

# The Socioeconomic Impact of Retinitis Pigmentosa (RP) in Japan

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## OBJECTIVES

- Inherited retinal dystrophies (IRDs) are a group of genetic diseases that can lead to visual impairment (VI) or blindness.<sup>1</sup> Retinitis pigmentosa (RP) is the most common form of IRD.
- While studies on the economic costs of VI and blindness for any reason exist, the specific broader societal economic burden of RP-induced VI has been largely overlooked.
- In this study, we attempted to assess the socioeconomic impact of RP in Japanese patients.

## METHODS

- Initial data collection was carried out in an online/phone survey in the autumn of 2021. The survey included 37 caregivers and 118 patients with RP (**Table 1**). Patients and caregivers were recruited via the Japanese Retinitis Pigmentosa Society (JRPS). A telephone interview was provided as an option for patients who had difficulty completing a web-based survey.
- The visual impairment of patients was graded according to the visual impairment class (AMA).<sup>2</sup>
- A post-hoc analysis was conducted to identify the socioeconomic burden of VI and blindness in RP patients, their caregivers, and other health system stakeholders.

**Table 1. Methods of interview stratified by VI severity<sup>2</sup>**

Method of interview	Near-normal vision	Moderate low vision	Severe low vision	Profound low vision	Near-blindness	Total blindness	Total
Web	5	10	18	21	32	11	97
Telephone	-	-	3	2	11	5	21
Total	5	10	21	23	43	16	118

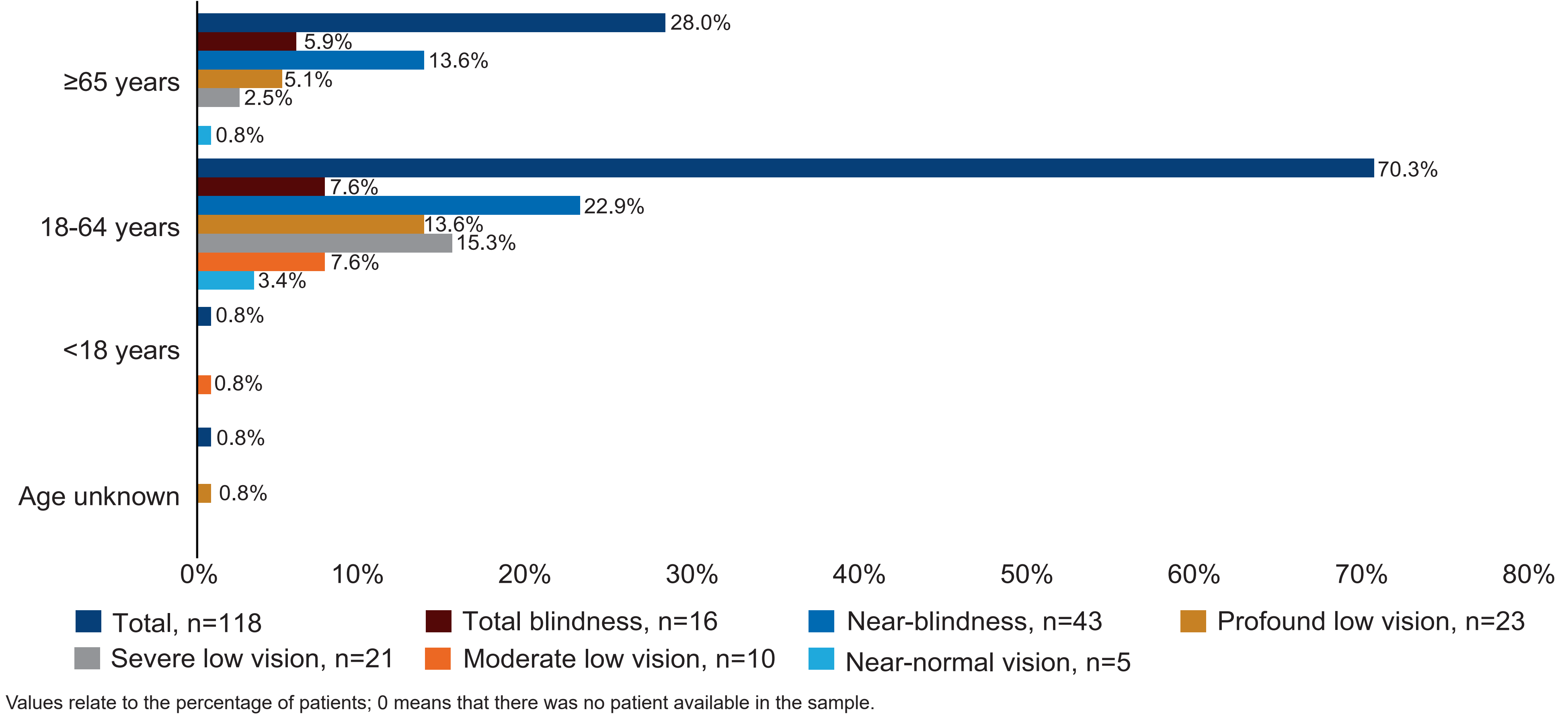
- Our model quantified the relationship between productivity losses, governmental spending, associated deadweight losses and total societal costs. This was based on the approach used by Nakamura et.al. paper<sup>3</sup>, while incorporating spending from further stakeholders.
- The approach accounted for net transfers of funds between stakeholders, while excluding potentially duplicative costs (for example, productivity losses and tax losses were not both included in societal costs).
- The societal costs were estimated by the following formula: (relevant productivity costs) + (deadweight loss rate [DWLR] \* social benefits) + (a separate DWLR for other governmental spending \* other governmental costs) + (net patient level costs) + (employment rate of patients \* cost to employer) + (rate of patients with informal carers \* cost to carer) + (donations from friends and family).
- Results were presented by each perspective (e.g., patient-level, government-level, society-level), by different levels of VI severity and by age group.

## RESULTS

### Patient distribution

- Out of the 118 patients surveyed, 13.56% were totally blind and a further 55.93% had profound vision loss or could only count fingers (**Figure 1**).
- Many of the surveyed patients were from the 18 to 64 years age group (70.34%) (**Figure 1**).

**Figure 1. Distribution of N=118 patients by age groups and level of visual impairment**



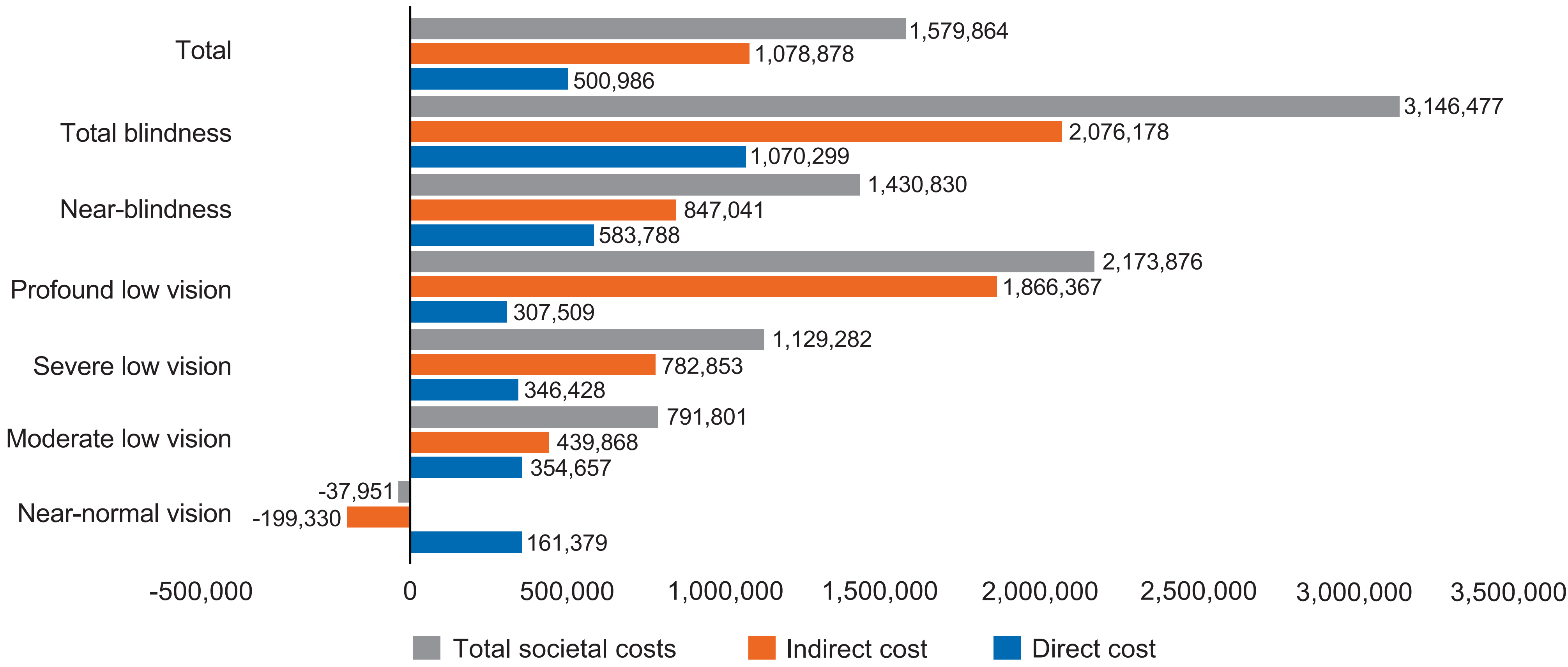
Values relate to the percentage of patients; 0 means that there was no patient available in the sample.

### Annual costs

- The average annual societal cost of RP patients was 1,579,864 Japanese Yen (~15,440 USD at 2021 PPP exchange rates<sup>4</sup>) (**Figure 2**).
- This is the sum of direct costs (medical and social welfare costs) of 500,986 yen and indirect costs (reduced productivity of patients and opportunity costs for caregivers) of 1,078,878 yen (**Figure 2**).
- Total societal costs generally rose with increasing severity of VI, as did productivity costs. For blind patients, the annual cost to society was 3.15m yen, whereas for patients with near-normal vision the total cost was actually slightly negative due to apparent productivity gains (**Figure 2**).

- Overall, the average annual productivity loss for all patients was 1,087,046 yen.
- The average annual social welfare cost burden for RP patients was 1,066,013 yen, which was relatively flat across severity groups.

**Figure 2. Annual societal costs of RP in JPY**



0 means that the patient reported these as zero costs or very minimal costs; negative values denotes societal benefit; otherwise societal costs are shown.

### Lifetime costs

- Assuming age of onset of 11.2 years and a life expectancy of 84.77 years, the overall lifetime average cost before discounting was calculated at 116,230,564 yen (~1,135,963 USD at 2021 PPP exchange rates<sup>4</sup>) for each RP patient, of which the direct cost was 36,857,546 yen (~360,222 USD<sup>4</sup>) (**Table 2**).
- Using a discount rate set at 2% based on the guidelines of C2H, the Japanese HTA agency<sup>5</sup>, the discounted average lifetime cost was calculated to be 61,279,075 yen (598,902 USD<sup>4</sup>) for each RP patient, of which the direct cost was 19,432,035 yen (~189,916 USD<sup>4</sup>) (**Table 2**).
- Average productivity and total societal costs were (as expected) highest in the working-age group.
- According to medical experts, genetic testing is required only once in a lifetime. The cost of genetic testing was therefore included in the lifetime costs once, and was excluded from the annual costs.

**Table 2. Lifetime undiscounted and (discounted) costs for all patients group in USD**

	Near-normal vision	Moderate low vision	Severe low vision	Profound low vision	Near-blindness	Total blindness	Total
Productivity	-143,323 (-75,563)	498,122 (262,620)	565,295 (298,035)	1,333,117 (702,846)	727,695 (383,655)	884,576 (466,366)	781,614 (412,083)
Government	166,029 (87,888)	843,299 (444,958)	0 (505703)	1,046,098 (552,370)	1,266,785 (668,533)	1,496,622 (790,155)	1,112,767 (587,408)
Patient	-317,304 (-167,282)	-689,373 (-363445)	-756,121 (-398,628)	-725,975 (-382,733)	-854,703 (-450,605)	-741,356 (-390,838)	-766,463 (-404,081)
Employer	0	0	15,408 (8,123)	10,785 (5,686)	2,301 (1,213)	35,951 (18,954)	10,880 (5,736)
Caregiver	0	169,608 (89,421)	425,966 (224,578)	413,622 (218,070)	252,563 (133,156)	1,053,472 (555,412)	404,496 (213,258)
Friends/family	51,770 (27,294)	0	41,087 (21,662)	3,751 (1,978)	0	0	10,237 (5,397)
Healthcare	85,837 (45,255)	175,467 (92,510)	172,227 (90,802)	141,419 (74,559)	322,625 (170,095)	683,645 (360,431)	276,052 (145,540)
Total Societal costs	-27,288 (-14,387)	569,325 (300,160)	811,983 (428,094)	1,563,073 (824,083)	1,028,803 (542,406)	2,262,398 (1,192,782)	1,135,963 (598,902)
Direct costs	116,036 (61,176)	255,007 (134,445)	249,091 (131,326)	221,107 (116,572)	419,759 (221,305)	769,572 (405,734)	360,222 (189,916)
Indirect costs	-143,323 (-75,563)	316,277 (166,747)	562,892 (296,768)	1,341,966 (707,511)	609,045 (321,100)	1,492,826 (787,048)	775,741 (408,986)

Values are undiscounted (discounted) costs in USD<sup>4</sup>; 0 means that the patient reported these as zero costs; negative values denote societal benefit; Gene testing costs were assumed to take place in year 1.

### Burden of costs in age group of 18 to 64 years (at all VI levels) – annual costs

- The details of the cost burden in the 18-64 age group are presented in **Table 3**, by way of example of the granular detail used to calculate the overall figures. Similar calculations were also carried out for other age group-VI severity combinations.
- The number of patients in this age group was 83.
- The proportion of working was 53%, and the depression rate in this group was 11%.
- The proportion of receiving informal care was 67%, and the proportion of receiving paid care was 46% in this age group.
- Social welfare payments of 1,157,576 yen from the government were balanced by the fact that patients received the same amount (plus a small contribution on average from friends and family). While this has a net zero impact, a further deadweight loss of 115,757 (10%) on a societal level was assumed.

- All perspectives (except for patients) had net financial costs associated with them. Patients' social welfare payments more than outweighed their other costs (ignoring opportunity costs represented by productivity losses, which were measured separately).
- Total societal costs do not equal the sum of costs from each perspective due to (1) removal of duplicative costs and (2) the need to weight perspectives (e.g., the contribution of employer costs to societal costs is a combination of the average cost to an employer multiplied by the employment rate).
- The highest costs were associated with government and productivity, and to a lesser extent to caregivers. Other perspectives had a relatively small impact on total societal costs.

**Table 3. Cost burden in the 18-64 years age group in JPY**

		Productivity (in JPY)	Other (in JPY)					Total societal cost (TSC) (in JPY)	
			Govt	Patient	Employer	Caregiver	Friends/family	Healthcare	
Visual health	Visual aids + supports		11,183	28,896	8,947			49,026	49,026
	Medical expenses			15,276				15,276	15,276
	Medication			13,052				13,052	13,052
Depression	Direct medical costs		10,534	1,170				11,704	11,704
	Depression related suicide costs	18,941						18,941	18,941
Other health costs	Paid caregivers		120,384	11,518				131,902	131,902
	RP research		67,795					67,795	67,795
	Gene testing (once off)		170,735	3,120				173,855	173,855
	Social benefits		1,157,576	-1,177,817			20,241	NA	115,758
Other patient level costs	Productivity loss	893,807						NA	893,807
	Tax loss		100,035					NA	0
Informal care	Opportunity costs					369,250		NA	369,250
	Social benefits		26,259			-26,259	0	NA	2,626
	Productivity losses	398,127						NA	0
	Tax loss		28,289					NA	0
Total costs from this perspective		1,310,875	1,522,055	-1,107,904	16,877	508,363	20,241	307,697	NA
Contribution to total costs from society		912,748	1,393,731	-1,107,904	8,947	342,992	20,241	0	1,689,138
Direct cost		18,941	1,393,731	-1,107,904	8,947	-26,259	20,241		426,081
Indirect cost		893,807	0	0	0	342,992	0		1,263,057

0 means that the patient reported these as zero costs; negative values denote societal benefit.

### Limitations of the study

- Participants in this study were identified in collaboration with JRPS and may be biased toward those with high involvement in patient support groups or high use of health care services. Therefore, they may not be representative of the entire RP patient population, and this selection bias should be noted.
- In this study, there may have been differences in question comprehension and response accuracy between online survey respondents and telephone interview respondents. In addition, there may be a higher frequency of misunderstandings and typing errors in online surveys of visually impaired respondents than in general online surveys.
  - Indeed, one patient's salary was excluded from calculations of average productivity due to a presumed typo. No other data were censored or removed.
- Additionally, this analysis assumed an age of onset of 11.2 years, and the average life expectancy of 84.77 years, which might not reflect the diverse heterogeneity in the phenotypes of RP.

## CONCLUSIONS

- We have calculated the costs of RP-associated VI from the perspectives of various stakeholders combined them to calculate total societal costs. This has been further carried out by age group and VI severity level.
- RP is associated with a significant financial burden from a societal perspective in Japan. Costs rise with severity level.
- In the overall population, indirect costs were about twice as high as direct costs and are primarily related to productivity losses and impact on opportunity costs.
- Additional research is needed to gain a holistic understanding of the burden of disease from the perspective of the health care system and society.

## Abbreviations

AMA, American Medical Association; DWLR, deadweight loss rate; IRDs, inherited retinal dystrophies; JPY, Japanese yen; JRPS, Japanese Retinitis Pigmentosa Society; n, number of patients; LCA, Leber Congenital Amaurosis; N, total number of patients; PPP, purchasing power parity; RP, retinitis pigmentosa; USD, United State Dollar; VI, visual impairment.

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