Letters to the Editor
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Cost effectiveness of neonatal ECG screening for the long QT syndrome

We read with interest the recent article by Quaglini et al.,1 in the European Heart Journal, concerning cost efficacy of proposed neonatal ECG screening for long QT syndrome. We all agree that undiagnosed cases of LQTS likely play a role of as yet undetermined magnitude in sudden death in very young children, including sudden infant death syndrome. A similar study has been previously published.2 We have some concerns, however, which we would like to voice.

We are sceptical concerning the issue of costs related to the process of establishing a correct diagnosis. The cost of a cardiologist visit is likely underestimated, and the contention that there are no costs associated with training adult cardiologists to read neonatal ECGs is unrealistic. We are most concerned, however, about the very real implications of making an incorrect diagnosis leading to inappropriate treatment.

Neonatal screening has been an area of active discussion and health policy deliberation in the paediatric community for decades. Unfortunately, as a screening test for LQTS, the ECG is disappointing when compared with other accepted neonatal screening tests. For example, congenital hypothyroidism (CH) has a similar incidence to LQTS (1:1800) and neonatal screening has a sensitivity of ~95%, a specificity of 99.9%, and a positive predictive value of 29%. In a recent review of 430 764 infants screened from the Netherlands,3 this amounted to 772 positives of which 224 were true CH patients. In this case, however, evaluating infants with positive screening tests to rule out the disease is fairly simple—a repeat blood test.

For LQTS, on the other hand, there is no comparable, simple, and definitive follow-up test to rule out the disease in an asymptomatic individual with an initially prolonged QT. Genetic testing is far too expensive for wide application and is certainly not comprehensible enough to allow one to rule out the diagnosis of LQTS. Paediatric cardiologists and electrophysiologists face the problem of asymptomatic QT prolongation regularly, and the process is typically time consuming and anxiety provoking for the physicians and families concerned, as it involves a careful study of family history, ECGs obtained from parents and other relatives, and often other testing. Accepting the authors’ figures of an incidence of 1/2500, a positive rate of 1% for a QTc cutoff of 470 ms, and a sensitivity of 80% yields a positive predictive value of only 3%. In the USA, with a birth rate of 4 million/year, one would predict a cohort of 1280 LQTS patients hiding among 40 000 infants with prolonged QT intervals, each and every year. It is not clear that the resources exist to manage such an onslaught of patients in whom the possibility of a life-threatening illness has been raised. Inevitably, a significant number of normal infants would be treated for long QT syndrome, and such treatment involves more than just beta-blocker therapy with its potential side effects. One can expect a lifelong restriction from competitive physical activity, difficulties obtaining life insurance, avoidance of a host of medications, and the potential for more invasive therapies such as ICD implantation. These costs may be difficult to quantify but are significant and not addressed in the article by Quaglini et al. Most distressingly, the psychological and emotional impact on the family of a child incorrectly labelled as having LQTS is ignored in the authors’ analysis. We support the focused use of ECGs in subjects for whom the prior probability of arrhythmic risk is elevated (such as those with a positive or suggestive family history), but we cannot support a recommendation for mass screening in the general population. Currently, at least half of LQTS patients are diagnosed by familial screening, and another large fraction of patients are diagnosed by ECG after an initial seizure or faint. We would be very interested in a recalculation of the cost efficacy of ECG screening in which the realistic costs of initial screening are included, as well as the downstream costs associated with investigating LQTS in all false positives and those costs associated with unnecessary lifelong treatment. A comparison of the two approaches, mass screening vs. focused screening, could then be assessed.

References

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