary diagnosing physician. For the years of focus, there were 127 1-5-year-old patients who underwent an ADOS assessment; nine cases were excluded because of missing address data, so the final sample was 118.

Before identifying district-level clustering, we tested the covariates that could predict the presence of cases in a district. Out of 468 districts, 90 had at least one case, and the maximum number of cases was 6. Thus, the distribution was heavily weighted to zero. We determined that there were not excessive zeros (which would occur if older children were included; see Coxe, West, and Aiken 2009), and the dataset was not overdispersed, so Poisson regression was used to predict district-level cases.

The covariates included at-risk population, percentage of households below poverty, percentage of the population over 15 years old with at least a secondary (middle school) education, percentage of the population in urban areas, distance to the HNN, percentage of children under five years old attending daycare or preschool, and population density. With the exception of distance to the HNN, all values were obtained from the INEC website (http://www.inec.go.cr; accessed on May 15, 2014) from the 2011 census.

At-risk population was calculated by counting backwards by the average age at the time of the ADOS to determine the birth year for each assessment cohort (2005, 2006, 2007, and 2008); for each birth year, we used INEC data to determine the number of births in each district (the total "at-risk" population for all four years was 290,335; min: 15, max: 5596). We expected that at-risk population would be an important predictor of diagnoses at the level of district, but an association with at-risk population says little about causality. Therefore, at-risk population was used as the null model.

We also determined the district size, and as a proxy for travel time, we used geocoded district centroids in ArcGIS 10.1 to calculate straight-line distances to the HNN. Stata 13.1 was used to calculate descriptive statistics, correlation and regression coefficients, pseudo $R^2$,

Bayesian Information Criterion (BIC) and deviance goodness-of-fit values, and incident rate ratios (IRRs).

To test for district-level clustering, we used Kulldorff's Spatial Scan Statistic (Kulldorff 1997) with the software SaTScan (Kulldorff 2009), which was also used in the California studies (see Mazumdar et al. 2010). Briefly, the software assumes a Poisson distribution to identify the "most likely cluster" of cases in a geographic area, using scanning windows to compare relative risks (RRs) across space. For each scanning window, the software maximizes the likelihood function based on the observed and expected (given the at-risk population) numbers of cases and the total number of cases in Costa Rica. After the most likely cluster is identified (the window with the highest log likelihood ratio (LLR)), Monte Carlo simulations are used with a Lehmer random number generator to repeatedly redistribute the data across the space, each time identifying the most likely cluster. To calculate an unbiased p-value, 999 simulated LLRs are compared with the observed LLR. The p-value is simply an indicator of where the observed LLR ranks among the 1,000 total LLRs, so a p-value of 0.001 indicates that the observed LLR is higher than all of the simulated ones.

We began by conducting a purely spatial test, and we compared the medical data of cases inside and outside the most likely cluster. If enough parents within the cluster were exposed to information that prompted them to act, we would expect to observe a lower average age of diagnosis and a lower average age of symptom recognition compared to the underexposed areas; relatedly, "exposed" children may display less severe symptoms, as parents may be more aware of symptoms that would have otherwise gone unrecognized.

To explore whether parents inside the cluster identify symptoms earlier and consult a clinician sooner than parents outside the cluster, we used t-tests to identify differences in the age

at the time of autism screening and diagnosis inside and outside the cluster. As proxies for symptom severity, we compared the age at first words, the difference between the developmental age and chronological age (lower values indicate closer-to-normal development), and the ADOS score. Finally, we used a chi-square test to compare four diagnostic outcomes (not diagnosed, low severity autism, moderate severity autism, and high severity autism) between cases inside and outside the cluster. To further explore differences in symptom severity, and given that an information effect may be more prevalent among parents of children with mild symptoms (King and Bearman 2011), we conducted an additional spatial test with only low severity and not diagnosed cases.

Next, we used the "temporal trends" feature of SaTScan to search for trends over time and across geographic space. The tool is effective for locating areas in which the number of cases is temporarily low but later catches up with the country-level rate, which can mean that the RRs do not differ on a global level. These types of patterns could be apparent if a social or environmental mechanism began to affect parents arriving at the HNN at an identifiable point in time. For this feature, we used increasingly small scanning windows with population ceilings of 10%, 5%, and 2% of the at-risk population. Because of the district shapes around the highly populated areas, we used ellipses instead of circles (the results were similar).

To conclude the cluster detection at the level of district, we repeated the detection tests by controlling for the best Poisson regression model, thereby controlling for the meaningful covariates. For this step, we used Stata to calculate the expected number of cases in each district, and we used these data to conduct a covariate-adjusted scan for clusters (see Klassen, Kulldorff, and Curriero 2005). We conducted the scan (a) for all cases, (b) for only low severity and not diagnosed cases, and (c) with temporal variation.

For an information effect to operate, parents would need to live in close proximity. Thus, to consider an even smaller geographic scale, we returned to the medical files of the cases in the San Jose metropolitan area and extracted the following information: referral information to the HNN and the developmental unit, dates and clinical notes from all HNN appointments, and address information over time.

We used the address at the time of screening to identify a precise address for each case. Addresses in Costa Rica are often relative to local landmarks, so in several cases, we identified the local landmark by asking around in the neighborhood and then using a GPS device to obtain the UTM coordinates, which we located in Google Earth before identifying the address to within approximately 50 meters.

There are no population data smaller than the district level, so we began by creating an ArcGIS raster of the included districts. Next, we created a 100×100 meter grid and numbered each gridpoint (n=60,283). Because San Jose is bordered by mountains and several of the districts are largely uninhabited, we identified uninhabited areas in Google Earth and removed them from the raster. Assuming an even population distribution, we divided the district populations by the number of district gridpoints and assigned those values across the entire district.

The San Jose metropolitan area is smaller than the cluster detected by Mazumdar et al. (2010), so the detection of spatial patterns at this small scale may be especially indicative of the contextual drivers that were discussed by subsequent authors (e.g., Liu, King, and Bearman 2010). We searched for the SaTScan window with the most likely cluster for (a) all cases (n=46) and (b) only low severity and not diagnosed cases (n=19). We also searched for the window with

the most likely area of *low rates*, which simply maximizes the likelihood function for a RR below 1. Finally, we reviewed the medical file referral information for the identified clusters.

**RESULTS**

*District-level patterns*

Using bivariate Poisson regression to predict district level cases, the pseudo $R^2$s for poverty (0.07), distance to the HNN (0.06), and daycares (0.02) suggest limited improvement over the intercept model; the pseudo $R^2$s for education (percentage over 15 years old with at least middle school education) (0.11), urbanicity (0.15), and population density (0.09) are slightly higher. Notably, poverty is strongly correlated with education (r=-0.80), urbanicity (r=-0.67), distance to the HNN (r=0.57), and daycares (r=-0.63), so districts with many households living in poverty tend to be much less educated, live in more rural areas, and have fewer children attending daycare. Education is strongly correlated with urbanicity (r=0.79), daycares (r=0.59), and population density (r=0.58).

Considered together, the preferred model for predicting cases includes urbanicity rather than poverty or education. However, urbanicity is likely a stand-in for patterns of poverty and education, as the HNN is located in an urban center associated with relatively high levels of education, low levels of poverty, and a high percentage of children in daycare.

Four models are shown in Table 1, with model 3 being the preferred model based on likelihood ratio tests. Model 3 includes at-risk population, urbanicity, and distance to the clinic. Of the remaining variables, daycares improved model 3 the most but not enough to warrant the additional variable. For model 3, the IRRs show that for every 100-person increase in the at-risk population ($\bar{x}$=620), the expected rate ratio of total cases increases by 0.06, so approximately 1,700 additional children would suggest one additional case in the district; likewise, each

additional percentage of households in an urban area ($\bar{x}$=52.44%) would increase the expected rate ratio by 0.02, and each additional kilometer from the HNN ($\bar{x}$=60.65 km) would decrease the expected rate ratio by 0.01, so the effect sizes are relatively small. The pseudo $R^2$ is 0.25, but most of the improvement is accounted for by the at-risk population (bivariate pseudo $R^2$=0.18).

**Table 1** Poisson regression coefficients for various models of the effect of selected covariates on the presence of cases of 1-5-year-old children suspected of autism in 468 administrative districts of Costa Rica, 2010-2013 (SEs in parentheses).

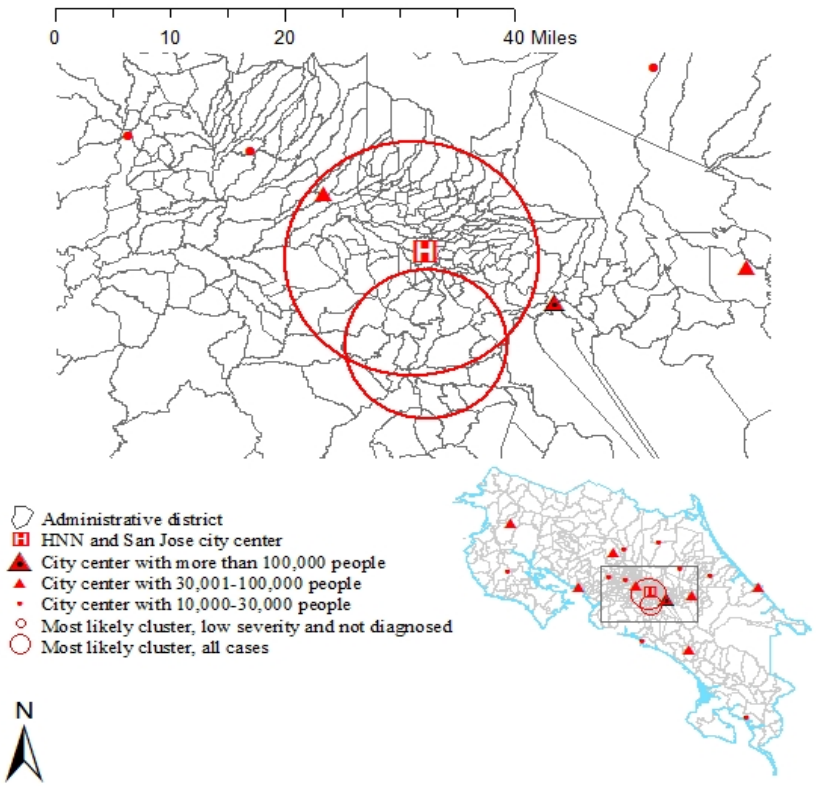
| Variable | Model 1 | Model 2 | Model 3 | Model 4 |
|---|---|---|---|---|
| At-risk population[a] | 0.072*** | 0.054*** | 0.059***[b] | 0.056*** |
| | (0.005) | (0.006) | (0.006) | (0.007) |
| Percent urban | | -0.020*** | 0.014***[c] | 0.016*** |
| | | (0.004) | (0.004) | (0.005) |
| Distance to the HNN (km) | | | -0.006*[d] | -0.007* |
| | | | (0.003) | (0.003) |
| Percent daycare/preschool | | | | -0.028 |
| | | | | (0.024) |
| Intercept | -2.091 | -3.339 | -2.728 | -2.729 |
| Pseudo $R^2$ | 0.181 | 0.239 | 0.247 | 0.250 |
| Log likelihood | -250.69 | -233.12 | -230.51 | -229.79 |
| BIC | 513.68 | 484.68 | 485.62 | 490.32 |
| Likelihood ratio $\chi^2$ | | 35.15*** | 5.21* | 1.45 |

*p<0.05; **p<0.01; ***p<0.001.
[a] For greater ease of interpretation, we divided the at-risk population by 100 (range: 0.15-55.96).
[b] IRR: 1.060 (0.007).
[c] IRR: 1.015 (0.004).
[d] IRR: 0.994 (0.025).

Turning to SaTScan, we identified a large area surrounding the HNN in which the population has 2.55-times the risk of undergoing an ADOS assessment compared to the population outside the area (p=0.002) (Figure 2). When only the low severity and not diagnosed cases are considered (n=41), a cluster of cases is centered in south San Jose, no longer including the HNN, and the cluster involves a six-fold increased risk (RR=6.18; p=0.001). The most likely

cluster for only moderate and high severity cases (n=77) is not statistically significant (RR=2.33; p=0.232; cluster not shown).

**Figure 2** Most likely country-wide cluster of all 1-5-year-old cases suspected of autism (large circle), and most likely country-wide cluster of only low severity and not diagnosed cases (small circle), Costa Rica, 2010-2013.




With the available assessment data, we report the differences between inside and outside the largest cluster in Table 2. We found no evidence that patients in the cluster were younger at the time of autism screening or diagnosis than children outside the cluster, but there is some evidence of less severe symptoms inside the cluster, as the age at first words is slightly earlier (20.89 vs. 24.74 months; p=0.070). Also, there is a smaller difference between the developmental age and chronological age (32.28 vs. 38.34 months; p=0.008) and a lower ADOS score (13.11 vs. 16.09; p=0.003) among cases inside versus outside the cluster, respectively.

**Table 2** Comparison of means and standard deviations of various clinical features[a] between ADOS assessment cases inside (n=74) and outside (n=44) the most likely spatial cluster of cases, Costa Rica, 2010-2013.

| | **Inside cluster** | **95% CI** | **Outside cluster** | **95% CI** | **t-test (1-sided)** |
|---|---|---|---|---|---|
| Age at first words | 20.89 (11.44) | 18.18-23.60 | 24.74 (15.45) | 19.73-29.75 | t=1.49 p=0.070 |
| Age at screening | 45.04 (12.54) | 42.11-47.97 | 46.10 (11.70) | 42.45-49.74 | t=0.44 p=0.329 |
| Age at ADOS | 60.82 (15.34) | 57.27-64.38 | 58.18 (12.64) | 54.34-62.02 | t=-0.96 p=0.832 |
| Developmental difference | 32.28 (13.22) | 29.08-35.48 | 38.34 (12.08) | 34.67-42.01 | t=2.45 p=0.008 |
| ADOS score | 13.11 (5.53) | 11.83-14.39 | 16.09 (5.71) | 14.36-17.83 | t=2.80 p=0.003 |

[a] All units except ADOS score are months.

Finally, the chi-square test indicates a relationship among the diagnostic categories between cases inside and outside the cluster (p=0.023), where there are more low severity and not diagnosed cases inside compared to outside the cluster (Table 3).

**Table 3** Percent of cases in four diagnostic categories among ADOS assessment cases inside (n=74) and outside (n=44) the most likely spatial cluster of cases, Costa Rica, 2010-2013.[a]

| **Diagnostic category** | **Inside cluster** | **Outside cluster** | **Total** |
|---|---|---|---|
| Not diagnosed | 16.2% (n=12) | 11.4% (n=5) | 14.4% (n=17) |
| Low severity autism | 23.0% (n=17) | 15.9% (n=7) | 20.3% (n=24) |
| Moderate severity autism | 41.9% (n=31) | 27.3% (n=12) | 36.4% (n=43) |
| High severity autism | 18.9% (n=14) | 45.5% (n=20) | 28.8% (n=34) |
| **Total** | **100.0% (n=74)** | **100.1% (n=44)** | **99.9% (n=118)** |

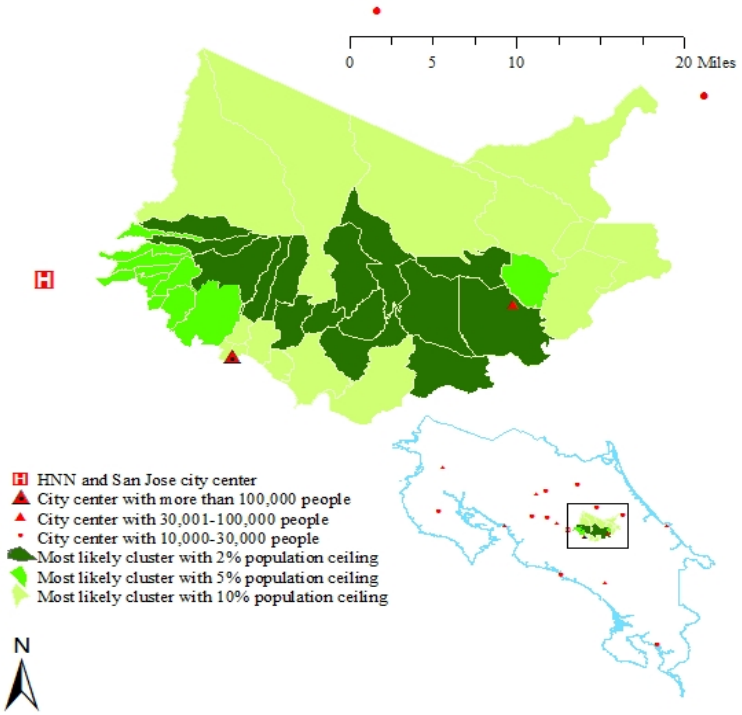
[a] Pearson $\chi 2$ (df: 3): 9.489; p=0.023.

Regarding temporal patterns, there is little variation in the numbers of assessments each year, as physicians at the HNN are limited to one day per week for ADOS assessments. Among

the included cases, ANOVA tests show that the differences across years are not significant for age at first words (p=0.118), time between screening and ADOS (p=0.555), age at ADOS (p=0.282), difference between developmental and chronological age (p=0.596), or ADOS score (p=0.330).

With the temporal trends feature in SaTScan, the elliptical scanning window (Goujon-Bellec et al. 2011) provides a slightly better fit than the circular window. With a 10% population ceiling, we identified an area (which includes all three shaded areas in Figure 3) that went from having two to 16 cases in one year (p=0.001). Thirteen of the new cases are included in the 5% population ceiling (p=0.004), and seven are included in the 2% ceiling (p=0.028) and occurred during a three-month period of 2013. These cases are outside the Central Valley, and because of the mountainous roads, involve more than a two-hour bus or car commute to the HNN.

**Figure 3** Most likely temporal-spatial clusters of 1-5-year-old cases suspected of autism, Costa Rica, 2010-2013.

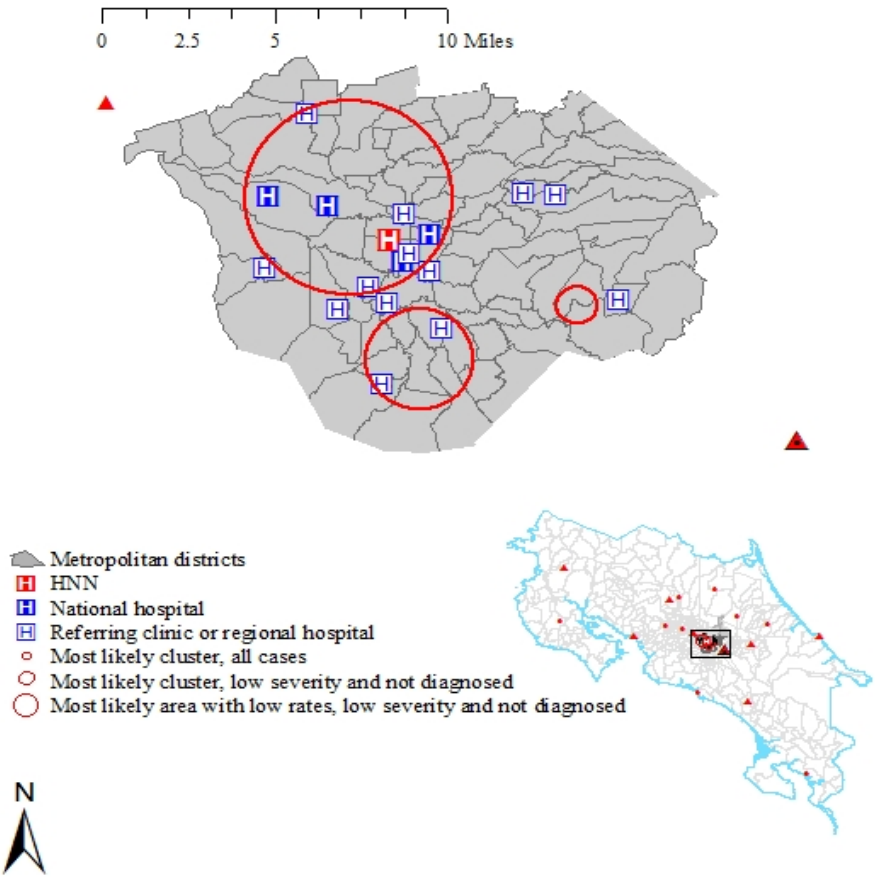


Finally, we used the Poisson regression results to scan for covariate-adjusted areas of increased risk. Considering all cases, the most likely cluster is no longer significant (p=0.482), but the cluster of low severity and not diagnosed cases remains significant (p=0.014). Similarly, the temporal trends do not substantively change after adjusting for urbanicity and distance to the HNN ($p<0.05$). These results suggest that the primary cluster in the country is largely accounted for by variation in urbanicity (potentially a proxy for SES) and distance to the HNN, but the cluster of low severity cases and changes over the four-year period remain unexplained by these covariates.

*San Jose metropolitan area*

We next turned to the San Jose metropolitan area to search for clustering at a smaller scale and around the HNN, using precise addresses for the 46 cases between 2010 and 2013. The cases involve several instances of "neighbors," where parents live within 500 meters of each other and may have occasion to make contact and share information. Figure 4 displays the clusters for (a) all cases (RR=18.05; p=0.086), (b) only low severity and not diagnosed cases (RR=5.77; p=0.166), and (c) low severity and not diagnosed cases but with low rather than high rates (Expected cases: 5.56; Observed cases: 0; p=0.041).

**Figure 4** Most likely San Jose metropolitan area clusters of 1-5-year-old cases suspected of autism for (a) all cases (small circle), (b) only low severity and not diagnosed cases (medium circle), and (c) all cases but with low rather than high rates (large circle), Costa Rica, 2010-2013.




The cluster of all cases includes several cases in a small neighborhood, and the cases involve different referring physicians. In the cluster of low severity and not diagnosed cases on the south side of San Jose, which is a concentrated area of the low severity and not diagnosed cluster in Figure 2, the cases followed different referral pathways. Only two cases attended the same clinic, but they were referred by different clinicians, for different reasons, and with no mention of autism.

For the low-risk cluster, a concentrated group of referring clinics, all the national hospitals, and the HNN are located within the area, suggesting that very close proximity to the major hospitals or the area with the highest air pollution is not associated with an increased risk

of diagnosis. This pattern is inconsistent with an information effect originating in these clinics after the public information campaign about autism.

**DISCUSSION**

The present chapter found evidence of spatial clustering of autism diagnosis in Costa Rica, where the diagnosis has been used for little more than 15 years. The setting presented a unique opportunity to study the distribution and early diffusion of the diagnosis when there is no variation in diagnostic practices, children have universal healthcare, and two potential environmental toxicants—mercury (from coal-fired power plants) and air pollution—are absent or concentrated in a limited area. These factors allowed us to explore new terrain with regard to findings of clusters in the U.S. and the possibility of clustering patterns as children around the world are increasingly identified as potential cases.

The spatial clustering in Costa Rica displays similar characteristics as the clustering in the U.S. For example, the primary cluster in Costa Rica is also characterized by relatively high income districts, and more people in those districts have completed secondary school compared to people in other districts. For our regression model, the urbanicity variable was a better fit than education or poverty, but urbanicity was strongly and positively correlated with these variables. Furthermore, when education and poverty were used instead of urbanicity in the covariate-adjusted scan, the results were comparable (data not shown). These findings were consistent with studies in other settings. In other settings, positive relationships between SES or urbanicity and autism incidence were thought to be related to information spread (King and Bearman 2011) or access to diagnostic services (Chen et al. 2007; Malcolm-Smith et al. 2013; Baird et al. 2006; Lauritsen et al. 2014).

The possibility of an information effect was especially plausible because of a public relations campaign in the early 2000s to recruit cases for a genetic study on autism. Nevertheless, the clusters provide limited support for an information effect. The information campaign focused on physicians who happen to work at clinics in close proximity to the HNN, and the large circle in Figure 4 shows that the area of *lowest risk* actually includes many of these clinics, suggesting that physicians in the information-exposed areas are not drivers of spatial patterns. This is further supported by the infrequent mention of autism in the clinical files. Most children do not appear to have been suspected of having autism before arriving at the HNN. Also, while the low severity cluster in south San Jose could have been caused by information spread (again, the clinical files do not support this), the temporal cluster is outside the Central Valley where the information campaign attempted to recruit cases. Finally, the information campaign also focused on teachers and educators, but the percentage of daycares and preschools in each district was not significant in the prediction of cases. Also, the primary special education school in the country, where many individuals with autism attend school, was well outside the low severity cluster.

Our results do not show the expected relationship with urbanicity that would be present in the case of an information effect or an effect related to parental action. Namely, because of universal healthcare access, we did not expect to see a relationship with urbanicity for the moderate and severe cases in the large cluster around San Jose, as these cases should be easy to identify as requiring care at the national hospital, regardless of parental education or income. However, the primary cluster around San Jose was not statistically significant in the covariate-adjusted scan, suggesting that education and income explain the clustering when all cases are included. But when only low severity cases were considered, the primary cluster remained significant, suggesting that education and income do not explain the clustering of cases that are

relatively more difficult to identify, which is when education and income should provide an advantage. This finding, which is also consistent with patterns in California (Mazumdar et al. 2013), suggests the possibility of an alternative mechanism for the very cases that should display an information effect that would be associated with urbanicity, education, and income. Furthermore, contrary to our expectations if there were an information effect, children in the cluster did not arrive at the HNN at an earlier age than children outside the cluster.

Within the San Jose area, which is a scale that could capture parents talking to their neighbors about autism, we also found limited evidence of an information effect. The most likely cluster of all San Jose cases involved several cases in a neighborhood, where the cases may have had contact, but the cases were referred by different physicians and with no mention of autism. While an effect at a very small scale may explain this cluster, the small *n* within the city makes inference difficult.

Our results fail to support the most likely environmental cause of clustering: air pollution (recall that mercury exposure is unlikely). Neither the low severity cluster nor the temporal cluster includes the area with the highest levels of pollution around the HNN (Barrientos 2010), and the risk of diagnosis in close proximity to the HNN is actually lower rather than higher than would be expected by chance. Other potential environmental factors (for a review, see Kalkbrenner, Schmidt, and Penlesky 2014) would likely not include the areas east of the Central Valley. Costa Rica is known for its stringent environmental standards, and the small towns east of the Central Valley are some of the least likely areas in the country in terms of the potential for exposure to environmental toxicants.

Thus, the low severity cases in south San Jose and the temporal cluster outside the Central Valley remain unexplained. For the former, the cases within the cluster involved

different referral pathways from different clinics, so the cluster does not appear to be explained by greater propensity to refer from a small number of clinics. Taken together, these patterns suggest an alternative localized social mechanism that is unrelated to information about autism and is not influenced by income or education.

Importantly, this chapter shows that diagnoses of autism appear in spatial clusters different from the U.S. setting, and even when the diagnostic category is relatively new and information levels about autism are low. The clusters in Costa Rica appear in two areas outside the Central Valley, including an area nearly two hours from San Jose, and they remain statistically significant for low severity cases after controlling for SES. The clusters are not explained by access to healthcare, as children in Costa Rica have universal and relatively equal access to primary care. However, we hypothesize that differences in the types of care may explain some of the clustering patterns. For example, the public healthcare system in Costa Rica often involves long waiting times to see a physician, and some parents may prefer to pay for a private physician simply because they can schedule appointments and are guaranteed to receive more physician face-time. Private physicians may be more likely to refer to the HNN than physicians in the public system, and the decision to consult a private physician may depend more on occupation type than on income and education. This hypothesis is pursued in a subsequent chapter.

The early clustering of autism cases suggests that clustering in the U.S. may have occurred much earlier than it was detected, long before there were insurance supports and incentives that variously promote autism diagnoses, and long before information about autism was available virtually everywhere. This also suggests that other countries in which the diagnosis is new are already experiencing the variable diagnosis of autism.

## CONCLUSION

This chapter follows several studies in the U.S. that identified spatial clusters of autism, but in the present case, the clusters are shown to appear soon after the diagnostic category is adopted. These patterns do not appear to be entirely indicative of parental action, but to further pursue the hypothesis of an information effect, data are needed from parents who bring their children to the clinic for a possible diagnosis.

The methods in this chapter have several potential limitations. First, the number of diagnosed children in Costa Rica is quite small, and one can imagine that social or environmental mechanisms that caused clustering in the U.S. setting have not yet emerged because the affected children have not been detected. One possibility is that a full-fledged information effect requires a threshold number of cases before parents will be influenced in such a way that will implicate spatial patterns. However, we believe our cluster detection method was adequate to detect initial clustering patterns that occur very early in the diffusion of the diagnosis (see Neill 2009; Nkhoma et al. 2004; Van Meter et al. 2008).

Second, in California, Mazumdar et al. (2013) were astute to consider autism clusters at birth and at the time of diagnosis. We did not have address data at the time of birth, but among parents in the metropolitan area for whom we had address data for previous appointments at the HNN, we found no evidence of residential sorting that would explain the clustering.

Third, we lumped the “not diagnosed” category with low severity cases, and we have argued that these two categories are the most difficult to identify. In the U.S., Liu et al. (2010) argued that severe cases are also difficult to diagnose, as they historically overlapped with the mental retardation category. On the contrary, we chose to discriminate based on what we expected would be difficult for parents and referring clinicians to identify, which would include

children with mild language or developmental delay that could be managed with a "wait-and-see" approach. This being said, the not diagnosed and low severity categories can also involve severe developmental problems that simply do not satisfy autism criteria, so these children are not universally difficult to identify as requiring attention at the HNN. We do note that on average, the not diagnosed and low severity cases were 10 months closer to normal development (on the VBAS) than the moderate and severe cases, but the categories do not perfectly capture ease of symptom recognition among parents and referring physicians.

Fourth, the district sizes and populations in Costa Rica are extremely variable. The largest district, with nearly 223,000 ha (861 square miles), or just over two-thirds the size of Rhode Island, had a 2008 population of only 7555. The smallest district has a similar population and is less than 17 ha (42 acres), or just over the size of Yankee Stadium. Some of the predictive power of district variables is lost as a result of this variation, although we do note that variation is less pronounced in and around San Jose, which is where most of our cases were located.

Fifth, we did not directly consider the possible impact of environmental toxicants. In Costa Rica, the concentration of toxicants in west San Jose is suggestive that the clustering is not caused by those toxicants, and there are no known emitters of mercury (i.e., from coal-fired power plants) in Costa Rica. It is plausible that mercury can cause autism (Garrecht and Austin 2011), but we did not consider other sources of mercury exposure. One such source is the widespread consumption of shark meat in Costa Rica, but determining mercury exposure and effects from fish consumption is extremely difficult (Debes et al. 2006), and we have no reason to believe that shark consumption varies across space. The possibility remains that an unknown toxicant is related to the clustering, such as pesticide use in rural areas (see Kalkbrenner, Schmidt, and Penlesky 2014), but given that autism likely represents many conditions with

different etiologies (Rice et al. 2013), efforts to detect an aggregate spatial effect are likely premature, especially with a small *n*.

Finally, there may be unknown cultural factors that relate to parents in Costa Rica identifying symptoms, spreading information, and seeking care for their children. For example, a recent study in China found culturally-mediated variation in how parents respond to language delay (Sun et al. 2013). In Costa Rica, we are unaware of how parental concern for developmental milestones may differ from parental concern in the U.S. If cultural differences exist, they are surely more pronounced in rural areas outside San Jose, where families are not exposed to the cultural influence associated with substantial U.S. tourism and the highly educated and internationally diverse communities. The clusters were all located in relatively urban areas.

We have identified patterns in the distribution of autism different from those in the U.S., and several of the patterns remain unexplained and do not appear to be related to information spread. The patterns are also not likely the result of variation in healthcare access or diagnostic practices, and there are no known environmental toxicants that could cause clustering in the relatively rural areas outside the capital. Now, to precisely identify why even the early diffusion of autism leads to diagnostic variability, we follow Susser's (2004) suggestion of moving to different levels of causation, from the spatial patterns at the population level to processes that occur among families and within households that lead to diagnosis.

# CHAPTER 3

## Parental action and autism in areas of increased diagnostic risk

Chapter 2 showed that diagnostic clusters of cases of autism appear in two areas outside the San Jose metropolitan area: east of the Central Valley and stretching to the town of Turrialba, nearly two hours from San Jose, and south of San Jose beyond the poor urban neighborhoods of Desamparados. For the eastern cluster, despite no systematic effort on the part of the HNN to schedule or recruit cases based on geography (that is, since the genetic study in 2005), there was an unexpected number of cases that all appeared in 2013. The southern cluster involved consistently high numbers of low severity cases between 2010 and 2013. When controlling in various ways for SES, both clusters remained statistically significant but only for low severity cases, and these cases are well outside of the Central Valley area where the information campaign was conducted.

Recall that low severity cases are of particular interest because they are the very ones that should be more difficult to detect, so parents of these cases may be especially susceptible to the exposure of information about autism. In other words, children with severe problems may be universally easy to identify as requiring medical care, whereas low severity autism cases include ones in which the children can variably pass for being typically developing. It is these latter

cases for which parents with no information about autism may be less likely to seek a diagnosis compared to parents who are exposed to information, and patterns of information spread could explain much of the clustering phenomenon from Chapter 2. The aim of this chapter is to focus on the role of parental action in autism diagnosis in Costa Rica, including the possible influence of information spread through social networks of parents and the people around them.

## INTRODUCTION

Social networks and relationships have long been suspected to have a substantial influence on help-seeking behaviors, including how, why, and when individuals decide to seek professional care for their health concerns (Pescosolido 1991, 1992). Medical help-seeking and service utilization studies have consistently found that social networks are correlated with health inequities (e.g., Deri 2005; Sripada et al. 2015), and social networks are even seen as "the mechanism through which illness careers move" (Pescosolido, Gardner, and Lubell 1998:277). However, the assumption of a causal influence of social networks (e.g., Pescosolido 2006) is problematic, as social networks are strongly associated with a range of other personal and structural characteristics (McPherson, Smith-Lovin, and Cook 2001) that can also determine health outcomes. Nevertheless, in some cases, social networks have become a catch-all explanation for aggregate patterns that are assumed to have social roots.

Perhaps the most prominent example of claiming the a priori importance of social networks is in research on autism. First, psychologists and physicians accounted for a steeply rising incidence of autism by pointing to "greater public awareness" (Gernsbacher, Dawson, and Goldsmith 2005) and "growing awareness and knowledge among parents" (Wing and Potter 2002). Then, after identifying spatial clusters of diagnosed cases of young children (Mazumdar et al. 2010), sociologists suggested the presence of what we call a "cul-du-sac effect" on the

diagnosis. The idea is that parents are meeting in and around the neighborhood and talking about their children, and parents of diagnosed children are encouraging parents of symptomatic but undiagnosed children to seek professional medical care. In this view, "knowledge diffusion" among parents and within communities leads to the spatial clustering of cases (King and Bearman 2011). Subsequent studies (e.g., Keyes et al. 2012; Liu and Bearman 2012; Mazumdar et al. 2013) and popular accounts, such as Weintraub's (2011) commentary in the journal *Nature*, have variously argued that exposure to information among parents—potentially when parents congregate at focal points to discuss symptoms (Liu and Bearman 2012)—can increase the risk of diagnosis in their children. Yet there is no direct evidence of this social network mechanism, and researchers have not spoken directly with parents or searched within spatial clusters of cases.

*Social Networks and Help-Seeking*

Despite a long-held interest in how social networks affect health, sociological studies that directly identify specific mechanisms are rare (Smith and Christakis 2008), and especially so for studies that seek to understand the "social influence" pathway of the effects of social networks (Berkman et al. 2000). In terms of diagnostic outcomes, social influence includes processes by which individuals variably recognize symptoms and decide to seek help—what David Mechanic (1961, 1975) called "illness behavior." Given the variation in help-seeking, which exists for a wide range of illnesses and manifests across categories of race, class, and gender, the challenge is to describe social processes that facilitate selection into treatment (McAlpine and Boyer 2007).

Sociological research on selection into treatment was especially popular in the 1960s and 1970s, although much of the work was theoretical. For example, distinguishing between the lay and professional referral systems, Freidson (1960) described how future patients, after experiencing symptoms, most likely first self-diagnose and explore home remedies. If the

symptoms persist, individuals may consult with "friends, neighbors, relatives, and fellow workers" for advice, but only in the "daily intercourse, initiated first by inquiries about health and only afterward about the weather" (376). These casual advice-seeking interactions are what lead to lay referrals to the professional system of physicians and healthcare professionals.

Freidson (1961:104-5) described the "career of seeking help," where patients often begin with non-specialized contacts and move up a "hierarchy of consultants" before they penetrate the professional system. However, with empirical work partially involving "normal" child patients, who only have infrequent contact with the professional system, Freidson (1961) found that parents often *did not* consult with their friends and relatives outside the household before contacting a physician. For non-urgent problems that had no clear answer, parents were known to make use of the lay referral system, but there were class differences in whether lay consultants influenced their behaviors.

Specific findings of the relationship between social networks and help-seeking have varied by setting. In a sample of families in Scotland seeking health and welfare services, McKinlay (1973) found that service use was negatively correlated, rather counterintuitively, with the availability of relatives and friends for consultation. Similarly, at a community mental health center, Horwitz (1977) found that patients with weak kinship but strong friendship ties entered treatment earlier and with less severe problems than patients with strong kinship and weak friendship ties, and social network ties were better predictors of service use than social class. Among mothers participating in a public health food program, Birkel and Reppucci (1983) also found that kinship ties led to delays in and lower rates of service use, presumably because these contacts provided parents with information and advice that lowered their need for professional information. In contrast, in a prospective study involving health diaries, Scambler, Scambler, and

Craig (1981) argued that large kinship networks encouraged women to consult primary care physicians, whereas casual conversations with friends led the women to perceive their symptoms as being less severe, thereby delaying professional consultation. None of these studies captured specific mechanisms of social network influence.

In sociology, the popularity of help-seeking studies waned after those early studies (Pescosolido, McLeod, and Avison 2007). Recently, help-seeking research has primarily come from psychiatrists and epidemiologists, where there have been several studies on parental help-seeking behaviors for young children. A recurring argument is that disparities in help-seeking are caused by differences in how parents interpret symptoms (Zuckerman et al. 2014), which may or may not be related to child psychopathology (Zwaanswijk et al. 2003) but is consistently related to SES (Jackson 1993) and culture (Weis et al. 1988). In the U.S., low SES families (Pavuluri, Luk, and McGee 1996; Weckerly et al. 2005), Latinos (Gerdes et al. 2013), and Blacks (Bussing et al. 1998a,c; Hillemeier et al. 2007) are the least likely to seek help or receive diagnoses for their children, and one reason is thought to be a knowledge gap among parents (Bussing et al. 1998b; Gaziano and O'Leary 1998), presumably about specific diagnoses or symptom etiologies. It is unclear what this knowledge gap entails or what happens when information is diffused through areas with low levels of knowledge.

What is consistent in the help-seeking literature is that social networks, or "social circles" (Kadushin 1966), are thought to influence individuals in terms of how they interpret symptoms, seek information about those symptoms, and pursue professional help. It is also unclear from this limited research what occurs during the help-seeking process for specific diagnostic inequalities. With regard to autism outcomes, do parents of symptomatic but undiagnosed children surreptitiously complain about language delays to their friends and family, perhaps after pleasant

inquiries about the weather? And if so, do the lay consultants respond with a business card for the neighborhood neurologist? Or do other lay consultants broach the subject first, urging parents, or "sanctioning" them (Zola 1973), to take the delays seriously by consulting a professional? Are these lay consultants, as Peter Bearman repeatedly suggests, autism parents themselves?

## DATA AND METHODS

The present chapter focuses on a population of children with autism that has already displayed spatial clusters of cases that may be a consequence of information diffusing through lay social networks. Similar to studies in the U.S., children in higher SES districts in the area of focus, Costa Rica, were at a higher risk for diagnosis (Schelly, Gonzalez, and Solis 2015), potentially an indicator of "local resources and the availability of health-related information" (Mazumdar et al. 2013:88).

Within areas of increased risk of diagnosis, the strategy was to interview parents of diagnosed children about the specific social network connections that influenced their help-seeking behavior, and the interview data were supplemented with a complete medical file review. In doing so, we were able to cross-check names in respondents' social networks with population diagnostic data.

The aim was to describe the help-seeking behaviors among the parents, including the process by which they identify areas of concern in their children, navigate the healthcare system, interact within the contexts of their physical and social environments, and eventually receive a formal diagnosis. If lay social networks influence the diagnosis of autism and lead to diagnostic clusters, then we expect to observe evidence of meaningful social network contacts and information spread. The setting provides a unique opportunity to observe these processes.

*Methods*

This chapter relies on semi-structured interviews with 54 families[24] of children who were recently diagnosed with autism at the primary diagnosing clinic for young children, the HNN. The study was approved by the medical ethics board at the University of Wisconsin and the bioethics committee at the HNN.

We approached parents who attended an appointment in 2013,[25] which included newly diagnosed patients and follow-up appointments from diagnoses in previous years.[26] After their appointments, the parents were approached by the interviewing sociologist and asked if they were interested in participating. These parents, typically only the mother, were briefly told about the study, and all parents expressed interest in participating. After obtaining written informed consent, interviews were conducted in Spanish in a private room at the HNN.

In addition to demographic questions, open-ended questions explored two mechanisms: (1) other people influencing the interviewees to pursue medical advice for their children's behaviors (*incoming* information spread), and (2) interviewees influencing other parents to pursue medical advice (*outgoing* information spread). Following Freidson's (1960) classic "lay referral system," where eventual patients (in this case, parents of patients) are thought to first

[24] A total of 57 families were interviewed, but three interviews were excluded from some analyses because the patients had older siblings with a diagnosis of autism, making consideration of help-seeking behaviors very different.

[25] Using two-sided t-tests, there were no significant differences between the interviewees and the country-wide population of children who were assessed for autism from 2010-2013 for age at first words ($p=0.480$), difference between developmental age and chronological age ($p=0.992$), age at ADOS ($p=0.576$), or ADOS score ($p=0.741$). However, the percentage of low severity cases was high compared to the country-wide sample (40.0% vs. 20.3%), which if anything will accentuate any cul-du-sac effect because information is thought to especially influence parents of children with mild symptoms (Mazumdar et al. 2013). Finally, the geographic distribution of cases appears to be similar to the country-wide sample, where the majority of cases occur in the Central Valley but several occur over 50 miles from the HNN. Importantly, there are several instances of cases in close geographic proximity, including cases living in the same town of less than 5,000 people. This ensures the plausibility of parents having contact with other parents of diagnosed children.

[26] Most patients were diagnosed after 2010, but three patients were diagnosed during the information campaign.

consult a range of acquaintances and family members before ultimately seeking help from a professional, both lay (family, friends, neighbors, and colleagues) and professional (clinicians and teachers) networks were considered.[27]

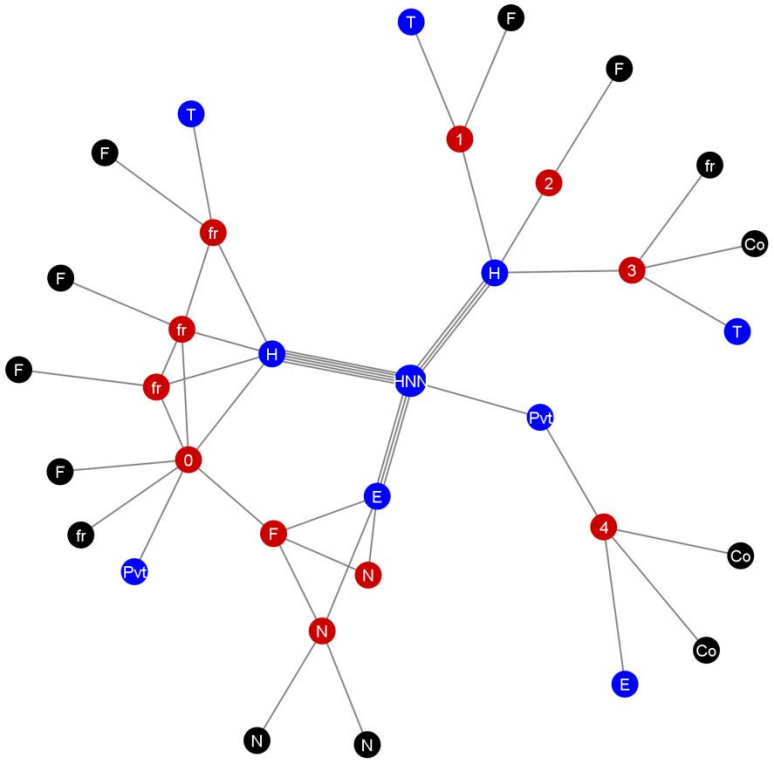
For incoming information that may have influenced the respondents, we asked parents to detail the process of symptom recognition, including what led them to notice something different in their child, who they talked to during the process, and whether anyone suggested that they receive medical advice. They were asked to create a timeline of their visits to clinicians in which they discussed or were asked about the symptoms, and they were asked to expound upon the referral information that was available in the medical files. For outgoing information that respondents may have spread to parents of subsequent cases, interviewees were asked to recount conversations about autism with other people (potentially including other interviewees). They were asked if they had ever seen other children who might have autism and whether they had said anything to the parents. Finally, they were asked to identify children with autism who they knew before or after their child was diagnosed and how often and in what circumstances they spoke to the child or the parents; if applicable, they were asked to provide names for later cross-checking in the medical records.

A plausible consequence of information spread via social network connections with other parents is illustrated in Figure 5, where the software Visone (Brandes and Wagner 2004) was used to represent steps in the lay and professional referral process as a network diagram. We hypothesized that the referral pathways would show interconnected clusters among cases, such as those on the left and bottom of the figure. For illustrative purposes, the parents of patient zero consulted with two family members, two friends, and a clinician before ultimately deciding to

[27] The present findings suggest that a more sophisticated classification of lay and professional networks is unnecessary.

visit a local hospital that referred to the HNN. The connections led a friend and a family member to have their own children diagnosed, and these parents in turn spoke with other friends, family, and neighbors, some of whom were also later diagnosed. On the right side of the figure, the parents of four patients also consulted with their lay networks, but these connections failed to influence subsequent cases, leaving the cases unconnected other than by the local hospital where three of them were referred onwards. Patient 4 shows an instance of parents consulting with colleagues and visiting a primary care clinic, known as Basic Teams for Integrated Healthcare (EBAIS) clinics, before seeing a private clinician who referred directly to the HNN.

**Figure 5** An example of possible network connections[a] in the lay (red and black) and professional (blue) referral process of children (red) who are eventually diagnosed with autism.[b,c]




[a] The nodes are distinguishable as follows: patients, shown in red, are assigned numbers unless they are linked to other patients; lay network nodes, in black or red, include family signified by "F," friends by "fr," neighbors by "N," and colleagues by "Co"; professional networks, in blue, include hospitals marked with "H," EBAIS clinics with "E," teachers with "T," private clinicians with "Pvt" and the HNN in the center with "HNN."
[b] The network is displayed to visualize "stress minimization."
[c] The left side of the figure shows lay network connections that lead to subsequent cases, whereas the right side shows lay network connections that fail to lead to subsequent cases.

Previous research found no evidence of an environmental effect and ambiguous evidence for an information effect. Because there is only one diagnosing clinic and that clinic makes no effort to locate cases, the social network mechanism of parents being exposed to important information, either through lay or professional networks, is extremely compelling. We note that information spread need not be related to autism specifically but would simply have to increase the likelihood that exposed parents would seek help for their children.

In the following section, we highlight three notable cases, chronologically—from birth to symptom recognition, help-seeking to medical consultation, referral to diagnosis—that are representative of patterns in the larger sample. We describe the characteristics of the families and compare the experiences of the parents with those of the other interviewees and district-level demographic data from INEC. Throughout the section, short parental narratives that have been adapted from the interviews are indented in the text.

**AUTISM CASES**

Our first patient, Alberto,[28] was 6 years old at the time of the interview and a resident of a rural area two hours east of where the information campaign took place in the years before he was born.

> pt15.2.10
> When Alberto was born in July 2007, we knew nothing about autism. He was our second child, the youngest by two years, and like all of us, he was born at the Hospital de Turrialba. His father works as an event promoter in the area, and I take care of the house. Our monthly income is about $300.

With just over 60,000 inhabitants, the city of Turrialba is the largest population center in the area, and Alberto's family home in the town of La Suiza is far enough from the main road to see few tourists. La Suiza is an agriculture town, with coffee plantations on the hillsides and sugarcane in the flatlands, and few inhabitants speak English. While the road to the hospital

[28] Inconsequential details have been changed to protect the confidentiality of the families.

passes by two small universities, the university communities are worlds away from all but those who work there.

Based on 2011 census data from INEC, Alberto's family's modest income is enough to place them at more than double the poverty line for rural areas. With elementary school educations, Alberto's parents are also in the majority: only 37.2% of people in their district have attended some junior high. What sets them apart from many rural families is their two-parent household. Finally, the family's home district is considerably less "urban" than those of the other 53 interviewees, at 47.5% compared to an average of 84.5%, and the district's 48 inhabitants per square km is well less than the average of 3,778 in the interviewed sample.

The next patient, Carlos, is from a San Jose district that is 100% urban and has 17,292 inhabitants per square km, the highest density in the country.

> pt2.16.9
> Carlos was born at the Hospital de Mexico, just a short distance from our house in Tibas. I live with my older sister and her two children. I was also born at the Hospital de Mexico, but my family is originally from a small town about four hours south of San Jose. Carlos's father, also from there, moved to the U.S. for work. I attended university for a couple of years but now I work as a seamstress. My sister and I make about $300 every month.

Carlos's neighborhood, where 39% of residents live in extreme poverty, is one of the most dangerous in the city. The housing projects have swelled with makeshift cardboard and corrugated tin shelters that reflect its poverty and drug violence. Like La Suiza, the neighborhood sees few visitors, and the residents are predominantly less educated than average. Few of the residents speak English.

The third patient, Diana, grew up in a middle-income neighborhood in Desamparados, just under 10 km south of the HNN near the southern edge of the city but with over 5,000 inhabitants per km. Her family earns approximately $750 per month, and unlike Tibas, fewer

than 14% of the residents live in extreme poverty. As such, and as is typical of many Central American neighborhoods, families commonly walk the streets and spend parts of the day talking and watching passersby, often congregating around the local corner store. When several of the interviewed parents would report having seen children "on the street" who may have autism, this is what they meant. One may meet or talk about children who live nearby, and even without knowing the parents, the children's names, or exactly where the families live.

If interaction with one's neighbors is a means of passing and receiving information about medical treatments and diagnoses, it is difficult to imagine a meaningful difference among the three patients. Rural and urban, low income and middleclass alike, daylight hours are a time to be outside, even if only while going to appointments or running errands. One would be hard pressed to find a neighborhood where the people do not know each other.

A rural-urban difference appears in relation to healthcare-related travel. Both Carlos and Diana live near several clinics and the national hospitals, and each of their neighborhoods has an EBAIS clinic within walking distance. Alberto's parents also have easy access to an EBAIS clinic, and families in his community have access to a CENSINAI (Centers for Education, Nutrition, and Integrated Infant Health), where working mothers receive free daycare and health-related education.[29] The Hospital de Turrialba, a dilapidated building on a steep hillside, is close

[29] CENSINAI would seem an ideal place for information spread, but only one interviewee reported having met a child with autism at CENSINAI before her son was diagnosed. However, the encounter did not motivate her help-seeking. Her son, a hyperactive and aggressive child, was already under medical surveillance for several comorbidities, including a cleft lip, severe epilepsy, and a gastrointestinal disorder. A diagnosis of autism was the least of this mother's concerns, and unless a child at daycare had similar comorbidities, the two would appear to have very different problems.

to the community, but the area has limited opportunities for specialist care. Most specialists and the national hospitals require a full day of travel.[30]

*First health concerns*

With few exceptions, parents, and typically the mothers, were the first to recognize the first symptoms in their children.[31] This was the case with Diana.

> pt28.1.7
> My husband and I became concerned in the first few months when her movements didn't seem normal, so we took her to see a neonatologist at the big private hospital in San Jose. The doctor diagnosed her with hypotonia, and after that one appointment, the doctor referred us to a public clinic in Desamparados.

One of the main justifications parents provide to account for paying for private appointments is waiting times in the public clinics. People in EBAIS waiting rooms can look like they have forgotten why they are there, and the waiting times can be several hours for a brief appointment. Factors such as a strong bureaucracy, imperfect infrastructure, and deep respect for authority contribute to a culture of waiting. At the HNN, patients commonly fail to show up to their appointments, so for enterprising would-be patients with no appointment, showing up for an extended wait is known to be effective.

The waits in the lower-level public clinics are not always extremely long, but the EBAIS clinicians see substantially more patients (potentially as many as 20-25 per hour) than the specialists in the hospitals. Psychologists at the HNN, for example, have 30 minutes per patient (the department head spends 60 minutes), and in the Developmental Unit, patients receive 30-60 minutes of clinician face-time. The problem is that to be referred to the HNN, patients must move through the system, beginning with EBAIS and then a local clinic. Once a referral is

[30] Chen et al. (2008) reported a substantial disadvantage in service utilization among rural preschoolers with autism in Taiwan, which the authors attributed to a lack of specialists in rural areas. No doubt this disadvantage begins before diagnosis.

[31] When it was not the parents, it was a teacher (n=3), clinician (n=1), grandparent (n=2), or babysitter (n=1); it was never a stranger or neighbor who first noticed symptoms.

secured to see a specialist at a national hospital, the waiting time for an appointment can be six months to one year.

With this in mind, the main reason the private system is preferred is that specialists are accessible on short notice and with a firm appointment time, including evenings. Several patients first pursued a private consultation with the explicit intention of returning to a public specialist, as a diagnosis through the public system helps families receive financial support from the state and referrals to therapies and special education schools.

The main barrier to private treatment is cost. To see a specialist, the cost may range from $50-75 for an appointment. Even for middle-income families such as Diana's, a single appointment represents 7-10% of their monthly income, the equivalent for a family making the median U.S. household income of approximately $375. It is no wonder that families and physicians alike limit the number of private appointments when patients would be equally well-served in the public sector. This was the case for Diana, who was referred after one appointment to a public clinic less than 10 minutes away from her home. It would also be the case for our first patient, Alberto, but not before a brief stint in the public sector: at approximately six weeks of age, he displayed intense and persistent crying in the middle of the night, long after EBAIS clinics close. His parents drove him two hours to the HNN emergency room where he underwent emergency hernia surgery. The next day, they received a referral for follow-up at the regional hospital between Turrialba and San Jose.

Alberto is far from unique in having had medical problems long before characteristic autism symptoms—social delay and quirky behaviors and movements—had time to present, and persistent crying is a common concern. In fact, for 13 of 54 interviewees (24.1%), persistent

crying was one of the first three reported symptoms at the medical history interview upon first arriving at the HNN.

In the case of Alberto's hernia surgery, there would have been no benefit in going to a private clinic, as the reputations of physicians in both sectors are comparable (among the interviewees who visited a private clinician, none reported physician quality as a factor), and the HNN is a similar distance from their home as the private hospitals. For non-emergency care, one would imagine that to circumvent the delays associated with the public system, parents would need to have an idea of what kind of specialist would be appropriate in the first place. Otherwise, they would have to search for information on where to obtain professional help.

Carlos also provoked early concern about his health: his mother had been worried the moment he was born. He was so big, she thought, and he slept too much and hardly cried. She became especially concerned about his health at 6 months of age during a visit with his pediatrician at the small local hospital just 3 km from their rough neighborhood.

> pt2.16.9
> It was so terrible – the doctor said Carlos was prone to sudden death![32] He wouldn't give us a diagnosis, though. After that, I fought with the doctors every time I saw them to try to get a test for his condition. At about 8 months, I took him to EBAIS for normal control, and the doctor said he was late in development, but he told me it was just because he was a boy. He said boys develop later than girls.

Carlos's mother would later complain that the EBAIS doctors were "not as careful" as the doctors at the HNN.

*The referral process*

For the vast majority of parents (45/54, 83.3%), the early concerns about their children were related to developmental delay or difficult behaviors, such as Alberto's crying or, as we

[32] The interpretation of Carlos's "sudden death" risk may have been a misunderstanding. It is likely that the physician was educating the mother on sudden infant death syndrome (SIDS), warning her about sleeping arrangements.

will see, Carlos's aggression toward family members. Only nine of 54 patients (16.7%) complained primarily about unusual behaviors or social delays that are commonly associated with autism.

Diana's hypotonia also did not suggest autism, so her parents had begun searching for answers when even the most seasoned autism expert would have no reason to suspect autism. Beginning at approximately 6 months, Diana's parents had been taking her to a second-level clinic for follow-up, and soon thereafter, she started having seizures. Later, her development became noticeably delayed in relation to other children.

> pt28.1.7
> We noticed problems at around age 2. She was acting very isolated in public, and she basically only said mama and papa. She was also having difficulty learning to walk. We asked the doctor at the local clinic to refer us to the HNN for further treatment, which he did. He never mentioned autism.

Like other parents in the sample, Diana's parents did not talk with other parents about autism. In fact, nearly half of the families, 26 of 54 (48.1%), knew nothing about the disorder before their children were diagnosed. Of the remaining parents, 25 (46.3%) knew very little about autism or had only heard of the category (often only from watching the 1988 movie *Rain Man*), and only three parents (5.6%) knew about specific symptoms. One parent in the sample, whose neighborhood had several children with autism, was asked whether the parents had influenced each other to seek a diagnosis: no, she said, it was just a coincidence, and the parents did not know about each other until after their children were diagnosed.

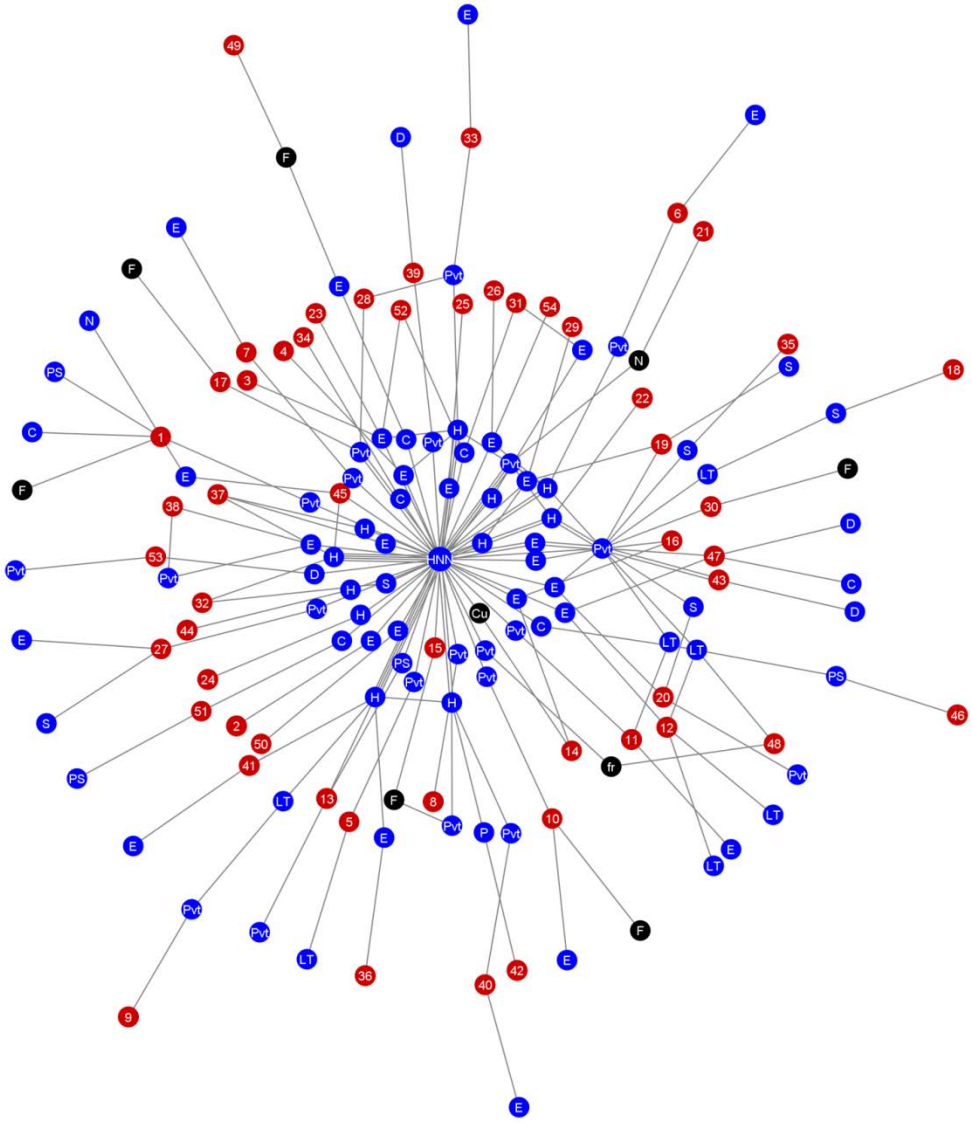
The three parents who had considerable knowledge of autism are university educated (note that 29.5% of adults in San Jose have some higher education). One of the parents had worked in a U.S. hospital, and another is a trained physiotherapist who has treated patients with autism. The remaining parent heard of autism via information spread: a radio program had

described the symptoms of a child with Asperger's, and the mother thought the behaviors were similar to the those of her 18-month-old son, who did not want to play with other kids or understand when he was spoken to. When she consulted her friends and family, people suggested she wait before acting on her concerns, as they thought his behaviors would return to normal over time. The problems persisted for another six months before she mentioned her concerns to her family doctor, who referred the case directly to the HNN for evaluation.

Surprisingly, the actual network connections among our sample of 54 patients reveal no direct network connections among families, meaning that none of the parents knew any of the other children (see Figure 6). Importantly, the figure shows that lay network connections, or parental reports of lay consultations, were much less common than expected,[33] reportedly occurring only nine times. In four of those nine cases, the connection did not lead directly to the next referral step, indicating that the connection influenced the parents to seek professional help about their concerns but without providing specific advice on how to do so. Only in one case did a neighbor suggest a clinician, but the neighbor did not have a child with autism or mention the diagnosis. The figure also shows that several clinicians and clinics are the center of small clusters of patients; the parents' reasons for choosing those healthcare options, though, were not because they received information from or were influenced by parents of previously diagnosed children.

[33] Lay connections are meant to capture consequential connections without which the parents may not have acted, so the connections here do not include generic reports of consulting with "friends and family." While it may seem counterintuitive that reports of consequential lay consultation were so rare, this result is consistent with McKinlay's (1973) finding that "underutilizers" of health and welfare services were much more likely than "utilizers" to consult with relatives and friends prior to using the services. Utilizers tended to turn to "semi-professionals" rather than people in their lay networks. Similarly, in a recent study, Boulter and Rickwood (2013) found that only 40% of parents of children with mental health problems had sought informal help (but including the internet and books) before the diagnosis.

**Figure 6** Actual network connections[a] in the referral process of a sample of children who were eventually diagnosed with autism at the HNN, Costa Rica.[b]




[a] Patients are shown in red and numbered in the order they were diagnosed. Lay network nodes, in black, include family signified by "F," a friend by "fr," a neighbor by "N," and a customer by "cu" (note that no patients are connected); professional networks, in blue, include hospitals marked with "H," EBAIS clinics with "E," teachers with "T," language therapists with "LT," preschools with "PS," daycares with "D," private clinicians with "Pvt," and the HNN with "HNN."
[b] The network is displayed to visualize "stress minimization."

The most popular referring node, a private physician whose node in Figure 6 is to the right of the HNN, was directly connected to five patients and removed by two network nodes from another five. Notice that the indirectly connected patients first went to language therapists (LT nodes) or schools (S nodes). Two of those patients, 12 and 48, originated from the same

therapist. In the case of patient 18 in the figure, the referral came from a teacher. Having noticed anomalous behaviors in his son, the father was not seriously concerned, as he recalled having had similar behaviors when he was a child. Other patients were also first identified by teachers as being slow to socialize, and a handful of children's early problems went unnoticed by parents and EBAIS clinicians, presumably because the children were not observed playing in group settings. With this in mind, a private physician was often recommended by a teacher. Such was the case for patients 35 and 20, who were also both seen by the popular private physician.

The parents who visited the popular physician did not know each other or share lay network connections. On the contrary, Figure 6 shows that a range of teachers and therapists (all in the San Jose area) referred to a limited number of specialists. For the few parents who sought additional professional care without apparently knowing where to turn, their primary help-seeking behavior was to consult professionals from within their lay networks. For example, patient 12 had been seeing several therapists, starting at age 2, for emotional problems and language delay; when one of them suggested the need for a diagnosis, the mother consulted another therapist, purportedly also a friend, who referred to the neurologist. Likewise, upon developing serious concern for language delay at 2.5 years, a mother in a rural area several hours south of San Jose traveled an hour to see a language therapist, her cousin, who worked in a neighboring school system. In that case, the therapist did not refer to a private physician in San Jose, possibly because she was not connected to the Central Valley professional networks, so the mother received a referral through the public pathway.

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Long after the hernia surgery, Alberto's mother would also consult with her family. In his first 12 months, though, she had no additional concerns about his health. She took him to his

routine appointments at the EBAIS down the street, mostly for vaccines and public health information. He was fussier than his older brother, but not problematically so. He said his first words at close to one year, but then at 18 months, he stopped talking.[34] His behaviors also deteriorated: he frequently became angry, cried to extremes, and hit himself. When he was around other children, he hit them as well. He preferred to play alone, and there was something about his style of playing that seemed odd.

> pt15.2.10
> I never asked the EBAIS clinicians about his behaviors. I did talk with my family, but they all said his behaviors would pass. One day, I was visiting my mother's house in Turrialba and her boss, a dentist, stopped by. We were talking about Alberto and watching him, and the dentist told me about autism. He said that autism could appear in varying degrees and that it impacted how people relate to each other. He suggested going to a psychologist in Turrialba, so I made an appointment the next day.

The psychologist agreed that it could be autism but said he could not make the diagnosis. He suggested the parents visit a pediatrician at the Hospital de Turrialba.

We mentioned that for specialty care, parents may be hard-pressed to know where to turn for medical help. Alberto's mother, for example—despite her ongoing concern—may have further delayed pursuing medical help had it not been for the dentist. For others, private specialty care may have been a more likely consideration if somebody suggested a clinician or if parents knew what kind of care to pursue, suggesting a type of career contingency (Goffman 1961) that would explain some variation in diagnosis.

So with no idea of where to turn, Alberto's parents pursued private care only after an uninitiated encounter with an individual with professional authority. After first seeing the psychologist, Alberto saw a public pediatrician at the Hospital de Turrialba and was then referred

[34] We note that 10 of 54 parents (18.5%) reported language regression as one of the first symptoms of autism, which is consistent with reported rates of regression in the U.S. (e.g., Lord, Shulman, and DiLavore 2004; Volkmar and Cohen 1989).

(for mental retardation) to a psychiatrist at the regional hospital in Cartago. From there, he saw a public neurologist before being referred to the HNN. Alberto was seen at the Developmental Unit at the HNN at 2.5 years of age.

The diagnosis at this point would come more quickly than the typical year, as he only needed three appointments over a period of two months. At just under 3 years old, Alberto was assessed to be developmentally 10.5 months old and was diagnosed with autism. But like many of the patients, the key parental action in Alberto's pathway to diagnosis was visiting that first clinician shortly after he was 18 months of age.

*Potential for exposure to information*

Unbeknownst to his parents, the diagnosis made Alberto part of a cohort of Turrialba children who would be diagnosed with autism in the coming years, establishing one of the country's primary spatial clusters of autism diagnosis. His parents were, however, intimately familiar with the clinical observations from the day of his diagnosis: Alberto "does not control sphincters; eats everything he can; is anxious; emits sounds; does not say words, respond to the examiner, have joint attention, respond to social smiles, or anticipate games with objects." The good news was that he was not hitting himself anymore. He was referred to language therapy at the HNN and psychiatry at the Hospital de Turrialba for follow-up.

Also unbeknownst to Alberto's parents, two interviewees, who were both diagnosed after Alberto, lived down the road in Turrialba. The older of the two, the son of an accountant, was first taken to EBAIS for developmental delay. He had been having trouble crawling and walking by 1 year of age. The parents were uncomfortable with the advice from EBAIS to give him time, but instead of consulting with friends and family, they consulted a private physician in San Jose.

The physician shared their concern and guided them through the public system, beginning with a pediatrician at the Hospital de Turrialba.

The younger of the two proximate interviewees lived in a different neighborhood in Turrialba. At 2 years old, he was still not talking and laughed for no apparent reason. A public pediatrician said to give him time, but after four visits and over a year, the problem was not improving. The pediatrician referred the child to the Hospital de Turrialba where a psychologist mentioned autism and referred directly to the HNN. At this point, the child was well over 4 years old and was talking in the third person and presenting with obsessive behaviors and interests.

Despite living in close proximity to each other and living in one of the spatial clusters of autism diagnosis in Costa Rica, none of these parents knew one another.

Alberto's case is not unique with regard to strong potential for information spread to other parents of children with autism but no signs of contact. In fact, being convinced by anyone that their child may have autism was unique: only one other parent admitted to having been influenced before the diagnosis. The parent was cutting the hair of a client and talking about her child's symptoms. The client, a physician, happened to be a good friend of the diagnosing physician at the HNN and helped the parent obtain a direct referral.

Only one respondent recalled a stranger mentioning autism. At 8 months old, her child was banging his head on the window of a public bus. A woman asked if he had autism, but the respondent thought little of this interaction until she recalled it after receiving the diagnosis. In fact, at the time of the head-banging incident, her child had already begun early stimulation treatment, and even though the mother saw children with autism at the early stimulation clinic, she did not suspect her son of having it. He behaved too differently from the other kids.

Carlos's mother also had the potential to be influenced by other parents, as several other interviewees lived in an adjacent neighborhood. But none of the parents knew each other, either before or after the diagnosis. The diagnostic process for Carlos never left the public realm, and consequential communication with other parents never occurred. Approximately two years after hearing of his potential for sudden death, Carlos's mother received a written referral from an EBAIS pediatrician: Carlos showed "slow motor development, language delays, inability to form phrases, and aggressive behavior."

*Outgoing information spread*

After the diagnosis, Carlos's mother became a kind of lay expert on autism and even reported having "telepaticos" for detecting other children with autism.

> pt2.16.9
> I took every opportunity to learn more about autism. I attended talks at the HNN and found literature about the disorder. The HNN gave me a pamphlet on autism, and I began talking with other people about the diagnosis to warn them of Carlos's behaviors, beginning with his teachers at school. Sometimes people on the street ask about his behaviors, and I tell them about autism. One time, I saw a boy who I think had autism. He was writing numbers on the wall of my house. I told the mother that he might have autism, but she was stubborn and didn't believe me.

Carlos's mother also had a nephew who was diagnosed with autism. However, she believed that her nephew, who was two years older than Carlos, had problems in the mental domain, whereas Carlos's problems were "more of a social issue." She never imagined that the boys had the same problem. She also never mentioned autism to her sister.

On the south side of San Jose, Diana was also living in a spatial cluster of low severity cases, where there was a six-fold increase in diagnostic risk within the cluster. Her parents were unaware of this fact. Before Diana's diagnosis, they knew of only one person with autism, a

niece[35] who lived over the mountains in a rural area, over an hour away by car and double that by bus. Although the parents were siblings, they did not have a "strong relationship" and only talked about once a year. Diana's parents did report that they had discretely compared Diana's development to that of their 2-year-old niece. Unlike her cousin, who displayed aggressive behaviors, Diana was isolated in public and delayed in language and walking; Diana's most conspicuous problem was the onset of seizures as many as four times each day.

In Turrialba, Alberto's parents also had opportunities to spread information about the diagnosis to other parents. While his mother did not know anyone with autism before the diagnosis, she met as many as 10 mothers of children with autism when he started special education. She began a routine of taking Alberto to the school three times a week. She talked regularly with the other mothers about the behaviors of the children. Like the other interviewees, she also talked about the diagnosis with Alberto's teachers and her family. In fact, her husband's nephew displayed suspicious behaviors, but the boy's parents "did not want to acknowledge the problem." Alberto's mother would not dare mention autism to parents for fear of how they would react.

In all, 31 of 54 parents (57.4%) reportedly suggested the need for a medical evaluation to a parent of a symptomatic child. More than half of those 31 did not recall having mentioned autism and simply suggested going to a specialist or the HNN. Family was the most common relationship for these connections (n=7). The next most common relationships were children at school (n=5) and acquaintances (n=5); neighbors (n=3) and children in clinic waiting rooms (n=3); friends (n=2) and children of colleagues (n=2); and children at church (n=1) and on the street (n=1). The remaining parents (n=2) did not recall how they knew the other children.

[35] Three of 54 patients (5.6%) had a cousin with autism. Only one parent shared information with her sibling. The siblings were "really close" and talked about shared characteristics, but the mother "didn't accept that he had characteristics [of autism] because she just thought he was spoiled."

Remarkably, using the names of children and parents to whom the respondents suggested medical care, we cross-checked in the HNN database (from 2007-2013) to locate evidence of subsequent diagnoses: no matches were confirmed, suggesting that none of these patients later received a diagnosis.

Many respondents expressed reluctance to pass information to other parents for fear that "parents won't accept" and "don't like to be criticized," and several respondents, upon suggesting medical treatment, reported that the other parents "didn't believe" something was wrong. One respondent noted categorically that information-receiving parents "aren't interested" in receiving advice about their children. Of the respondents who mentioned healthcare options but not autism specifically, several justified the choice by noting that they "didn't know how the parents would react." As a cautious alternative to mentioning autism, one respondent pointed out to her sister-in-law that her child acted a lot like her son, but she "wouldn't dare" say anything more direct. Another respondent told her sister to take her 17-year-old daughter to a specialist because she was too dependent.

Given this reluctance to recommend that other children receive a medical evaluation, the circumstances in which parents were comfortable and willing to pass information were rather limited. In several cases, other parents approached the respondents to ask about their child, but there tended to be outside reasons for seeing the two children together, such as while standing in line or waiting in a medical waiting room. The children were often seen to be "doing the same things."

As with the cautious sister-in-law, parents reported conspicuously similar symptoms between their children and the other child when they initiated conversations: there was a nephew whose main symptom, like the respondent's child, was extreme language delay; a daughter of a

colleague who had the “same features” as the respondent’s daughter; and a female acquaintance whose female child was “equal” to the respondent’s daughter. A neighbor encounter involved a close friend whose child, curiously like the respondent’s child, also displayed stereotyped behaviors, hit his mother, and had epilepsy. In another, the neighbor child was seen running out in the street, apparently oblivious to the danger of cars. The respondent told the mother that her child had done the same thing and that they “had something similar.”

## DISCUSSION

Despite the high potential for social network connections among parents—namely because spatial clusters of cases were identified in high SES urban areas, and the genetic study spread information about autism in and around the capital—the interviews revealed no parent-to-parent ties among parents of diagnosed children, and parents knew very little about autism at the time of their children’s diagnoses, suggesting an absence of information spread about autism. These findings are inconsistent with the cul-du-sac model of information diffusion among parents (e.g., Liu, King, and Bearman 2010), although parents *were* engaged in the spreading of information to other parents. In the following sections, we discuss how incoming and outgoing information may have had such a minimal effect on the help-seeking behaviors of parents.

### *Incoming information*

Given the search for mechanisms that would lead to spatial clusters of cases, the limited number of reported lay network connections had little impact on the referral process for the interviewed parents. The four notable connections in which such influence was reported were the dentist in Alberto’s case, the physician who was having his hair cut, the therapist friend of the mother of patient 12, and the cousin (also a therapist) of the mother in a rural area south of San Jose. These examples all involved individuals with high levels of expertise and authority, which

is consistent with mental health research by Rogler and Cortes (1993) and early work by Freidson (1961), McKinlay (1973), and Horwitz (1978). These lay connections with professionals may well have a substantial influence on parents' help-seeking behavior, but there also may be something to Freidson's (1960) quip about only discussing symptoms in the casual context of conversations about the weather. Rather than seeking out the dentist or the physician, two of these parents found themselves discussing their children unexpectedly, and in the context of other everyday activities. The other two parents sought advice from trusted friends and family (see Levin and Cross 2004) with professional expertise.

Most often, advice that parents were exposed to was unhelpful, such as when parents were routinely approached about their children in public situations. When their children were seen to be behaving inappropriately, strangers were rather uninhibited about offering advice. A classic example is a respondent whose child routinely waved his arms and was "very corky," always running back and forth. On several occasions, passersby approached the mother and inquired as to why she did not see a doctor. The mother was annoyed by these public encounters but was later influenced by a language therapist to pursue medical help, suggesting that unlike individuals with some medical authority, strangers have limited ability to influence parents' help-seeking behaviors.

In ethnographic work about autism parents in New York, de Wolfe (2013) documents similar encounters. The U.S. parents were routinely inundated with unwanted attention, and they often mentioned autism as a way to account for inappropriate public behaviors that generated comments from strangers (see also Farrugia 2009). One father, whose son had complete "meltdowns" in the park, said that other people commonly proposed solutions or criticized his parenting. Citing Goffman's (1963) work on stigma, de Wolfe points out the difficulty for

parents when they are seen by others to be engaging in bad parenting, such as when unconventional forms of discipline are the autism parent's only chance for reprieve. One of de Wolfe's parents, Richard, became so frustrated with these encounters with strangers, which often occurred in situations when he was already flabbergasted, that he printed business cards to settle misunderstandings about his son's tantrums. Another parent thought mention of the diagnosis to account for "bad" behavior was futile, believing that people would never understand unless they experienced the situation. In Goffman's (1963:129) terms, these parents were in the midst of the "normative predicament," adapting to the gap between normal childhood public behavior and that of their stigmatized children.

Other parents reported encounters that were less memorable. Recall the anecdote of the 8-month-old child who was banging his head on the window of a public bus. The respondent remembered that a stranger had asked if he had autism but recalled the encounter only after the diagnosis. Indeed, at the time, her child had already started early stimulation treatment that would be the source of his referral. The child had been born premature, and at 6 months of age, had apparently stopped developing. At 8 months, the wheels of referral were already turning down the road to diagnosis.

The sense from the accounts is that parents, including parents on the slow side of the spectrum of identifying or showing concern for anomalous behaviors, are aware of problems very early in their children's lives (for similar reports in the U.S., see Midence and O'Neill 1999), long before opportunities arise for parent-to-parent information spread. With this in mind, we note that early symptoms are poor predictors of later symptomology (Tolbert et al. 2001), and many stereotypical symptoms of autism, such as hand waving, are not present in younger children (Guinchat et al. 2012; Stone et al. 1999). If it does present at all, classical autism often

does not appear until 5 years of age (Lord and Risi 1998), and parents of younger children may not report such behaviors (Howlin and Asgharian 1999).

In our sample, stereotypical autism behaviors did emerge in several cases but typically long after generic symptoms were identified. This is consistent with research from before (e.g., Rogers and DiLalla 1990; Volkmar, Stier, and Cohen 1985) and after (e.g., Baghdadli et al. 2003; Tolbert et al. 2001) the U.S. autism "epidemic," from other low- and medium-income countries (e.g., Chakrabarti 2009; Samms-Vaughan 2014; Sun et al. 2013) and from the U.K. (De Giacomo and Fombonne 1998): parents seem universally capable of identifying anomalies before 2 years of age, but the early symptoms of concern are primarily related to language delay. When problems are social in nature—a step closer to classical autism—many people still do not associate the anomalies with autism (Mitchell and Locke 2014). As in our sample, many children provoke concern because of general medical concerns or day-to-day challenges, such as sleeping problems (Chawarska et al. 2007) or excessive crying. These types of generic developmental problems do not require information from other parents to elicit concern.

*Outgoing information*

Given the small number of parents in our sample who reported being influenced by incoming information (3.7%), it was surprising that reports of outgoing information spread were relatively common (57.4% of families). The outgoing information appears to have made no difference to the targets, as no subsequent diagnoses could be confirmed in the medical records.

A likely explanation for an absence of matches is that the incoming information from other parents occurred but went unnoticed, which implies that outgoing information spread also had little effect.[36] This would explain respondents' complaints that other parents do not want to

[36] It is possible that the information-exposed parents had their children assessed but did not receive a diagnosis. However, if this occurred with any regularity, the children of the information-exposed parents would have had to all

hear “criticism” about their children. The implication here is that our respondents knew that recommending that a parent take her child to a clinic for a diagnosis would be seen as criticism. In the least, the interaction would involve a delicate conversation, perhaps especially if the other parent was a friend or close relative.

*Rain Man effect*

While the early symptoms in our sample were consistently generic, the symptomology had tended to widen by the time a diagnosis was received. Yet these parents did not have the advantage of seeing hundreds of children with autism, so even then, as experts on their own autistic children, they had a limited perspective on symptomology. The implications of this are seen in several of the parents who knew about autism because they had watched *Rain Man*, and not surprisingly, they never thought their 2-year-olds resembled the 50-year-old Dustin Hoffman.

While some parents developed a more nuanced view of autism as a spectrum of behaviors that may be present to varying degrees, the majority viewed the diagnosis as a concrete description of the restricted set of behaviors in their children. The consequence is that when respondents recalled encountering diagnosed children before their children were diagnosed, typically at school or through a relative, the symptoms were so drastically different that they did not consider the possibility of a shared diagnosis, and this occurred even when the parents were relatively knowledgeable about autism. The same phenomenon occurred between two interviewees, both with a previously diagnosed child: the younger children had symptoms that were very different from the diagnosed siblings, so despite being lay experts on autism, the diagnoses of the younger siblings were a complete surprise.

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be low severity cases with borderline symptoms, which seems somewhat implausible given that the low severity cases in the diagnosed sample reported such an absence of influence.

What we call a *Rain Man effect* describes the strong and lasting influence of exposure to memorable depictions of a diagnosis, including the assumption that one's diagnosed child is representative of the typical case. The implication is that even with several neighbors with autism or a child with diagnosed peers at school, the symptom variability may be so extreme that the potential for consequential information spread is low. This is consistent with what we know about the genetic causes of autism, where autism may represent many different conditions that have various causes (Rice et al. 2013), and the diagnosis is associated with a nearly unlimited array of symptoms (Hughes 2009) even though they might be fitted with the classic "triad" of impairments.

In the best case scenario for parent-to-parent information spread, parents of diagnosed children with autism may learn that autism can present in numerous forms and that symptoms can vary. But as they observe other children and find themselves in conversations about child behavior, as they are consulted by friends and family about behavioral delays in other children, the likelihood of suggesting that another child has autism may depend on whether those symptoms fit the classical "stigmata" of the diagnosis (e.g., Shorter 1987:71), or at least their early representations of the diagnosis. If another child presents the same behaviors as one's own child, such as the child who ran in front of cars, a parental lay referral is easy to imagine. But how often do such peculiar "symptoms" overlap?

*Lay and professional referral networks*

Consistent with several previous reports (e.g., Boulter and Rickwood 2013; Scambler et al. 1981), connections with lay social networks potentially delayed rather than hastened the referral process. Similarly, a common complaint among parents who pursued the public referral pathway—especially in the first level of care at the EBAIS clinics—was that physicians showed

little concern for early symptoms, often suggesting a wait-and-see approach. Physicians choosing a wait-and-see approach early on for children later diagnosed with autism has also been reported in the U.K. (Smith, Chung, and Vostanis 1994), the U.S. (Hutton and Caron 2005), and Sweden (Sivberg 2003). The exception in the public realm was for patients who received additional medical surveillance for co-morbidities, such as Diana's hypotonia and seizures. Because of receiving specialist care for their conditions, one possibility is that these patients received extra face-time from physicians, perhaps increasing the likelihood of eliciting physician concern to a degree comparable with private care.

Among parents who pursued the private pathway, there were no complaints of inattention to their concerns. This is noteworthy for two reasons. First, private clinicians in Costa Rica are concentrated in urban areas, which are exactly where autism diagnoses tend to be concentrated in other settings (e.g., Chen et al. 2008; Lauritsen et al. 2014). Given the evidence that proximity to neurologists and psychiatrists increases the risk of autism diagnosis (Kalkbrenner et al. 2011), it may not be a coincidence that underdiagnosed populations also have limited access to services (Liptak et al. 2008). Second, in the present sample, parents who pursued private care had more education and higher incomes than parents who only followed the public pathway, and referrals appeared to be more likely from the private pathway than the public pathway, which is consistent with research showing an increased risk of diagnosis among high SES families (Durkin et al. 2010). Thus, the positive relationship between urbanicity/SES and diagnostic risk (Schelly et al. 2015) may provide a piece of the puzzle of variability in diagnosis. Specifically, in Costa Rica, the private healthcare pathway may be driving the spatial patterns, suggesting the importance of supply side (clinics) rather than demand side (parents seeking diagnosis) mechanisms, which is consistent with some research in the U.S. (Mazumdar et al. 2013).

Information moving through professional networks of referring clinicians may be a factor in the diagnosis of autism, but in the present location at the present time, it seems unlikely that information moving through lay networks is a meaningful component of the social diffusion of autism diagnosis.

## CONCLUSION

Following the work of Pescosolido (1991, 1992) on social networks and health service utilization, the classic Liu et al. (2010) paper was the first among many studies to assume the presence of a cul-du-sac effect, where parents spread information about autism and influence each other's help-seeking behavior. In the present setting, information spread was highly plausible: an information campaign in the mid-2000s targeted individuals to obtain referrals for a genetic study, and autism continues to be underdiagnosed; during the information campaign, there was only one diagnosing clinic, an absence of insurance incentives, and universal healthcare for children, thus removing clinical variation in diagnostic procedures as a possible explanation for disparities. Finally, spatial clusters of the diagnosis displayed similar characteristics as those previously identified in the U.S., where information was blamed as the cause (Schelly et al. 2015). Even after focusing in on the areas of increased risk—the very locals where an effect would be most conspicuous—we found no evidence that information moving through lay social networks is driving the diagnostic disparities.

The present chapter interviewed the parents of a treated sample of children, which means that we could not consider how information influences, or would have influenced, parents of symptomatic children who remain undiagnosed. However, if social networks were a factor in how the early diagnostic clusters appeared, we would have seen evidence of their effects in the

present sample. Thus, we are confident that information is not presently driving the spread of autism diagnosis in Costa Rica.

We also did not directly consider professional networks. The interviews and medical file information suggest that professionals are driving at least some of the variation in diagnosis of autism, but it remains unclear how these physicians are connected or what role information about autism plays in their referrals. In the medical referrals, autism was rarely mentioned explicitly, but are physicians who are aware of autism more likely to refer than physicians who know little about the diagnosis? Are there social networks of clinicians that are especially good at recognizing developmental delay in young children? What are their relationships?

Whatever the answers, a potentially more important question is whether these findings apply to the U.S., where the spread of autism diagnosis (and information) has reached mature stages that Costa Rica may not reach for years. Indeed, even the U.S. clusters from the 1990s may represent a more information-receptive environment than that of Costa Rica. The parents in Costa Rica were unreceptive to hearing advice from individuals without some medical authority, which may be dissimilar to the U.S. setting where respect for medical authority may be relatively low. What is likely not different, though, is that parents were on the path to diagnosis, albeit unknowingly, long before there were opportunities for consequential advice-giving; furthermore, the plurality of autism symptoms made it unlikely for two children to be attributed to the same diagnosis. These factors are not unique to Costa Rica.

Nevertheless, we cannot rule out the possibility that a cul-du-sac effect has yet to occur in Costa Rica. There may be a threshold effect that involves a minimum density of diagnoses before parent-to-parent contact is meaningful, and the parents of the undiagnosed cases may be especially in need of information in ways that differ from the treated sample. We pointed out a

tendency among parents to be reluctant to discuss the condition with the parents of some symptomatic children for fear that the parents will be unreceptive or even offended. This may be a realistic fear if the information-receiving parents have never heard of autism, as the diagnosis and related symptoms may not even be in their repertoire (Shorter 1986, 1987) of possible problems in a child. But as autism becomes better known and the number of cases increases so that, as in the U.S., everybody knows somebody with autism (Grinker 2007), the topic may become easier to broach, even outside the most forgiving settings such as clinic waiting rooms. In our study, it seems that autism-naive parents, clinicians, and teachers who are proactive in seeking answers for language- or developmentally-delayed children may nevertheless be the source of spatial clusters; but what patterns will emerge if those individuals are all aware of autism symptomology?

Most of all, this chapter suggests a need for caution in the often-invoked attribution of disparate health outcomes to social network effects. Studies have consistently identified disparities across categories of income and education, with low income, low education families receiving substandard care and experiencing unsatisfactory outcomes. But theories that account for these disparities need to dig deeper than network ties to consider how parents categorize their children in terms of normality or typicality, how they explicitly and implicitly compare their children with others, and how they negotiate encounters with clinicians and other experts whose advice is sought. In the present chapter, neither information nor lay social networks were the drivers of aggregate patterns of diagnosis, but further research is clearly needed. We encourage sociologists to renew their interest in the “social influence” pathway of the effect of social networks on health, especially using empirical evidence in specific settings and for specific outcomes of interest.

# CHAPTER 4

## Patterns of referral in the diagnosis of autism

The previous chapter focused on how lay and professional social network connections influenced Costa Rican parents' decisions to seek care. Relying on a complete medical file review, a network diagram of the referral pathways of 54 cases was created. The network diagram showed that there were no direct network connections among parents, and lay social networks did not appear to substantively affect the help-seeking process. On the contrary, there was evidence that parents were on the path to a diagnosis before advice-giving—perhaps especially from other parents—would have been able to influence their help-seeking behaviors. Furthermore, there was some initial evidence of variation in how much professional face-time patients receive at the private clinic, where children receive more medical scrutiny and, accordingly, are potentially more likely to be referred. This suggests that the pathways to diagnosis reported in Chapter 2 may be routed not through parental networks but via the referring clinic and clinicians. With this in mind, a referral mechanism that leads to variation in clinical identification may be to blame for the diagnostic variation by locale that was observed in Chapter 2. In particular, we consider whether this referral mechanism simply relates to physician face-time with patients.

Like Chapter 3, this chapter continues to focus on the 54 families of children who were diagnosed with autism and later interviewed about their experiences obtaining care for their children. The interviews included questions about symptom recognition and the referral process, which provides an opportunity to understand what actually happens before parents arrive at the HNN for a possible diagnosis. The chapter begins by detailing the healthcare system and the referral process in Costa Rica, which is in many ways quite unique compared to the U.S. because children in Costa Rica have universal healthcare access; however, there are discrepancies between families that exclusively follow the public pathway and families that pursue private care. These discrepancies may affect how much scrutiny patients receive from their physicians, a factor that, in turn, may lead to diagnostic variation. Special attention is paid to the public and private pathways, and the pathways are mapped out for all 54 patients, including their pathways to lay network connections that were mentioned in Chapter 3.

Again using data from these 54 patients, the chapter then develops regression models predicting (1) the early identification of symptoms among parents and (2) the referral duration, or the time between symptom recognition and the first visit to the HNN. The aim of this section is to begin to differentiate between factors involving parental action and those involving a referral mechanism related to clinical detection. For example, if variation in clinical detection drives diagnostic variation, then parental knowledge or features of the household should not predict the speed with which parents obtain an appointment in the developmental unit at the HNN for a possible diagnosis.

The chapter next considers differences between mild and moderate/severe cases of autism. If this referral mechanism explains diagnostic variation, it likely has a stronger effect on mild cases that are more difficult to identify, as these cases may necessitate additional medical

scrutiny. Cases were classified as mild if the children either attended or were later able to attend a regular school (n=22). We use t-tests and z-tests to compare means and proportions between mild and moderate/severe cases, again considering differences that would be suggestive of the referral mechanism.

Next, the referral pathways are considered for mild and moderate/severe cases, and household incomes are displayed to highlight differences between the public and private healthcare pathways. Finally, the chapter considers hypothetical scenarios predicting how many mild cases will arrive at the HNN given expectations about private healthcare utilization and detection probabilities between the public and private sectors, and these scenarios are compared with the findings from the 54 cases.

Before we discuss the Costa Rican healthcare system, we begin with a review of literature on referral mechanisms related to diagnostic variation.

## REFERRAL MECHANISMS: A REVIEW

In a range of settings, studies have consistently shown that the risk of autism diagnosis is higher in urban areas (e.g., Daley 2004; Lauritsen et al. 2014), especially for pre-school-aged children (Chen et al. 2007). In the U.S., Mandell, Novak, and Zubritzky (2005) found that children living in rural areas were diagnosed nearly five months later than urban children, although “near-poor” children were diagnosed nearly 11 months later. These findings are consistent with those of Chen et al. (2008) in Taiwan, where both suburban and rural children were diagnosed at an older age and experienced a longer delay between symptom recognition and diagnosis. The authors suggest that these patterns can be attributed to shortages of medical specialists in suburban and rural areas, which may be a pattern that is duplicated throughout much of the world. Consistent with this idea, service utilization studies have shown that racial

and ethnic minorities and families living in rural areas are the least likely to utilize services for autism (Thomas et al. 2007). There seems to be consensus within these studies that "identification-related factors" explain these patterns – specifically related to the clinical detection of developmental delay in early childhood (Lauritsen et al. 2014).

With clinical detection in mind, there are many factors that could lead to diagnostic variation. In countries with substantial disparities in access to care, such as the U.S., the low rates of diagnosis among Latinos and Blacks, for example, can be attributed largely to insurance-related disparities that hamper referrals (Krauss et al. 2003) and the difficulty with which parents receive access to specialty care (Liptak et al. 2008; Bisgaier and Rhodes 2011). Unfortunately, the early difficulties associated with autism, including behavioral problems that often require a parent who would otherwise work to stay home, may exacerbate access issues associated with insurance disparities, further hindering parents who traditionally have worse insurance (Kogan et al. 2008) or even access to primary care (Newacheck, Hughes, and Stoddard 1996).

Interestingly, for ADHD, when insurance status is controlled for, nonpoor children continue to be much more likely to receive treatment compared to poor children (Bussing et al. 2003), which may suggest factors occurring at the clinic. For example, the amount of trust parents have for their physicians (Jegatheesan, Fowler, and Miller 2010) has been shown to be associated with whether families obtain important services for their children, which can be related to whether families have their own family physician. Potentially relatedly, among parents with mental health concerns for their children, Sayal et al. (2010) found that shorter appointment times were a barrier for parents because they were less likely to initiate delicate topics related to their children's problems. And among children with autism in the U.S., immigrant parents were

more than twice as likely as nonimmigrant parents to complain that physicians do not spend enough time during the appointments (Lin, Yu, and Harwood 2012).

During the actual appointments, communication between parents and physicians also matters. For example, several studies have shown that whether physicians even take note of conspicuous symptoms depends heavily on whether parents discuss their concerns or initiate complaints about their children (Palmer et al. 2005; Dulcan et al. 1990). Although parents consistently acknowledge the importance of discussing concerns with their physicians, most parents fail to actually do so (Horwitz, Leaf, and Leventhal 1998).

## DATA

### *The Costa Rican Healthcare System*

The Costa Rican Health Ministry is the "steward" of the healthcare system, which includes the Social Security Fund (Caja Costarricense de Seguro Social), widely known as the "Caja"; the National Insurance Institute, which provides private insurance plans; and a small number of private providers (Muiser et al. 2012). The Caja was founded in 1941 and is one of the few national organizations in Latin America to provide near-universal health coverage, serving nearly 90% of the population (Muiser et al. 2012) and accounting for health outcomes that essentially mirror those in the U.S.[37] (e.g., World Health Organization 2013).

Prior to the 1990s, the system had been highly inefficient and filled with redundancies; with this in mind, World Bank loans mandated a series of reforms in the 1990s (Rosero-Bixby 2004). The EBAIS clinic was created as the Caja's first level of care, with each clinic designed to be managed by a physician and to serve approximately 1,000 households; by 2000, there were 664 EBAIS facilities nationwide, and the percentage of individuals with inadequate access to

[37] For example, in 2011, life expectancy was 79.0 in both Costa Rica and the U.S., and the under-5 mortality rate was 10/1,000 in Costa Rica and 8/1,000 in the U.S. (World Health Organization 2013).

healthcare services had dropped from 22% in 1994 to 13% (Rosero-Bixby 2004). Half of the population lived within 1 km of an outpatient facility and within 5 km of a hospital, with the worst access to services occurring primarily in a large rural region over two hours south of San Jose (Rosero-Bixby 2004).

EBAIS clinics are complemented by a number of private facilities, including five private hospitals; however, the private facilities only provide primary and secondary care (Muiser et al. 2012). For cases that require specialty care, the private insurance plans and EBAIS clinics refer directly to hospitals that are managed by the Caja, including 20 regional hospitals and six specialized hospitals in the San Jose metropolitan area (Muiser et al. 2012). The HNN is one such hospital.

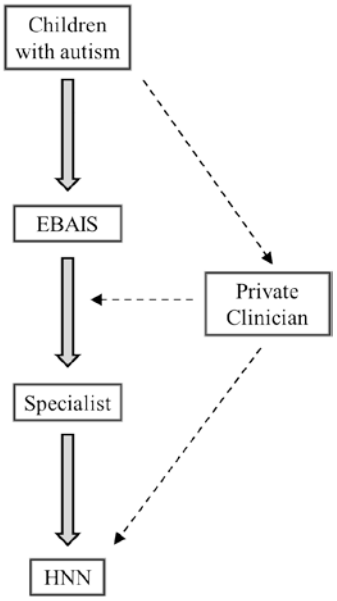
One complication associated with the Costa Rican healthcare system is that the Health Ministry regulations are somewhat unclear, so coordination between the public and private sectors is largely informal (Muiser et al. 2012). For example, physicians are allowed to work part-time in the public sector while also working in private clinics, but the rules governing their referrals between the sectors are ambiguous. Even though the Caja provides free healthcare to much of the population, one of the primary complaints about the public system is that there can be extremely long waiting times, so some patients are inclined to use the private system to avoid long waits (Rosero-Bixby 2004).

*The referral process*

With the end of the explicit recruitment efforts for the 2005 genetic study on autism, there are two pathways children can follow to the developmental unit at the HNN (Unger et al. 2008): (1) The state healthcare pathway, in which children have universal access (Rosero-Bixby 2004), involves consulting a general physician at a local EBAIS clinic (level 1), being referred to

a specialist (level 2) at a local public hospital or clinic, and then being referred to the HNN for a diagnosis (level 3); (2) The private clinician pathway involves consulting a practitioner who is permitted to refer to the HNN, which includes clinicians who also work within the public system. In cases in which private clinicians do not work in the public system, the clinicians are known to write an informal referral note to be taken by parents back to the EBAIS clinic or specialist, where the public physicians provide a formal referral (see Figure 7).

**Figure 7** Referral pathways for children who are eventually diagnosed with autism at the HNN in Costa Rica.



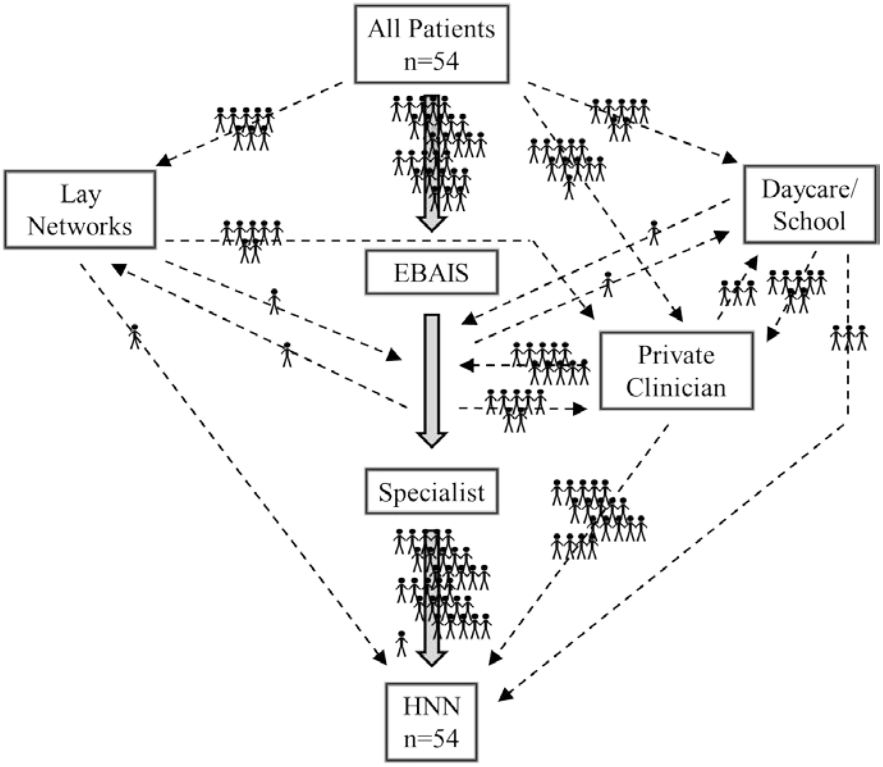

The actual pathways of families, including consultations with lay networks, are shown in Figure 8, where each child is represented and followed throughout the diagram until they arrived at the HNN (note that some children followed a rather circuitous route, sometimes involving double-counted steps).

Notably, most patients (n=28) first consulted a clinician at an EBAIS clinic,[38] but before arriving at the hospital from the public pathway (n=31), there was considerable movement from

[38] EBAIS clinics are distributed throughout the country, including in small towns that in other settings would likely be associated with very limited access to primary care.

the public to private (n=7) and from the private to public (n=10). The former, outgoing movement, commonly involved complaints that EBAIS physicians were inattentive to the symptoms of concern, whereas the latter, incoming movement, included patients who first visited private clinicians that were not allowed to refer directly to the HNN.

**Figure 8** Professional and lay referral pathways for 54 patients who were eventually diagnosed with autism at the HNN in Costa Rica.




Among the 10 patients who moved from private to public care, several were first informally referred from daycare/school (n=7) or from lay networks (n=7). Importantly, the parents who consulted lay networks did so primarily to decide which private clinician to consult with. These parents consulted with family (n=5), a neighbor (n=1), and a friend (n=1); the one patient whose parents consulted their lay network before pursuing public care was the mother mentioned in Chapter 3 who visited her language therapist cousin in a nearby rural town. The town is located far south of San Jose, where there is a noticeable lack of local specialty care.

Among the 11 families that went directly to a private clinician, nine (81.8%) lived in metropolitan areas; on the contrary, among the 28 families that first sought help on the public route, only 15 (53.6%) lived in metropolitan areas; the remaining families lived at least two hours from San Jose. The 11 families that first pursued private care did not often recall their reason for pursuing a particular clinician, although they did express having done so to avoid the delays in the public system.

*Predicting symptom recognition and time to the HNN*

The first models of interest predict the age at symptom recognition ($\bar{x}$=16.03 months[39]) for the 54 patients whose parents were interviewed. Explanatory variables include parental education and income[40] (SES); severe symptoms, comorbidities, and autism symptoms[41] (patient characteristics); number of people in the household, older sibling, the presence of the father, and whether the mother works (household characteristics); parental knowledge of autism, whether the parents knew individuals with autism before the diagnosis, maternal age, and paternal age (knowledge characteristics); and distance from the HNN, population density, and the percentage of households living in poverty (district characteristics). All patient characteristics, household characteristics (with the exception of people in the household), and knowledge variables (excluding parental age) are dichotomous, and the remaining variables are continuous.

We expect several patterns. SES and patient characteristics should be significant predictors throughout. Household characteristics may be significant if parenting experience helps parents to identify symptoms (e.g., having more people in the household and an older sibling

[39] This age is right in the middle of the range reported in the U.S. by Frith and Soares (1993) and very similar to the 18 months reported by Howlin and Asgharian (1999).

[40] Household income in hundreds of dollars (US) per month.

[41] Autism symptoms were classified as behaviors that are typically (and specifically) associated with autism, such as hand flapping, walking on toes, or extreme social delay.

may mean that developmental delays become more conspicuous); also, the father being home or the mother working may indicate household types that are associated with higher income families.

If the findings from Chapter 3 are indicative of larger patterns that knowledge among parents and information spread do not influence the diagnosis, then the knowledge variables, especially knowledge of autism and knowing individuals with autism, should not be significant. (Maternal and paternal age would not necessarily have to be related to knowledge.) Finally, absent an information effect, district characteristics should not influence the age of symptom recognition.

Table 4 shows the five models predicting age at symptom recognition.[42] As expected, there is limited evidence for patterns that would indicate the importance of parental knowledge or information spread: the relatively small coefficients of knowledge of autism and knowing individuals are nonsignificant, and even autism symptoms becomes nonsignificant in models 3, 4, and 5, with the slope dropping to less than a one-month effect on symptom recognition. Having an older sibling (p=0.005) and the father being home (p=0.051) are important in model 3, where the predicted time of symptom recognition decreases by 7.8 and 6.9 months, respectively, but these effects appear to be attenuated when maternal age is included in model 4. This is likely because maternal age is associated with both variables: specifically, older mothers are more likely than younger mothers to have multiple children, and fathers are more likely to be in the household when the mothers are older.

[42] Initially concerned about possible type I errors associated with adding many predictors with a relatively small *n*, the models were first run unnested. We then kept SES in all models, and finally, we nested all models such that the final model included all 16 predictor variables. The results were consistent in terms of which variables appeared to be meaningful, so only the nested models are reported. Note that with the current sample size, we are cautious about making too much of the estimates.

**Table 4** Five models predicting age (months) at first symptom recognition among 54 children later diagnosed with autism, Costa Rica (SEs in parentheses).

| Variable | 1. SES | 2. Patient char. | 3. Household char. | 4. Knowledge | 5. District char. |
|---|---|---|---|---|---|
| Parental education | -0.41 (0.24) | -0.41 (0.24) | -0.89**(0.25) | -0.83**(0.23) | -0.73**(0.23) |
| Income | 0.49 (0.27) | 0.45 (0.27) | 0.79**(0.25) | 0.66**(0.23) | 0.66**(0.22) |
| Severe symptoms | | 0.00 (2.85) | 4.77 (2.85) | 5.34* (2.47) | 5.57* (2.47) |
| Comorbidities | | -5.83* (3.17) | -7.65**(2.82) | -10.28**(2.53) | -10.07**(2.46) |
| Autism symptoms | | 4.74* (2.74) | 2.87 (2.49) | 0.84 (2.20) | 1.10 (2.20) |
| No. in household | | | 0.47 (0.91) | 0.41 (0.79) | 0.42 (0.77) |
| Older sibling[a] | | | -7.80**(2.64) | -3.37 (2.58) | -3.73 (2.51) |
| Father home | | | -6.89 (3.44) | -1.81 (3.20) | -2.43 (3.14) |
| Mother works | | | 5.26 (3.66) | 5.76 (3.19) | 5.39 (3.19) |
| Knowledge | | | | 3.87 (2.36) | 2.48 (2.42) |
| Knew individuals | | | | 1.25 (2.47) | 0.56 (2.55) |
| Maternal age[b] | | | | -0.65**(0.23) | -0.55* (0.23) |
| Paternal age | | | | 0.04 (0.26) | -0.03 (0.26) |
| Distance (km) | | | | | -0.05 (0.05) |
| Pop. density[c] | | | | | -0.84* (0.35) |
| Poverty | | | | | 0.10 (0.12) |
| Intercept | 20.13 | 19.29 | 30.24 | 39.65 | 39.77 |
| Model F | 1.92 | 2.12* | 3.92** | 4.27** | 4.12** |
| Partial F | | 2.16 | 5.25** | 3.30* | 2.02 |
| R2 | 0.070 | 0.181 | 0.445 | 0.587 | 0.647 |
| Adj R2 | 0.034 | 0.095 | 0.332 | 0.450 | 0.490 |
| BIC | 413.51 | 418.64 | 413.53 | 397.07 | 400.74 |

*p<0.05; **p<0.01.

[a] Age at first symptom recognition for patients with older siblings (n=29) and patients without older siblings (n=25): 12.97 months vs. 19.58 months (two-sided t-test, p=0.019), respectively.

[b] Age of the mother at the time of birth of the child later diagnosed with autism. As expected, older mothers were more likely to have had previous children (i.e., an older sibling); for example, 64% of mothers over 35 and 79% of mothers between 29 and 35 had had a previous child, whereas only 13% of young mothers and 48% of mothers in their early-to-mid 20s had had a previous child (Chi2=9.69, p=0.021).

[c] Thousands of persons per square km.

In the district characteristics model (model 5), population density is significant, although the district characteristics as a whole do not improve the fit of model 4 (partial F=2.02; p=0.129), and BIC indicates no improvement over model 4. It is certainly plausible that parents in urban areas are slightly better than parents in rural areas at detecting symptoms (every additional 1,000

people per square km would be associated with less than a month of earlier detection), but this cannot be flushed out with the current data. However, population density was not significant in a bivariate model, when district characteristics were considered alone, or when district characteristics were considered only with SES.[43] Thus, district characteristics appear to contribute little to improving model 4.

Thus, model 4 is the preferred model (partial F=3.30; p=0.020), although maternal age appears to be driving much of the improvement over model 3. Altogether, model 4 explains 58.7% of the variation in symptom recognition (Adjusted $R^2$ = 0.450). Maternal age (p=0.008) appears to be the strongest predictor: specifically, a 35-year-old mother is predicted to notice symptoms nearly 10 months earlier than a 20-year-old mother, presumably because older mothers have more experience observing typical developmental milestones in other children. This is consistent with previous studies, where older mothers are better at identifying symptoms compared to younger mothers (Russell et al. 2011). It is also possible that an older child and the presence of the father are related to maternal age.

In addition to maternal age, other important predictors of symptom recognition include education (p=0.001), income (p=0.006), severe symptoms (0.037), and comorbidities (p<0.001). An additional eight years of parental education is expected to decrease the time of symptom recognition by 6.6 months, and the presence of comorbidities, as expected, decreases the predicted time by 10.3 months. However, an extra $500 of monthly household income is expected to *increase* the time of symptom recognition by 3.3 months. Likewise, having severe symptoms also increases the predicted time of symptom recognition by 5.3 months. For income, it is possible that some families were somewhat delayed in recognizing symptoms because both

[43] It seems very likely that population density is related to parental education and maternal age, as parents in the city may have relatively more education and also tend to delay childbearing.

parents were working, but even if the estimate is accurate, the effect is relatively small. Regarding severe symptoms, the inverse relationship may have been a result of how the interview question was worded. Parents of children with more severe symptoms may have attempted to indicate the first autism symptoms rather than the first symptoms overall, inflating the estimate.

Finally, also in model 4, mother works (p=0.078) contributes a relatively large effect. In households with working mothers, the first symptom recognition is delayed by 5.8 months, which could be explained by busy working mothers being less able to discern minor developmental anomalies than their stay-at-home counterparts.

* * * * *

The second models of interest predict the time (in months) between symptom recognition and the first visit to the HNN developmental unit ($\bar{x}$=45.01 months[44]). All of the five sets of variables from the symptom recognition models are included, and in addition, referral variables have been added as model 6. The referral variables include a dichotomous variable for whether private care was ever pursued and a continuous variable for the number of referral steps needed to reach the HNN ($\bar{x}$=3.26; max: 7).

The expectation is that income will be the biggest driver of a decreased referral time, as parents with more money may be able to bypass delays in the public system to seek a referral via the private sector. Patient characteristics should also decrease the referral time, as symptom severity should speed the referral process. If the mechanism for detecting cases begins at the clinic rather than among parents, then household characteristics and knowledge should not speed up the referral. However, district characteristics should be important in reducing the time, as

[44] This amount of delay is consistent with delays observed in Sweden by Sivberg (2003) and slightly longer than the delay reported in the U.S. by Wiggins, Baio, and Rice (2006).

distance to the HNN and population density are strongly associated with living in the San Jose area near the hospital, which is also where there is a high concentration of second-level clinics and hospitals that may be common sources of referrals to the HNN. For parents living in rural areas far from the HNN, there may only be one option for obtaining a referral (the local EBAIS clinic), that is, unless the parents are willing to travel the long distance to the capital. Finally, referral steps (model 6) should also reduce the referral time, as parents pursuing private care may be able to avoid delays.

Table 5 displays the results for models predicting the referral time. The results are notable for the absence of a good fit: none of the model F tests are statistically significant. Only for the SES-alone model is the F test close to being significant ($p=0.054$). As expected, household characteristics and knowledge do not speed the referral time, but contrary to expectations, district characteristics and referral variables are unhelpful. Having severe symptoms does appear to decrease the referral time ($p=0.051$), and the effect is strong (over 1 year faster with severe symptoms). But again, the patient characteristics model is not significant.

**Table 5** Six models predicting time (months) between symptom recognition and first visit to the HNN Developmental Unit among 54 children later diagnosed with autism, Costa Rica (SEs in parentheses).

| Variable | 1. SES | 2. Patient char. | 3. Household char. | 4. Knowledge | 5. District char. | 6. Referral |
|---|---|---|---|---|---|---|
| Parental ed. | 0.44 (0.53) | 0.70 (0.54) | 0.58 (0.59) | 0.62 (0.58) | 0.51 (0.54) | 0.34 (0.55) |
| Income | -1.40* (0.60) | -1.62** (0.60) | -1.63* (0.66) | -1.75** (0.63) | -1.57* (0.61) | -1.41* (0.66) |
| Severe sym. | | -12.96 (6.47) | | | | |
| Comorbidities | | 7.53 (7.21) | | | | |
| Autism symp. | | 1.00 (3.42) | | | | |
| No. in house | | | 1.42 (2.33) | | | |
| Older sibling | | | 10.89 (6.94) | | | |
| Father home | | | 5.00 (9.16) | | | |
| Mother works | | | 7.73 (8.79) | | | |
| Knowledge | | | | -3.12 (6.83) | | |
| Knew ind. | | | | 9.97 (7.54) | | |
| Maternal age | | | | 1.10 (0.69) | | |
| Paternal age | | | | 0.10 (0.69) | | |
| Distance | | | | | -0.18 (0.15) | |
| Pop. density | | | | | -0.99 (1.10) | |
| Poverty | | | | | -0.29 (0.37) | |
| Private care | | | | | | 0.28 (7.63) |
| Referral steps | | | | | | -2.12 (2.18) |
| Intercept | 47.98 | 49.59 | 28.40 | 11.69 | 61.88 | 56.65 |
| Model F | 3.10 | 2.20 | 1.70 | 2.21 | 1.89 | 1.75 |
| AIC | 494.2 | 495.2 | 497.8 | 486.4 | 496.7 | 497.2 |
| R2 | 0.108 | 0.187 | 0.178 | 0.224 | 0.165 | 0.125 |
| Adj. R2 | 0.073 | 0.102 | 0.074 | 0.122 | 0.078 | 0.054 |

*p<0.10; **p<0.05; ***p<0.01.

One other pattern needs emphasis: the signs for parental education and household income switch as compared to the symptom recognition models. Specifically, education increases the referral time, whereas income decreases it. Income does appear to be tied to seeking private care, but the private care variable is far from being significant and does not drastically change the

income coefficient. Given that even the SES model is nonsignificant, though, we hesitate to overinterpret this pattern.

*Mild cases*

Out of 54 cases, 22 were classified as mild. Again, mild was conceptualized simply as cases in which the children either already were attending or later attended regular school (i.e., for interviewees with children diagnosed in previous years). This cutoff was used because the intention was to determine why difficult-to-detect cases are geographically distributed in clusters, and fitting in with typically-developing children is a good proxy for these cases.

We consider the same variables that were included in the models from the previous section, beginning with the continuous variables, for which we use two-sided t-tests to compare the means between moderate/severe and mild cases. If our interpretation of the patterns thus far is correct—that information among parents does not drive the identification of mild cases—then education and income should be similar between moderate/severe and mild cases. Next, if older mothers are better at detecting developmental abnormalities, then mothers should be older on average in the mild case group. The only other expected difference is referral steps: if parents are driving the diagnosis of mild cases, then we might expect additional steps among mild cases, as they require additional effort among the parents.

Table 6 shows the results of the comparisons between moderate/severe and mild cases for the continuous variables. There are two surprises: first, maternal age is similar between the two groups (28.3 vs. 27.8 years; p=0.785), so there is no evidence that older mothers are better at identifying difficult-to-detect behavioral anomalies in their children. Second, there are actually more referral steps for the moderate/severe cases (3.72 vs. 2.59; p=0.006), and this is the only statistically significant difference. One possibility is that moderate/severe cases are associated

with more co-morbidities than mild cases, and these co-morbidities require additional appointments that push the number of steps higher (there is some evidence for this in Table 7). Table 6 shows no other surprises, and as expected, education is somewhat similar between moderate/severe and mild cases.[45] Income appears to be slightly higher for mild cases ($911 vs. $759), and distance to the HNN is slightly lower for mild cases (20.50 km vs. 29.15 km), but these differences are not statistically significant.

**Table 6** Means and standard deviations of various family and patient characteristics between moderate/severe (n=32) and mild (n=22) cases of autism, Costa Rica.

| | **Mod/severe** | **95% CI** | **Mild** | **95% CI** | **t-test (2-sided)** |
|---|---|---|---|---|---|
| Parental education (years) | 20.03 | 17.06-23.00 | 18.91 | 15.87-21.95 | t=-0.53 p=0.601 |
| Household income | $759 | $536-981 | $911 | $573-1,249 | t=0.81 p=0.423 |
| No. in household | 4.44 | 3.77-5.10 | 4.36 | 3.96-4.76 | t=-0.17 p=0.863 |
| Maternal age (years) | 28.28 | 25-88-30.68 | 27.82 | 25.55-30.09 | t=-0.27 p=0.785 |
| Paternal age (years) | 32.78 | 30.45-35.12 | 31.67 | 28.84-34.49 | t=-0.62 p=0.536 |
| Distance (km) | 29.15 | 18.68-39.62 | 20.50 | 11.05-29.95 | t=-1.19 p=0.239 |
| Pop. density | 2.80 | 1.65-3.95 | 3.00 | 1.35-4.66 | t=0.216 p=0.830 |
| Poverty | 22.33 | 18.82-25.85 | 20.85 | 17.06-24.64 | t=-0.577 p=0.567 |
| Referral steps | 3.72 | 3.18-4.26 | 2.59 | 2.03-3.15 | t=-2.88** p=0.006 |

For the dichotomous variables, two-sided z-tests are used to compare the proportions of moderate/severe cases with those of mild cases. Table 7 shows several differences of interest.

[45] Education may be pulled up slightly for mild cases, but if there is a real effect, it is likely rather small, and a bigger n would be needed to detect it.

First, many more mothers work outside the home in the mild group compared to the moderate/severe group (54.5% vs. 18.8%; p=0.006), which is likely a result of moderate/severe cases demanding extra care, requiring that some mothers either leave their jobs or decide to stay home as care providers. This is also supported by the interview data: when asked about their job, many mothers reported having worked in the past, and when asked directly, the majority said that they took care of the house "because of the child." Unsurprisingly, these mothers who stay home also tend to have less money than mothers who work.

**Table 7** Percentages of various family and patient characteristics between moderate/severe (n=32) and mild (n=22) cases of autism, Costa Rica.

| | **Mod/severe** | **95% CI** | **Mild** | **95% CI** | **z-test (2-sided)** |
|---|---|---|---|---|---|
| Comorbidities | 31.3% | 15.2-47.3% | 18.2% | 2.1-34.3% | z=-1.08 p=0.282 |
| Autism symptoms | 56.3% | 39.1-73.4% | 54.5% | 33.7-75.4% | z=-0.12 p=0.901 |
| Older sibling | 56.3% | 39.1-73.4% | 50.0% | 29.1-70.9% | z=-0.45 p=0.651 |
| Father home | 84.4% | 71.8-97.0% | 68.2% | 48.8-87.6% | z=-1.41 p=0.160 |
| Mother works | 18.8% | 5.2-3.2% | 54.5% | 33.7-75.4% | z=2.74** p=0.006 |
| Knowledge | 53.1% | 35.8-70.4% | 50.0% | 29.1-70.9% | z=-0.23 p=0.821 |
| Knew individuals | 28.1% | 12.5-43.7% | 22.7% | 5.2-40.2% | z=-0.44 p=0.657 |
| Private | 56.3% | 39.1-73.4% | 45.5% | 24.6-66.2% | z=-0.78 p=0.435 |

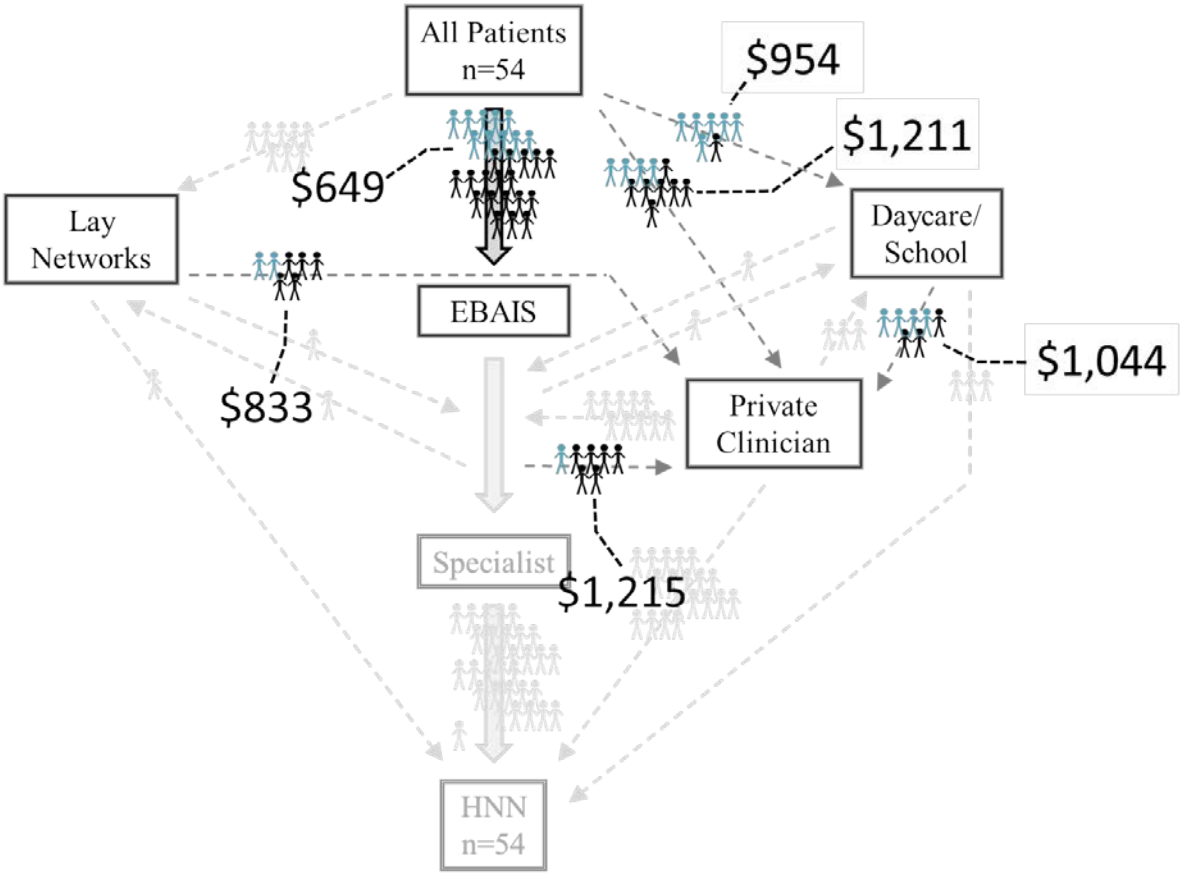
Next, as expected, there are more comorbidities in the moderate/severe group, although autism symptoms are almost identical between the groups. Interestingly, there are more fathers in the households of moderate/severe cases compared to the mild ones (84.4% vs. 68.2%), which is also inconsistent with the idea that the detection of mild cases is more likely if there are two

parents in the home. However, this difference is too small to be statistically significant with the current sample size. Finally, inconsistent with the parental action mechanism, autism knowledge, knowing other individuals, and seeing a private physician are somewhat higher in the moderate/severe group, although these differences are far from statistically significant.

*Referral pathways of mild cases*

We want to return to the figure that shows the public and private referral pathways that lead to the HNN. Figure 9 highlights all patients incoming to EBAIS and all patients incoming to the private pathway, with other pathways grayed out for ease of visualization. Note that mild cases are shown in blue.

**Figure 9** Professional and lay referral pathways and average household incomes for 54 patients, mild (shown in blue) and moderate/severe, who were eventually diagnosed with autism at the HNN in Costa Rica.



First, notice that the average monthly incomes of families that at some point pass through the private pathway are substantially higher than those of families that started out in the public pathway (among the public families, those with more money were more likely to leave the public pathway to see a private clinician). And in fact, in logistic regression models predicting private care, the only significant variable was income (for the SES model, income OR: 1.39; p=0.008; data not shown). Families with more income are the ones that are more likely to see a private clinician.

Next, notice that the proportion of mild cases appears to be similar between private and public, especially if you note the proportion in the ones who started public (10/28, 35.7%) versus the ones who started private (4/11, 36.4%). However, there are also four mild cases that were referred to private clinicians from school, and there are another two cases that were referred from lay networks. Finally, the parents of one of the mild cases from the public pathway, a case in which the child was displaying “weird gestures,” complained that the public pediatrician “didn’t give enough attention” to their complaint, so they visited a private clinician three times before being referred back to the public system. Given that one mild case was referred directly from school without entering either the public or private pathway, the proportion of mild cases that went private (12/22, 54.5%) is higher than the proportion of mild cases that went public (9/22, 40.9%).

This difference is not substantial if the expectation is that the proportions should be the same, which would be the case if (a) there were an equal proportion of families pursuing public versus private care, and (b) the detection probability, or likelihood of referring onwards to the HNN, were the same between the public and private sectors. These may not be reasonable assumptions. Regarding families pursuing public versus private care, Muiser et al. (2012)

suggest that the number of families who purchase private healthcare services is as low as 30%, and given that the limited data on this topic describe adults who do not have the universal healthcare that children do, the estimate could even be inflated for children. Certainly in our experience, it is a minority of families that pursue private care for their children.

Table 8 displays four hypothetical scenarios of the proportions of families that pursue public versus private care, as well as detection probabilities of those cases being identified as needing further care and being referred onwards through the system. The first two scenarios show equal detection probabilities, where public and private clinicians are equally likely to identify cases. If most families (say 60%) always remain in the public sector, which we believe is a conservative estimate, then there would be three mild cases from the public sector for every two from the private sector (Scenario 1). With a less conservative estimate of the proportion using solely private (70%), again with equal detection probabilities, there would be seven cases coming from public for every three private (Scenario 2). Scenario 3 shows the same 70% estimate of public use but with double the detection probability from the private sector, which follows our hypothesis that longer appointment times may make private physicians more likely to detect mild problems in children. Still, Scenario 3 has slightly more children being referred through the public sector. Only with Scenario 4, which uses the conservative estimate of 60% of families remaining in the public sector, do the cases arriving at the HNN (three cases public for every four private) look similar to what we observed in our small sample, where 12 cases came from private and nine from public. If the percentage of parents who pursue private care is even lower than 40%, which we believe it is, then the detection probabilities would need to favor private care even more to lead to more cases arriving from the private sector.

**Table 8** Four hypothetical scenarios showing the expected percentage of mild cases arriving at the HNN for a possible diagnosis, given two possible proportions of general population families pursuing private care and two detection probabilities of referring to the HNN, Costa Rica.

| | Families in general population in pathways | Detection probability/likelihood of referral | Percentage of cases arriving at HNN |
|---|---|---|---|
| **Scenario 1: conservative estimate, equal detection** | | | |
| Public | 60% | .25 | 15% |
| Private | 40% | .25 | 10% |
| **Scenario 2: moderate estimate, equal detection** | | | |
| Public | 70% | .25 | 17.5% |
| Private | 30% | .25 | 7.5% |
| **Scenario 3: moderate estimate, unequal detection** | | | |
| Public | 70% | .25 | 17.5% |
| Private | 30% | .50 | 15% |
| **Scenario 4: conservative estimate, unequal detection** | | | |
| Public | 60% | .25 | 15% |
| Private | 40% | .50 | 20% |

Returning to Figure 9 with this in mind, one reasonable possibility is that parents who pursue private care are inadvertently increasing their chances of receiving a referral for their children.

## DISCUSSION

Chapter 3 provided theories to explain the absence of information-sharing at a scale that could explain spatial diagnostic clustering, and what was left was the possibility that private physicians may be more likely to refer than public ones. There was some evidence for this in the network connections shown in Chapter 3, where a handful of private clinicians provided more than their share of referrals. One neurologist in particular saw a large number of the cases and advised many of those parents in how to obtain referrals to the HNN; most of those cases were

referred to the neurologist by schools and daycares. This is consistent with two U.S. studies: Liu and Bearman (2015) found evidence that proximity to schools was a predictor of an increased risk, and Kalkbrenner et al. (2011) found that proximity to neurologists and psychiatrists was a better predictor of an autism diagnosis than proximity to primary care physicians. One possibility is that specialists are more willing to refer borderline cases than are primary care physicians, and there is some evidence that they recognize symptoms that primary care physicians fail to recognize or do not take as seriously.

In the interview data from Chapter 3, several parents mentioned not wanting to wait in the public system for the series of referrals that would be necessary to eventually have an appointment at the HNN, and others complained that EBAIS was not adequately attending to their concerns or had encouraged them to wait and see. We mentioned that EBAIS appointments are relatively short compared to those of specialists (the private physicians consulted were all specialists), where the appointments are typically 30 minutes or more. The extra time allotted to specialists may lead to improved detection of early language and developmental problems.

In our sample, the wealthiest parents were more likely to purchase private services for their children, which is in line with other reports (Cercone and Jimenez 2008). For the other parents, private care was primarily sought when public care was seen to be inefficient or when a private specialist was suggested by a trusted source. That 28 of 54 patients (51.9%) in our sample eventually consulted with a private clinician is higher than expected, especially given that the symptoms were similar between the public and private groups. However, a relatively high detection probability in the private sector relative to the public sector could explain why the proportion is in favor of private: in short, many additional children remain undetected in the

public system, whereas the private system is sending a high number of cases onwards for further diagnostic scrutiny.

Recall the important patterns from earlier: (1) Table 4 shows that other than maternal age, household characteristics and knowledge fail to explain variation in when parents notice problems in their children, and additional income actually increases the time of recognition; (2) Table 5 shows that only income reduces the time between symptom recognition and the first appointment at the developmental unit at the HNN; (3) Tables 6 and 7 show that mild and moderate/severe cases are mostly similar for our variables of interest, but families of mild cases have slightly higher incomes, live a bit closer to the HNN, and are far more likely to involve a working mother (54.5% vs. 18.8% are working). Combined with the findings that private care is more likely to be sought among higher income families, we can add to our narrative: mothers of low severity cases are more likely to still be able to work compared to moderate/severe cases; these working mothers have more money, but less time, so they are a bit slower at identifying symptoms in their children. However, they use their extra money to pursue private care more frequently, which reduces the referral time slightly. It happens that incomes are higher near the hospital, and there are also more private care options in the Central Valley than elsewhere. This causes variation in that there are more working mothers from higher income households near rather than far from the HNN, which would explain why the mild cases live slightly closer to the HNN.

This narrative is in line with findings from Chapter 3, where parents consistently suggested pragmatic reasons for pursuing private care, including that public appointments involve long waiting times; we also mentioned that some patients pursue private care because the appointment times are more predictable, so if a working mother must take time off work for an

appointment, a private appointment can be scheduled precisely in advance and is predictable in terms of how much time will be needed. On the contrary, a public appointment can take from 30 minutes to most of the day, depending on the waiting times on a given day.

* * * * *

The patterns presented in this chapter may explain why several of the previously identified spatial clusters are located in higher income locations, where families pursue private care to avoid delays in the public system, and in doing so, they receive additional medical scrutiny because the appointment times are longer and potentially more focused, simply providing them with more physician face-time. Alberto's case from Chapter 3 may be illustrative, where his low income and relatively uneducated parents went to a private clinician after a dentist suggested it. The psychologist there spent the entire appointment considering Alberto's behaviors. On the contrary, at an EBAIS appointment, clinicians have limited time for their appointments, and EBAIS clinicians face constraints on what they can address because of public health mandates such as Dengue fever and nutrition education.

During the clinical encounter, there is additional evidence of a face-time benefit for private clinicians: when parents are first interviewed during consultations at the HNN, they overwhelmingly indicate that "everything is fine" at home, apparently inhibited about complaining about their children's behaviors. Only when asked, "Is your child ever aggressive?" do parents describe the problem behaviors, sometimes detailing daily aggressive behavior that is actually their primary concern. Some children display the aggressive behaviors after 30 minutes or more of the clinician speaking with the parents, only after the children become impatient from sitting so long. These behaviors are often unobservable in the early minutes of any appointment, making even the extremely aggressive child appear tranquil at first.

There is no reason to think that the same thing does not also happen at EBAIS, with the only difference being that the clinicians there may not have time to obtain more accurate answers or observe concealed behaviors. With this in mind, there is simply no evidence that EBAIS physicians are different than the private ones. As an effort to staff rural clinics, medical students receive free tuition in exchange for being placed around the country. The important difference is that private clinicians are able to focus on identifying and addressing the complaints that necessitated an appointment, whereas EBAIS physicians have other mandates and less time.

Research in primary care has shown that time during the appointment can determine whether certain topics are broached by parents (Sayal et al. 2010; see also Liptak et al. 2008), a finding that may be especially relevant to immigrant families who have difficulty with a non-native tongue (Lin et al. 2012) – a group that has a lower risk of being diagnosed with autism (see Fountain and Bearman 2011; Mandell et al. 2009; Palmer et al. 2010; Zuckerman et al. 2014). In other studies, parents have been shown to have a strong influence in the recognition of symptoms among pediatricians (Palmer et al. 2005; Dulcan et al. 1990; Horwitz, Leaf, and Leventhal 1998; Palfrey et al. 1987; Shevell et al. 2001). In our sample, parents appeared to notice problematic behaviors even when EBAIS clinicians remained unconcerned, suggesting that the problem is with the appointment time rather than with parents being able to notice problems.

The same attentiveness can be observed in private daycares and schools, which comprise the majority of the referrals in Figures 8 and 9. In the private setting, the teacher-to-student ratio is higher, and students who are isolated from their peers are given more attention. In our sample, it was primarily private schools that suggested the neurologist who was so prominent in Chapter 3's data and analysis. In the public sector, students can get lost in the larger groupings, and

teachers may not have the time to notice slightly anomalous social behaviors. Notably, the Desamparados area, where there is a low severity cluster, is saturated with private daycares and schools, and in our sample alone, five patients from that area were referred by schools.

## CONCLUSION

Our data suggest that an increased likelihood to refer in the private sector, both at clinics and daycares/schools, is related to an increased attentiveness to anomalous behaviors that may involve generic symptoms. We mentioned in the last chapter that a similar phenomenon may occur when parents witness early severe problems in their children, leading to heightened surveillance both at home and during routine appointments. In our experience, private clinicians are not more likely than public clinicians to mention autism, but they do appear to notice and show concern for relatively minor language and developmental problems that EBAIS is known to write-off as unproblematic.

Rather than a cul-du-sac effect explaining patterns of autism diagnosis, it seems that the decision to pursue private care has unintended consequences that may explain the clustering of cases. Mothers are much less likely to work when their child has severe symptoms, which means they have less money to pursue private care. However, they also presumably have more time to wait in line at EBAIS. In comparison, mothers who work outside of the household are balancing the need to obtain answers about minor behavioral anomalies in their children (not quite severe enough to necessitate quitting their jobs) with time constraints associated with working. Furthermore, for a minority of the cases but more often for mild cases compared to the moderate/severe ones, the fathers are no longer in the picture, which places further constraints on working single mothers.

There is one additional important factor that was developed in Chapter 3 (and implied in Chapter 1): tight professional networks may have consequences for referrals, and some physicians within those networks may receive a higher-than-average number of incoming referrals and send a higher-than-average number of referrals to the HNN. These tight professional networks appear to have the most consequences in the private system, which is where the referral pathways can be circumnavigated and public delays bypassed. These professional networks in the private sector mean that parents who intend to avoid the public sector for practical reasons—the working mothers with children with mild symptoms especially—experience variation in their likelihood of seeing one of the physicians who refers more cases. What seems to be clear, though, is that mothers are not choosing specific physicians because they know it will get them to the HNN, and the families are not influenced by information about autism or their knowledge of other children who were previously diagnosed. In Costa Rica, spatial clusters of cases of autism appear to be a direct result of seeking private rather than public care, leading to the inadvertent outcome of an increased risk of referral.

# CONCLUSION

The crisis of psychiatry peaked in the 1960s and 1970s and began to wane with the DSM-III in 1980. On one hand, changes in the DSM have led to increased medicalization and pharmaceuticalization (Frances 2013), but on the other hand, the changes have accompanied technological, medical, and epidemiological advances that are associated with improvements in identifying disease etiology. No doubt, we have been in a new era of biomedical psychiatry for some time, yet sociologists of medicine have been slow to adapt. The tendency is for sociologists of diagnosis to eschew consideration of biological or even behavioral factors associated with disease and illness categories, instead preferring to focus exclusively on social mechanisms.

Focusing exclusively on social mechanisms of disease and illness has its roots in the highly influential work of the 1960s and 1970s, when figures such as Thomas Szasz (1960, 1961, 1963, 1970), Erving Goffman (1961, 1963), Irving Zola (1972, 1973), David Mechanic (1972, 1975), and Peter Conrad (1975, 1977, 1979; see also Conrad and Schneider 1980) argued quite compellingly for the value of studying the social aspects of medical and psychiatric outcomes. These arguments came at a time when medicine and psychiatry tended to take the social aspects entirely for granted, whereas today, even the American Medical Association has a policy (H-295.874) to educate its students on the social determinants of health. Thus, the critiques of the

1960s and 1970s were timely and appropriate, whereas some of the same critiques applied today would be superfluous.

Consider the widely cited 1960 essay by Thomas Szasz, “The myth of mental illness”: Szasz, a renowned psychiatrist himself, argued that the concept of a mental illness was essentially flawed. Instead of actually pointing to a “neurological defect,” he argued that the label was used by psychiatrists to point out individuals’ “problems in living,” suggesting that maladaptive behaviors were “inextricably tied to the social” world rather than the biological or neurological world of the individual. This argument was in contradiction to the leading view at the time that all mental illnesses would eventually be understood in neurological and “organic” physical terms (Szasz 1960).

Recall that 1960 had not even seen a decade since the DSM I in 1952, and the widespread use of psychopathic drugs was only just beginning. Mainstream psychiatry was firmly committed to the realm of psychoanalysis and talk therapy, and this was separated quite distinctly from medicine and even neurology. Within psychiatry, suicide was perhaps the only definitive sign of mental illness (e.g., Szatz 1960) and a prominent consideration for treatment. Treatment was only beginning to change: the 1950s involved experimentation with pharmacotherapy, but often only in consort with electroconvulsive therapy, still the leading treatment at the time. Even as late as the mid-1960s, following early indications of its effectiveness (e.g., Denber and Bird 1957), pharmacotherapy for the treatment of depression was weighed heavily against the risk of suicide (Kalinowsky 1964). Drugs, after all, failed to have dramatic and immediate effects, although arguably, pharmacotherapy allowed the patient to become “a participating partner” in recovery (Freyhan 1960:1057).

During that time, pharmacotherapy or electroconvulsive therapy were considered to be successful in the absence of suicide or with the reduction of depressive symptoms, but little was known about how the two treatments worked, much less what was happening psychologically or neurologically for a patient to be at risk of suicide in the first place. Only by the late 1950s was there a plausible hypothesis to explain the treatment effects associated with electroconvulsive therapy (e.g., Rosenblatt et al. 1960). For pharmacotherapy, the mechanism driving the effects was only just being articulated, with suggestions that antidepressants (primarily reserpine) had a stimulatory effect on catecholamines (e.g., norepinephrine), which were observed to be deficient in some depressed patients; this "catecholamine hypothesis of affective disorders" (Schildkraut 1965:509) was just that: a hypothesis.

It was in this context that Szatz argued that mental illness was a myth, that many of the conditions targeted by psychiatrists were rooted not in the brain but in "problems with living." He was very clearly apprehensive of the mainstream view that all conditions would eventually be understood neurologically, but this apprehension was not without merit at a time when mental illness was so little understood.

For depressive symptoms in particular, our understanding of the chemical and neurological underpinnings of the related conditions has substantially improved. First, it is clear from many studies that depression has genetic components that even outweigh environmental ones (Sullivan, Neale, and Kendler 2000). Second, we have come a long way in understanding the neurological underpinnings of depression, which involve neurotransmitter and hormonal irregularities within several areas of the brain, including the hypothalamic-pituitary-adrenal axis and the noradrenergic, serotonergic, and dopaminergic systems; the release of hormones occurs in response to stress, including physical activity and illness, among other things, and depression

occurs when abnormal hormonal responses lead to structural changes in these areas of the brain (Nestler et al. 2012). Specifically, depression can occur when the noradrenergic system is overresponsive to stress while the serotonergic system fails to counteract this stress response; antidepressants, then, inhibit the noradrenergic stress response but promote the serotonergic reaction to it (Ressler and Nemeroff 2000).

Much remains to be known about depression-related conditions and their symptomology, especially given that there continue to be "inconsistencies in the data" and in the effectiveness of antidepressants, including those that are thought to influence parts of the brain not known to be related to depression (Ressler and Nemeroff 1999:1219). However, there is agreement on the source of the anomalies and in the complexity of the chemical and neural interactions involved, and authors point out that one serious problem in identifying any specific mechanism is that even the diagnosis involves many subtypes that are not differentiated in their diagnosis (Ressler and Nemeroff 1999). Yet research is currently capable of disentangling genetic from environmental (and even epigenetic) and neurological mechanisms to describe the spectrum of normal responses to stress (Feder, Nestler, and Charney 2009).

In some sense, then, Szatz's (1960, 1961) critique of the use of the term mental illness remains mildly relevant: for depression, there is a fine line between normal and abnormal responses to stress, and there are social origins and implications for drawing that line in one place rather than another. However, what has changed since the 1950s is that the evidence is now overwhelming that the cause of depressive symptoms does in fact exist in the "psysicochemical processes" (Szatz 1960:113) of the brain – an outcome that Szatz seemed confident would never occur.

Even for a condition with an etiology that continues to be elusive—conversion disorder, formerly known as hysteria—recent advancements suggest that the neurological mechanism is within reach (Harvey, Stanton, and David 2006). For Szatz (1961:155), hysteria exemplified mental illness categories getting out of control, as people could simply fake the symptoms, thereby receiving the special treatments afforded to sick individuals. Today, the diagnosis remains extremely difficult to make, but physiologists and physicians are making progress using functional imaging to better understand brain functioning (Nicholson, Stone, and Kanaan 2010). No doubt, scholars in 50 years will look back at our current knowledge and think us naive, but our current state of knowledge continues to advance in the direction of understanding the neurobiological mechanisms causing anomalous behaviors that are associated with psychiatric diagnostic categories.

**IMPLICATIONS FOR MEDICAL SOCIOLOGY**

Despite his often neutral language regarding psychiatric outcomes, Goffman (1959) was also very critical of the mental hospital. His primary criticisms can be summarized as follows: (1) career contingencies mean that supposed symptoms of mental illness fail to determine whether one will be hospitalized, and many patients are hospitalized unwillingly (125); (2) hospital staff do not employ moral neutrality to patients, and much of what the staff note is actually denigrating and only focuses on the types of negative behaviors that can account for the hospitalization in the first place (137); (3) the custodial role of the hospital is meant to serve people with whom the patient has had some "special kind of trouble," but this does not necessarily serve the community or the patient (1961:353); (4) the hospital is alienating and stigmatizing, both of which can have "more significance for the patient and his personal circle than do his original difficulties" (356); (5) psychiatric staff are ill-equipped to treat mental illness

but nevertheless are a position with even greater influence than the psychiatrist, who only infrequently sees the patient (1961:357).

In the era of *One Flew Over the Cuckoo's Nest*, these criticisms were invaluable, and Goffman was an important early voice to point out the unintended consequences of the psychiatric system. But as we have pointed out earlier, the psychiatric era has changed; progress has been made, and much has been learned about the causes of mental and behavioral abnormalities, not to mention the treatments. Presently, the most prescient criticism for psychiatry may be that too few individuals with mental health problems are being treated by psychiatrists or hospitalized in mental health facilities; as a result of deinstitutionalization, many individuals are being cycled in and out of jails and prisons, accounting for 15% or more of the population of inmates (Lamb and Weinberger 1998; James and Glaze 2006).

So what is the medical sociologist to do? One option is to combine forces with psychiatry and medicine, sharing unique insights that can serve medical progress. For example, the social determinants of health, or Goffman's career contingencies, suggest that patients have unequal changes of being identified and included in studies of the conditions of interest to psychiatrists. Furthermore, as Fava et al. (2014:136) note,

> The current clinical taxonomy in psychiatry, which emphasizes reliability at the cost of clinical validity, does not include effects of comorbid conditions, timing of phenomena, rate of progression of an illness, responses to previous treatments, and other clinical distinctions that demarcate major prognostic and therapeutic differences among patients who otherwise seem to be deceptively similar since they share the same psychiatric diagnosis.

This means that current efforts to understand the neurobiological mechanisms driving anomalous behaviors are impeded by poorly demarcated categories, and funding mechanisms along with limitations in study design tend to encourage focusing on one disease entity, potentially even

when researchers know better than to assume their population of patients is homogenous in terms of etiology.

For autism specifically, one only needs to consider research supported by Autism Speaks, a leading funding agency for autism, to understand the dilemma: in bold at the top of the grants webpage, the foundation is said to support "global biomedical research into the diagnosis, causes, prevention, and treatment of autism" (https://www.autismspeaks.org/science/grants-program; accessed March 24, 2016). The problem is that studies of the causes of autism then rely on samples of cases without considering social aspects of diagnosis, including the possibility that the diagnostic boundaries are somewhat problematic – least of all because diagnostic reliability and even validity remain persistent concerns.

Our recent experience discussing autism with a researcher searching for autism biomarkers is an example of such an approach: On a project that recruits cases simply based on which ones are diagnosed using the ADOS, one researcher, a tenured professor in a chemistry department at a research university, was hard-pressed to imagine that the ADOS could still have reliability problems. Specifically, this researcher discounted the possibility out of hand that two clinicians could disagree about a diagnosis of autism. It is just this kind of ignorance for social aspects of diagnosis that lead to bad study designs and likely sampling problems.

Arguably, the biological and natural sciences are beginning to consider these factors. One example is a recent article (Rice et al. 2013) published in the journal *Public Health Reviews* by several big names in autism research, including an epidemiologist and a physician with the Centers for Disease Control and a clinical psychologist with Autism Speaks. The article emphasizes that autism likely represents multiple conditions with multidimensional causes. Other studies point out that there is considerable overlap—especially in the low severity cases—

with other diagnoses, especially ADHD (Sturm and Fernell 2004). Genetic studies have shown that autism has commonalities and shares risk factors with schizophrenia (Cross-Disorder Group of the Psychiatric Genomics Consortium 2013a), suggesting again that a fruitful approach would be to stop thinking in terms of diagnoses and focus instead on disease causes that may involve overlapping psychiatric categories (Cross-Disorder Group of the Psychiatric Genomics Consortium 2013b).

It seems the missing link in identifying causal mechanisms related to specific conditions is to disentangle biological mechanisms from social and environmental ones. For this to happen, what is critically needed is a collaborative, interdisciplinary approach to understanding biological mechanisms of interest within and between diagnostic categories, and to do so, sociologists of medicine can identify social pathways that inhibit the improved understanding of patient populations. Social pathways start with the diagnostic process, including how variation in diagnosis is created in the first place. The most obvious causes of diagnostic variation involve (a) help-seeking behaviors of parents and (b) detection practices of referring and diagnosing clinicians.

The study design of this dissertation was unique compared to other studies in the sociology of diagnosis, as the approach was to focus on a category in its early stages of adoption in a new setting, allowing us to observe in real time the early stages of an “epidemic.” The work involved collaboration with the primary diagnosing physician and the centralized diagnosing hospital for an entire country, which allowed us to conduct complete medical file reviews to consider factors such as variation in symptomology, including symptom severity, comorbidities, and even family history. By focusing on a small setting with one clinic, the study was able to consider population data, thereby eliminating recruitment biases and clinical variation that exist

in larger settings. Finally, to the extent possible, we also considered environmental causes. The study first tracked the adoption of the autism category (Chapter 1), considered the distribution of cases over time (Chapter 2), diagrammed the social network connections among parents of patients (Chapter 3), and considered referral pathways that could explain aggregate patterns (Chapter 4).

Study design can always be improved, but we learned that autism symptomology per se is not an important feature of the help-seeking process. Pathways to diagnosis do not appear to depend on education or exposure to information about autism. Naive or not, our parents reported early concern for symptoms that would be difficult even for an autism expert to initially associate with the disorder, which is consistent with reports of the first symptoms of concern for parents around the world. Thus, the help-seeking behaviors of our parents were unrelated to autism.

At the clinic, it appears that private physicians are much more likely to refer than public physicians, which may be a consequence of these physicians having more time to notice borderline symptoms in their patients or to discuss sensitive complaints with the parents, who are somewhat reluctant to complain about their children. Parents choose private care because they hope to avoid delays in the public system, and the private system allows working parents to make an appointment beforehand and better plan how much time to take off from work. This is of special interest to sociologists for a category like autism because parents—in Costa Rica and possibly elsewhere—do not appear to orient to the diagnostic category until after the diagnosis. With this in mind, social mechanisms for autism diagnosis transcend the category of autism and relate instead to the types of generic developmental problems parents primarily notice.

For autism patients, at least in Costa Rica, career contingencies often begin with families living in areas with many private physicians; with parents with financial means hoping to avoid delays in the public system; with physicians with fewer institutional constraints having sufficient time to notice behavioral red flags. This suggests the importance of ascertainment and referral practices for understanding variation in autism diagnosis. The next step is to open up the dialogue with the medical and biological sciences.

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