A proposed approach to estimating excess mortality associated with cleft lip / cleft palate

This note describes a simple and cheap method that may be useful for the estimating excess mortality associated with cleft lip / cleft palate in settings where reporting of congenital anomalies is incomplete or absent. It is intended as a request for comments.

The method uses:

> Readily available survey data (e.g. census tables, DHS, MICS) or simple survey data that can describe the age-distribution of the general population.
>
> Data on the ages of found cases from clinical settings (may suffer a selection bias) and from community-based case-finding exercises (e.g. using active and adaptive case-finding).

The method is designed to be very low cost (i.e. compared to a cohort study) and to increase the impact of programs treating cleft lip / cleft palate through the introduction or expansion of community-based case-finding and referral.

Mortality in the general population is estimated from the population age distribution using a simple exponential decay model in which the proportion of the birth cohort surviving at each age follows:

$$p = e^{-zt}$$

where e is the base of the natural logarithm (approximately 2.7183), z is the mortality rate, and t is age. Given an age-distribution (e.g. as shown in Figure 1) we can fit the model:

$$\log_e(n) = \alpha + \beta \times t$$

where n is the count of children in each age group. The absolute value of β is an estimate of the mortality rate (z) in the general population. For the data shown in *Figure 1* this is $z = 0.028205$ which is equivalent to about 0.77 / 10,000 / day. The expected counts for the model:

$$p = e^{-0.028205 \times t}$$

are shown in red in *Figure 1*. We can fit a similar model to the ages of cases of cleft lip / cleft palate found in clinical data and by community-based case-finding. Given the age distribution of cases shown in *Figure 2* we estimate z to be 0.36690 which is equivalent to about 10 / 10,000 / day.

Excess mortality can be estimated as the ratio of the two mortality rates:

$$\textit{Mortality rate ratio} = \frac{z_{Cases}}{z_{General\ population}}$$

with the example data this is:

$$\textit{Mortality rate ratio} = \frac{0.36690}{0.028205} = 13.01$$

Mortality in cases is about thirteen times higher than the general population.

A 95% confidence interval could be calculated using the bootstrap.

It is envisaged that data for children and cases aged between 6 months and 5 years (the simple exponential decay model may not hold with older children and adults) will be used and that age will be recorded to best possible accuracy and precision.

Figure 1 : Example age distribution (0 to 5 years) in the general population

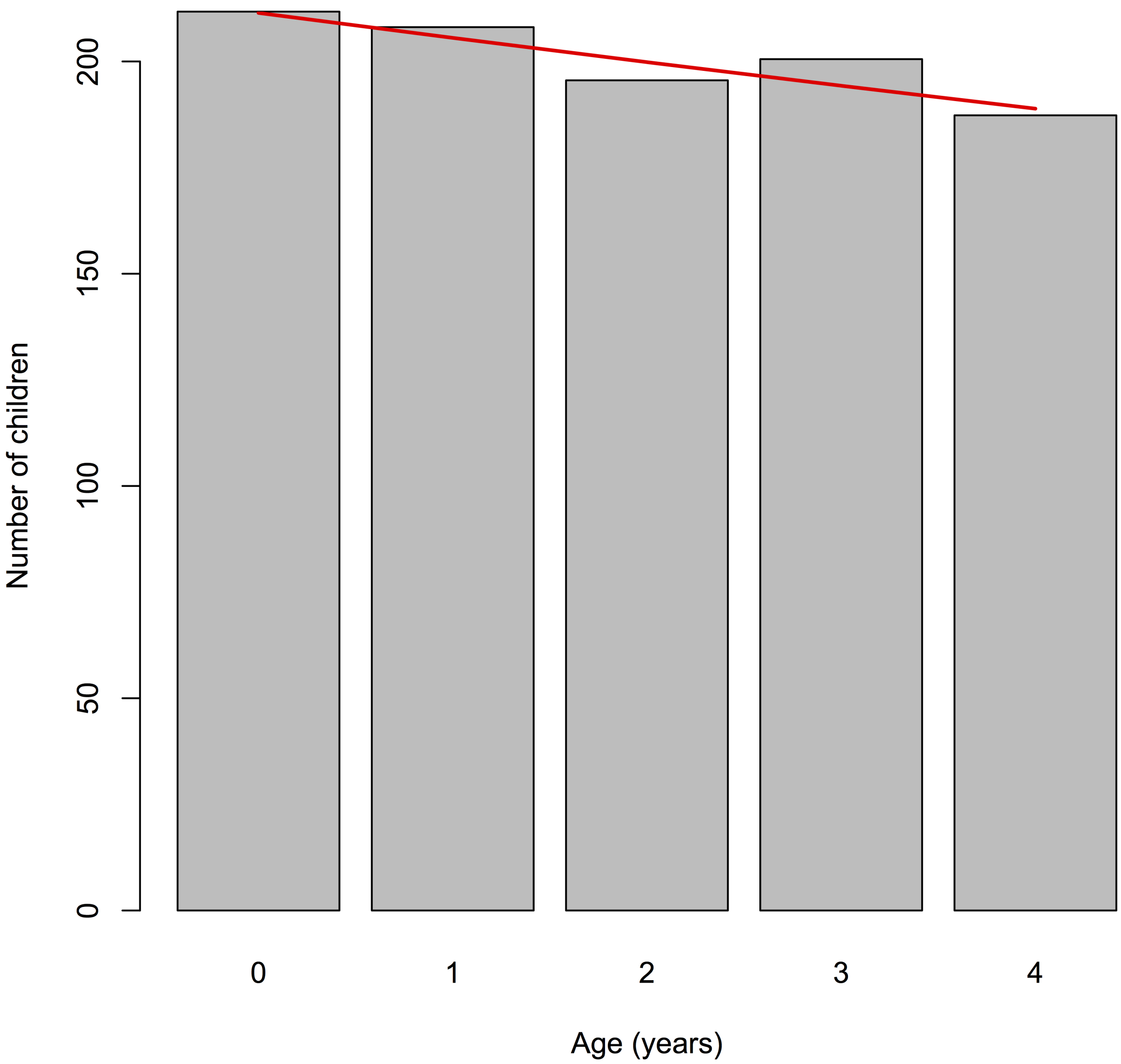

Figure 2 : Example age distribution (0 to 5 years) in cases