



## Measuring use and cost of health sector and related care in a population of girls and young women with Rett syndrome

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### ABSTRACT

This study measured use and cost of health sector and related services in Rett syndrome and effects of socio-demographic, clinical severity and genetic factors on costs. The study population consisted of individuals with Rett syndrome registered with the Australian Rett Syndrome Database in 2004. Descriptive analysis was used to examine patterns of resource use and costs, and logistic regression to analyse factors associated with higher costs. We found the use of health sector and related resources varied by type of resource. Mean annual cost per case was \$21,158 (range \$238–\$85,776). High cost items included long-term residential care, therapy services out of school and paid home and community care. Factors associated with increasing odds of being above the median cost were clinical severity and the p.R255X mutation. Compared with the youngest cases, cases in the 5–14 year age group and the 15–24 year age group were at lower odds of being above the median cost. Needs relating to health sector and related resources can result in considerable costs for individuals with Rett syndrome. Many households caring for dependents with Rett syndrome are like to be facing undue financial pressures from the additional costs of disability.

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## 1. Introduction

Rett syndrome is a severely disabling neurodevelopmental disorder primarily affecting girls. It is caused by mutations in the *methyl-CpG binding protein 2 (MECP2)* gene (Amir et al., 1999) and has an estimated incidence of diagnosis in 1 in 8500 female births (Laurvick et al., 2006). Although there is great variation in the outcomes for females with this condition (Colvin et al., 2003), Rett syndrome is usually associated with significant physical and intellectual disability (Dyke & Leonard, 2006).

As a result of their unique health needs, children with disabilities generally use more health services than other children (Newacheck, Inkelas, & Kim, 2004). International studies comparing the expenditure on health services of children with or without disabilities have shown that children with disabilities have mean health care expenditures three to four times those of other children (Newacheck et al., 2004; Newacheck & McManus, 1988). Among the children with disabilities, expenditure on health care has tended to be skewed, with a relatively small group accounting for a disproportionate share of total spending (Newacheck et al., 2004; Newacheck & McManus, 1988). A study of the cost of autism in the UK found that factors

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that were associated with a higher annual average cost per child were accommodation type, whether or not the child was intellectually disabled, and age group (Knapp, Romeo, & Beecham, 2009).

Health service use in Rett syndrome has been examined three times. Leonard, Fyfe, Leonard, and Msall (2001) conducted a pilot study in 1999 trialing the use of the Internet for data collection in Rett syndrome. The study questions focused on the burden and impact of Rett syndrome, and factors such as functional abilities, medical needs and the use of medical therapy and accommodation services were explored. Participants consisted of 86 mainly US families. Significant differences were found in the use of medical services by age group, with visits to paediatricians, geneticists and neurologists decreasing with age and visits to general practitioners increasing with age. In a population-based study in Australia, Moore et al. (2005) examined the use and pattern of health service utilisation in Rett syndrome. Multivariate analysis indicated that genetic, clinical and socio-demographic factors were all significant predictors of the utilisation of medical services. In the most recent study, health status and health service use were examined by age and by mutation type (Young et al., 2011). Although health status declined with age, health service use was also shown to decline in parallel. However, these patterns differed by mutation type thus demonstrating important variability.

The purpose of this study was to examine the use and cost of health sector and related services in Rett syndrome in Australia, to compare the cost of service utilisation in Rett syndrome with that of the Australian female population of the equivalent age, and to investigate factors associated with higher costs. No similar data are currently available regarding the added health system costs of caring for people with Rett syndrome, in Australia or internationally, and no studies have explored the cost implications of socio-demographic and other factors. Understanding the nature of health care use and its cost for people with disabilities provides a measure of the burden that disability places on families and the community. From a policy perspective, these data are important for health planning purposes and for developing strategies to protect families against high out-of-pocket expenses in caring for children with disability.

## 2. Methods

### 2.1. Study population

Families whose daughters were registered in 2004 with the Australian Rett Syndrome Database, a population-based registry established in 1993 of Australian cases born since January 1, 1976 (Laurvick et al., 2006), provided the data for this study. The Australian Rett Syndrome Database uses multiple sources of ascertainment, including a parent support group and a rare diseases surveillance unit. Data were collected from families and clinicians. As of 1st January 2004, 268 verified cases had been entered into the database.

### 2.2. Data collection of resource use and costs

After excluding cases submitted in a de-identified form by clinicians, those who had died and those with whom the registry had lost contact, a follow-up questionnaire was administered to families or carers of 221 individuals in the study in late 2004. This questionnaire could be completed on paper or over the internet and gathered information relating to a range of functional and behavioural abilities, medical and therapy service utilisation, schooling and daytime activities, accommodation and special product and equipment needs. This study focused on the use and cost of health sector and related resources required for the care of people with Rett syndrome. These resources included medical and dental services, out-of-school therapy services, paid home and community care, hospital admissions, medications and supplements, non-durable health-related products, therapeutic devices and special equipment, respite care and long-term residential care (Table 1). Families were asked to report on the use of these resources over a specific period of time, say in a typical year or, in the case of therapeutic devices and special equipment, whether the item was used or not. All resource use reported for periods shorter or longer than a year was converted to an annual basis. Families were also asked to report on the cost of resources and the amounts paid out-of-pocket and by third parties. Where the cost reflected market prices (e.g. dental services), no adjustment was made to these costs. For resources that were publicly provided or the cost did not reflect market prices (e.g. prescription medications subsidised by the Federal government), unit costs were obtained from official sources such as the Pharmaceutical Benefits Scheme (PBS) for PBS-listed medications and the National Hospital Cost Database Collection for hospital visits. Missing data were estimated based on unit cost data obtained from official sources or directly from providers or retailers (Table 1). The costs of therapeutic devices and special equipment were converted to an equivalent annual cost using a 5% discount rate and estimates of their useful lifespan. All costs were expressed in 2004/05 Australian dollars, with price indices used to adjust prices expressed in other years (Australian Institute of Health & Welfare, 2007). In addition to the cost as a continuous variable, the cases were classified into two groups based on total annual cost per person: either above or below the median (to compare the 'top half' of cost cases with the 'bottom half').

### 2.3. Comparison of resource use with a previous study

Resource use in this study was compared with the corresponding use reported in 1999 in the study conducted by Moore et al. (2005).

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