Author’s response to reviews

Title: Effects of financial support on treatment of adolescents with growth hormone deficiency: a retrospective study in Japan

Authors:

Eri Maeda (erimaeda@med.akita-u.ac.jp)
Takahiro Higashi (thigashi@ncc.go.jp)
Tomonobu Hasegawa (thaseg@a6.keio.jp)
Susumu Yokoya (yokoya-s@ncchd.go.jp)
Takahiro Mochizuki (m4601256@yahoo.co.jp)
Tomohiro Ishii (tomishii@mac.com)
Junko Ito (jun-ito@toranomon.gr.jp)
Susumu Kanzaki (smkanzak@med.tottori-u.ac.jp)
Akira Shimatsu (ashimats@kyotolan.hosp.go.jp)
Koji Takano (ktakano-ty@umin.ac.jp)
Toshihiro Tajima (tajeari@med.hokudai.ac.jp)
Hiroyuki Tanaka (hrtanaka@saiseidr.jp)
Yusuke Tanahashi (yutanaha@asahikawa-med.ac.jp)
Akira Teramoto (a-tera@nms.ac.jp)
Toshiro Nagai (toshironagai@gmail.com)
Kunihiko Hanew (hanew-endo-clinic@juno.ocn.ne.jp)
Reiko Horikawa (horikawa-r@ncchd.go.jp)
Toru Yorifuji (t-yorifuji@osakacity-hp.or.jp)
Naohiro Wada (naohiro-wada@i.shizuoka-pho.jp)
Toshiaki Tanaka (toshi_tnk@tanaka-growth-clinic.com)
Author’s response to reviews:

September 20, 2016

Dear Dr. Petra Baji,

Thank you for reviewing our paper, “Effects of financial support on treatment of adolescents with growth hormone deficiency: a retrospective study in Japan (BHSR-D-16-00461).” We have revised the paper in line with the suggestions by the reviewers. Please see our replies below. We marked the changes in red in the manuscript.

Reply to reviewer comments:

Reviewer #1:

1. In the background section, it would be helpful to include a lit review of research on the topic of financing mechanisms for rare disease treatment. It would be useful to refer to the lit review to better contextualized the current study.

Insurance has generally had positive impacts on the use of pediatric specialty care in the United States, but little is known about the association between out-of-pocket payment and pediatric general or specialty care in Japan. Following the reviewer’s advice, we have included this point in the background section (page 5, lines 33–39).

2. In the same section, it would be helpful to explain what new insight from an economic theoretical point of view would the study add to current discourse.
In Japan, where 70% of medical charges are covered by universal health insurance, little attention has been paid by research to the geographic variation in subsidy for the remaining 30% out-of-pocket payment. This study was conducted to reveal possible effects of geographical inequality in financial support for specialty care in children. We showed this point clearly as a secondary aim (page 5, lines 43–46), which corresponds to the fourth paragraph in the Discussion (page 11, lines 9–44).

3. in the first paragraph of the result section, it may help to explain what percent of patients are not eligible for subsidy. I suspect readers would appreciate it.

We have explained that 15.5% of the children were eligible for financial support after they reached the MAPChD height limits (page 8, lines 48–53), and the rest (84.5%) were not eligible.

4. it would be interesting to explain whether the geographic variation in subsidy showed a dose-response with regards to treatment duration. in other words, more co-pay, fewer sustained treatments?

We performed the additional analysis according to the reviewer’s advice, but we did not find any dose-response associations. Although it is a conjecture, we think the lack of association is due to unbalanced data; the percentage of children who continued treatment for more than two years was only 12% (47/388).

5. in the discussion section, readers would be interested how this study's result compares with other rare diseases and subsidy policy options.

7. Are there other rare diseases with subsidy in Japan the authors could compare with the current study?

Currently, the Medical Aid Program for Chronic Pediatric Diseases of Specified Categories (MAPChD) supports out-of-pocket payment to children with any of 704 chronic diseases. MAPChD defines subsidy criteria for some of the diseases, but there has been no report of the relationship between financial subsidy and access to care for the other diseases. This study is the first to describe the association, and it provides an important reference for the other chronic
diseases. For more common diseases, there is one report of survey results about parental attitudes toward care seeking for children with a severe cold. Since the authors found no difference in care-seeking attitudes regardless the presence of subsidy, caution should be taken in generalizing our results to the other diseases. We have added this point in the Discussion (page 11, lines 28–44).

5. What is the influence of co-pay and/or subsidy on rare disease treatment versus common diseases such as asthma, which was discussed in the section? Are parents having children with rare diseases more compelled to seek treatment compared to a common disease?

We could not find a description about the rareness of the diseases and treatment utilization in relation to financial support, but severity of colds might weaken the effect of financial support on attitude toward health care. We have added this point in the Discussion (page 11, lines 33–44).

6. How similar is the healthcare finance environment compared to other countries to allow the study's finding generalizable?

Although countries have different health financing systems, many developed countries subsidized most of the treatment costs for GHD through public health insurance or reimbursement by the governments (page 4, lines 9–14). The applicability of the findings to each country must be judged by the policy-makers responsible in that country. We have added this point in the Discussion (page 10, lines 9–15).

Reviewer #2: This is an interesting and well written article. As gender differences on a hormonal level are known (Pituitary, December 2002, Volume 5, Issue 4, pp 247-253) the authors should cite and discuss this aspect in more details. Otherwise the manuscript is ready to be accepted.

Based on the reviewer’s advice, we have added further discussion in view of the gender difference in clinical features and cited this publication (page 10, lines 16–22).
We believe that we have suitably responded to each comment and that paper has consequently been strengthened. In addition, a professional language editor checked and edited our manuscript as appropriate. If we can make further changes, please let me know.

Thank you for the opportunity to revise our paper.

Sincerely yours,

Eri Maeda, M.D., Ph.D.
Takahiro Higashi, M.D., Ph.D.