

# Client Service Receipt Inventory for rare genetic diseases in Hong Kong: abridged secondary publication

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## KEY MESSAGES

1. The Client Service Receipt Inventory for the rare genetic disease population in Hong Kong (CSRI-Ra) was validated to capture both direct and indirect costs of rare diseases from a societal perspective.
2. Pilot testing of the CSRI-Ra demonstrated moderate to good agreement between utilisation records and electronic patient records, indicating that criterion (concurrent) validity was acceptable.
3. The CSRI-Ra, alone or in combination with

electronic patient records, is valid for health- and social-care planning.

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## Introduction

Genomic medicine increases public awareness of rare diseases (RDs). In Hong Kong, one in 67 people is living with one or more RDs.<sup>1</sup> RDs have broad health- and social-care implications that extend beyond the public health system perspective with regard to effective health- and social-care planning.

The direct costs of RDs include healthcare expenditures; indirect costs include reduced productivity and lost opportunities. There is a lack of standardised tools to collect indirect cost-related data, hindering assessment of the true burden of RDs. In Hong Kong, no tool is available to capture service utilisation data outside of the Hospital Authority. The Client Service Receipt Inventory (CSRI) is a common resource-use measurement tool that evaluates service utilisation patterns to estimate associated costs across healthcare, social care, and community settings. It is a reliable and valid tool for the collection of data related to socio-economic costs.<sup>2-4</sup> The development and adaption of the CSRI in the RD population for the first time in any jurisdiction and language would allow a comprehensive assessment of RD's implications, providing meaningful results to both clinicians and healthcare policy makers. Acknowledging the need for the development of a resource-use measurement tool in Hong Kong, the current study sought to develop, contextualise, translate, and validate the CSRI for the RD population in Hong Kong (CSRI-Ra).

## Methods

The CSRI-Ra was developed through focus group

meetings, in-depth interviews, and data analysis using a thematic framework. Focus group participants included patients with RDs, family members/caregivers of patients, health professionals, and staff from non-governmental organisations (NGOs) and special schools. Two focus group meetings were conducted in August 2019. The first meeting included patients with RDs and family members/caregivers of patients to collect data specific to the RD population. The second meeting included professionals from health- and social-care settings to collect data concerning the accessibility and availability of services and resources. The meetings were conducted in Cantonese and audio- and video-recorded. Collected data were grouped into emerging semantic themes and analysed using a combined inductive and deductive identification approach.<sup>5</sup> Discrepancies were identified and resolved.

Previous CSRIs were used to guide the structure of the CSRI-Ra. All identified themes, subthemes, and variables were grouped into five sections: background information, household and caregiver support, healthcare service and resource utilisation, community support, and education and employment. An expert panel discussion was conducted to ensure that the CSRI-Ra was sufficiently standardised but sensitive to the local context.

The CSRI-Ra was forward-translated to traditional Chinese and backward-translated to English by four independent bilingual researchers. Discrepancies were identified and resolved.

To achieve face validity and semantic equivalence, the content and language of both Chinese and English versions were addressed among

focus group participants through telephone and email discussions. Face-to-face interviews with three additional allied health workers were conducted to ensure that both versions were sufficiently adapted to the Hong Kong setting. Ten bilingual university students were randomly selected to provide suggestions for improvement as the general public. Both versions were validated among eight bilingual patients with RDs and caregivers of patients to assess their alternate-form reliability using intra-class correlation coefficients (ICCs), along with two-way random-effects models based on single ratings and absolute agreement.

The criterion validity of the CSRI-Ra was assessed in a pilot cohort comprising 94 independent patients with RDs and caregivers of patients. Agreement between the self-/proxy-reported utilisation record collected from the CSRI-Ra and the actual utilisation record in the electronic patient record (ePR) was assessed using ICCs with two-way random-effects models. The numbers of accident and emergency (A&E), inpatient, and outpatient visits were compared.

## Results

In the first focus group, there were eight participants covering six RDs with heterogeneity in terms of disability type, treatment needs, and service and resource availability (Table 1). The expert panel considered these six RDs to be sufficiently representative of the RD population in Hong Kong. Additionally, the nine participants in the second focus group had a mean of 12 years of experience related to RDs (Table 2).

Data were analysed using thematic analysis and grouped into the five sections. Two versions were developed: self-completed (patient version) and proxy-completed (caregiver version).

Agreement between the English and Chinese versions of the CSRI-Ra was assessed in eight bilingual participants (two patients with RDs and six caregivers of patients), with a mean of 51.4 days between completion of the two versions. The overall ICC was 0.91 (95% confidence interval [CI]=0.89-0.92), which indicated excellent agreement between the two versions. The ICCs were 0.89 (95% CI=0.86-0.92) for the self-completed version and 0.93 (95% CI=0.91-0.95) for the proxy-completed version.

Agreement between the CSRI-Ra and the ePR was assessed in 94 participants (Table 3). Among them, 54 (57.4%) completed the patient version and 40 (42.6%) completed the caregiver version. In total, 45 RDs were recorded. The overall ICC between the self-/proxy-completed CSRI-Ra and the ePR was 0.69 (95% CI=0.56-0.78), which indicated moderate to good agreement and acceptable criterion (concurrent) validity. Both the patient version (ICC=0.67, 95% CI=0.50-0.80) and the caregiver

TABLE 1. Characteristics of participants in the first focus group meeting (n=8)

Characteristics	No. (%) of participants
<b>Role</b>	
Patient with rare disease (RD)	2 (25.0)
Family member/informal caregiver of patient with RD	6 (75.0)
<b>Sex</b>	
Female	5 (62.5)
Male	3 (37.5)
<b>Age group, y</b>	
18-25	0
26-30	1 (12.5)
31-35	0
36-40	2 (25.0)
41-45	1 (12.5)
>45	4 (50.0)
<b>Education level</b>	
Primary or below	0
Secondary	0
Post-secondary/associate's degree or equivalent	1 (12.5)
Bachelor's degree	2 (25.0)
Master's/doctoral degree	5 (62.5)
<b>RD</b>	
Achondroplasia	1 (12.5)
Marfan syndrome	2 (25.0)
Mucopolysaccharidosis type 6	2 (25.0)
Pompe disease	1 (12.5)
Tuberous sclerosis	1 (12.5)
Williams syndrome	1 (12.5)
<b>Experience completing research questionnaires</b>	
Questionnaires related to RDs	6 (75.0)
Questionnaires related to service use/resource use	2 (25.0)

version (ICC=0.70, 95% CI=0.50-0.80) demonstrated moderate to good agreement. Subgroup analyses of item performance based on the numbers of A&E, inpatient, and outpatient visits revealed acceptable convergent and discriminant validities. Agreement was highest for inpatient services (ICC=0.81, 95% CI=0.73-0.87).

## Discussion

The CSRI-Ra, available at <https://paed.hku.hk/e-form/csri-ra-registration-form.asp>, is validated for use in the RD population in Hong Kong. It is a standardised tool to measure RD costs from a

societal perspective.

The burden of RDs in Hong Kong has been assessed from a health system perspective. In 2015-16, the inpatient cost of RDs was estimated to be HK\$1 594 339 530, corresponding to 4.3% of total inpatient costs for the Hospital Authority.<sup>1</sup> However, the true burden of RDs has not been estimated. The CSRI-Ra enables quantification of the socio-economic burden of RDs, along with informed health- and social-care planning.

The CSRI-Ra relies on participant recall for data collection. The validation stage demonstrated that the CSRI-Ra had acceptable criterion (concurrent) validity, indicating that the collected data are comparable with the ePR. The reliability and validity of CSRIs in other health areas have been demonstrated.<sup>2-4</sup> The CSRI-Ra offers both self-completed and proxy-completed versions, allowing data collection across a wide range of patients with various ages and disease severities. The CSRI-Ra considers the uniqueness of RDs and can be generalised to other contexts and populations, allowing international comparisons of RD burden.

There were some limitations in this study. Despite its acceptable criterion validity, the present study only compared the number of A&E, outpatient, and inpatient visits between CSRI-Ra and ePR. We did not assess hospital and community services in private settings; nor did we explore caregiver support or employment situations; the corresponding datasets were inaccessible or unavailable in ePR and existing databases. Nonetheless, the acceptable agreement between the CSRI-Ra and the ePR reflects the reliability of the tool and suggests its application in areas beyond the health system perspective for the estimation of socio-economic burden related to RDs.

## Conclusion

The CSRI-Ra enables estimation of economic impacts from a societal perspective and better understanding of RD-related service and resource utilisation patterns in Hong Kong, thereby informing health- and social-care planning.

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## Disclosure

The results of this research have been previously published in:

TABLE 2. Characteristics of participants in the second focus group meeting (n=9)

Characteristics	Value*
<b>Role</b>	
Nurse (based in special school)	1 (11.1)
Social worker	2 (22.2)
Special school principal/teacher	3 (33.3)
Manager/coordinator/staff in non-governmental organisation	3 (33.3)
<b>Sex</b>	
Female	7 (77.8)
Male	2 (22.2)
<b>Age group, y</b>	
18-25	0
26-30	2 (22.2)
31-35	0
36-40	3 (33.3)
41-45	1 (11.1)
>45	3 (33.3)
<b>Education level</b>	
Primary or below	0
Secondary	0
Post-secondary/associate's degree or equivalent	2 (22.2)
Bachelor's degree	3 (33.3)
Master's/doctoral degree	4 (44.4)
<b>Experience related to rare diseases, y</b>	
<5	0
5-9	4 (44.4)
10-14	2 (22.2)
15-19	1 (11.1)
≥20	2 (22.2)
<b>Target patient type</b>	
Physical disability	6 (66.7)
Intellectual disability	9 (100.0)
Psychological/mental problems	3 (33.3)
Visual impairment	6 (66.7)
Hearing impairment	5 (55.6)
Others	1 (11.1)
<b>Target patient age group</b>	
Infants (<1 y)	1 (11.1)
Toddlers (1-2 y)	1 (11.1)
Children (3-12 y)	5 (55.6)
Adolescents (13-18 y)	6 (66.7)
Adults (>18 y)	5 (55.6)
Older adults (≥65 y)	3 (33.3)
<b>Experience completing research questionnaires</b>	
Questionnaires related to rare diseases	0
Questionnaires related to service use/resource use	0

\* Data are presented as mean±standard deviation or No. (%) of participants

TABLE 3. Agreement between the Client Service Receipt Inventory for rare genetic disease population in Hong Kong and the electronic patient record

	Intra-class correlation coefficient (95% confidence interval)		
	Overall	Self-completed (patient version)	Proxy-completed (caregiver version)
Overall service utilisation	0.69 (0.56-0.78)	0.67 (0.50-0.80)	0.70 (0.50-0.83)
Inpatient visits	0.81 (0.73-0.87)	0.81 (0.70-0.89)	0.79 (0.64-0.89)
Outpatient visits	0.60 (0.45-0.71)	0.46 (0.22-0.65)	0.67 (0.46-0.81)
Accident and emergency visits	0.58 (0.42-0.70)	0.66 (0.48-0.79)	0.33 (0.03-0.58)

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