Objective: Early childhood psychosocial experiences determine future health and health-care use. Identifying psychosocial predictors in cystic fibrosis may inform intervention strategies that can reduce health-care utilization.

Design: The study was designed as a prospective cohort study.

Setting: The study was set in the only cystic fibrosis clinic in Western Australia.

Patients: The patients were children up to 6 years diagnosed with cystic fibrosis in Western Australia between 2005 and 2011.

Main outcome measures: Psychosocial data collected for each year of life were compared with Australian population data and analysed as predictors of annual hospital, emergency and outpatient visits.

Results: Compared with the Australian population, cystic fibrosis families demonstrated lower socio-economic status and labour supply ($P < 0.001$), increased residential mobility ($P < 0.001$) and trends towards increased rates of parental separation ($P = 0.066$). Marital discord and maternal and child psychological stress significantly predicted increased hospital admissions, emergency and outpatient visits.

Conclusions: Social gradients may exist for families of young children with cystic fibrosis in Western Australia with potential implications for child health. Family psychological and relationship stress predicted increased child cystic fibrosis-related health-care use.

Key words: child; cystic fibrosis; mental health; parent; psychosocial determinant; social gradient.

What is already known on this topic

1. Little is known about psychosocial factors, other than socio-economic status (SES), in early life that may influence health-care use in young children with chronic disease and cystic fibrosis (CF).
2. There are no data describing psychosocial characteristics of families with young children (0–6 years) diagnosed with CF in Australia.
3. Chronic disease in childhood may beget socio-economic disadvantage through loss of social and economic assets, but whether a social gradient exits in early life in CF in Australia is not known.

What this paper adds

1. These are the first Australian data that suggest psychological distress in the family, and not SES per se, predicts increased hospitalisation and emergency visits in early life in CF.
2. A social gradient may exist for families of young children with CF compared with the general population.
3. CF families have increased residential mobility and reduced family labour supply, which may have important health implications for young children.

Psychosocial experiences in early childhood are important determinants of future health and well-being, and may significantly alter the clinical trajectory of children with chronic disease through numerous bio-psychosocial interactions. The role of psychosocial factors in the environment of young children as moderators of health-care use, and health outcomes are widely recognised, but poorly understood in cystic fibrosis (CF).

CF is a life-shortening, multisystem disorder with complex daily treatment regimens, need for frequent surveillance by multidisciplinary teams and high use of hospital resources.

Data linking socio-economic circumstances to health and chronic disease exists, and large registry-based cohort studies in CF demonstrate increased mortality, worse pulmonary and nutritional outcomes and increased health resource utilisation.
among patients with lower socio-economic status (SES). Conversely, the impact of chronic disease on psychosocial and SES of individuals or families can be considerable, with loss of financial and social assets that may adversely influence health-care use and health behaviour, amplifying risks of morbidity. While the importance of the psychosocial environment in early life on the future health of young children is acknowledged, whether a social gradient exists for CF in early life warrants further study.

This study aimed to examine psychosocial characteristics of families of young children with CF in Australia and to identify factors associated with increased health-care use during the first 6 years of life.

Methods

This is a prospective cohort study of all children with CF, to age six years, diagnosed by newborn screening receiving care at the paediatric CF centre for Western Australia (WA) between 2005 and 2011. Children included were undergoing early surveillance with the Australian Respiratory Early Surveillance Team for CF (AREST CF), over 95% of the total state population of children aged to 6 years with CF.

Data collection and measures

Psychosocial data were collected for each year of life to allow multiple time-point tracking of psychosocial variables, from detailed medical and nursing case notes and CF social worker records (diagnosis evaluation, annual review data, regular clinic review notes). Included were patient demographics; family structure, documented parent relationship status; housing and transport; socioeconomic data and caregiver mental health. Health-care utilisation data were obtained from hospital clinical management systems, hospital medical records, and social worker records.

Measures included:
- SES classified according to the Socio-Economic Indexes for Areas (SEIFA). Subject postcodes were allocated a SEIFA decile indicating degrees of comparative advantage/disadvantage: <5th decile (most disadvantaged), 5–9th decile, 10th decile (least disadvantaged).
- Index of Relative Socio-economic Disadvantage encompasses low income, unemployment and low levels of education.
- Index of Relative Socio-economic Advantage/disadvantage includes indicators of advantage.
- Index of Economic Resources, and index of Education and Occupation, encompasses income and housing and education and occupation, respectively.
- Parental occupation was classified using the Australian and New Zealand Standard Classification of Occupations.
- Reported parental mental illness was classified using the International Statistical Classification of Diseases and Related Health Problems 10th Revision.

The data were compared with published data from The Australian Bureau of Statistics and The Longitudinal Study of Australian Children (‘B cohort’ data collection age 0–1 in 2004, age 2–3 in 2006 and age 4–5 in 2008). Ethics approval was obtained from the Princess Margaret Hospital for Children Ethics Committee.

Statistical analysis

Binomial test for proportions was used to compare SEIFA indices with corresponding WA averages. Data were compared separately for three different categories of SEIFA index and were stratified by child’s age group. Parents’ home ownership levels were compared with national averages and stratified by child’s age group.

To examine associations between social, economic and psychological variables and health resource utilisation, hospital admission data were modelled using general estimating equations with Poisson distribution and robust error variance with repeated measurements allowed for each child. Associations of these predictors with outcomes were measured using multivariate models adjusted for body mass index (BMI) z-score (obtained at annual review visit), pancreatic status and presence of bronchiectasis to control for disease severity. Because of low sample size, data were analysed for all years combined.

Outpatient visits were grouped into presence or absence of outpatient visits/year, <4 or ≥4 CF outpatient visits/year, presence or absence of IV antibiotic days/year and presence or absence of CF-related emergency department visits/year. The cut-off of <4 or ≥4 CF outpatient visits/year reflected current benchmark standards of CF care that recommend quarterly outpatient visits for optimal surveillance. All analyses were performed using Stata 13.0 (Stata Corp., College Station, TX, USA) with P-values of <0.05 considered significant.

Results

Seventy-five children (51% male) were included (demographic characteristics in Table 1). The number with data from diagnosis available for analysis at year of life time-points 0–1, 2–3 and 4–5 were n = 75, n = 55, n = 33 respectively.

Geographical and residential mobility

The distribution of children and families paralleled Australian population data, with 69% living in metropolitan and 31% in regional areas (Table 1). There was no statistically significant
residential shift of the cohort towards metropolitan residence with increasing subject age ($P = 0.23$). Residential mobility was greater than the Australian preschool population. More study children moved residence at least once during infancy and preschool years than the general preschool population ($P = 0.001$). Compared with the Australian population, children with CF had trends towards significantly increased odds of moving house in the first year of life (odds ratio (OR): 1.7 ($95\%$ confidence interval 0.97–2.97), $P = 0.061$), and three times the odds of moving house between ages 2–5 years (OR: 3.2 (1.87–5.5), $P < 0.001$).

**Socio-economic status**

SEIFA index data were compared with the WA general population at three time periods (aged 0–1 years, 2–3 years, 4–5 years). $^{24}$ Compared with the WA population a significantly greater proportion of CF families were grouped into the lowest deciles, and a significantly smaller proportion of CF families were grouped into the highest decile indices of Education and Occupation, Economic Resources, and Relative Socio-economic Disadvantage (Table 2). These observations were consistent across each year-of-life time points.

**Caregiver occupation and family labour supply**

There were no significant differences in parental occupation classification when compared with the published Australian data ($P$ values $> 0.5$). $^{21}$ Two-parent families with CF had a significantly reduced labour supply in their child’s first year of life ($P < 0.001$) and at 2–3 years ($P = 0.041$). $^{21}$ More mothers of CF children were unemployed $^{21}$ during ages 0–1 (94.7% unemployed vs. 64.2%, $P < 0.001$) and 2–3 years (56.4% vs. 51.6%, $P = 0.042$). Less than one-third of previously employed mothers (29.5%) returned to work part-time following end of maternity leave, only one returned to fulltime employment. At child age 4–5 years, a smaller proportion of mothers were working (part-time or full-time) compared with the general population (39.4% vs. 55.1%, $P = 0.055$). The proportion of two-parent families experiencing unemployment was equivalent to the Australian rate of 6.7%; 6.25% at age 0–1, 5.41% at age 2–3 and 5% at 4–5 years, respectively (all $P$ values $> 0.5$). Fathers were the main income earners for two-parent families. Paternal fulltime employment did not differ significantly from general population data. $^{22}$ 86.7% child age 0–1 year ($P = 0.09$), 72.7% child age 4–5 years ($P = 0.66$).

**Homeownership and transport**

Complete data on homeownership were available in 81%, 75% and 64% of families at year-of-life time points 0–1, 2–3 and 4–5 years respectively. Homeownership was lower (not significant ($P = 0.24$) compared with national levels (Table 3) of around 70%, $^{13}$ The distribution of families across the accommodation categories did not differ statistically from Australian data (Table 3). Car ownership in the first 3 years of life was more common among study families than in the Australian population ($P = 0.002$).

**Family structure and relationships**

The biological mother was the primary caregiver in all families. Intact parental relationships were observed in 90% of families during the child’s first year of life, falling to 62.5% by the sixth year. The frequency of two biological parents in the home among CF families was lower than the general population, and

<table>
<thead>
<tr>
<th>Table 2</th>
<th>Western Australia (WA) Socio-Economic Indexes for Areas (SEIFA) index deciles by child age</th>
</tr>
</thead>
<tbody>
<tr>
<td>SEIFA Index of Education and Occupation</td>
<td>&lt;5th decile</td>
</tr>
<tr>
<td>Time point (age years)</td>
<td></td>
</tr>
<tr>
<td>0–1</td>
<td>24 (32.4)</td>
</tr>
<tr>
<td>2–3</td>
<td>21 (38.2)</td>
</tr>
<tr>
<td>4–5</td>
<td>11 (33.3)</td>
</tr>
<tr>
<td>WA State</td>
<td>6.3%</td>
</tr>
<tr>
<td>SEIFA Index of Economic Resources</td>
<td></td>
</tr>
<tr>
<td>Time point (age years)</td>
<td></td>
</tr>
<tr>
<td>0–1</td>
<td>10 (13.5)</td>
</tr>
<tr>
<td>2–3</td>
<td>12 (21.8)</td>
</tr>
<tr>
<td>4–5</td>
<td>9 (27.3)</td>
</tr>
<tr>
<td>WA State</td>
<td>4.7%</td>
</tr>
<tr>
<td>SEIFA Index of Disadvantage</td>
<td></td>
</tr>
<tr>
<td>Time point (age years)</td>
<td></td>
</tr>
<tr>
<td>0–1</td>
<td>14 (18.9)</td>
</tr>
<tr>
<td>2–3</td>
<td>16 (29.1)</td>
</tr>
<tr>
<td>4–5</td>
<td>11 (33.3)</td>
</tr>
<tr>
<td>WA State</td>
<td>5.4%</td>
</tr>
</tbody>
</table>

All data presented as frequency and (%) of study families grouped into each SEIFA decile according to child age. WA State: the proportions of the general Western Australian population grouped in each decile.
The highest incidence of parental separation occurred in the second year of life (10.4%) and relationship dissatisfaction was documented in 13.3% of primary caregivers during years 0–1, 9.1% during years 2–3, 18.2% during years 4–5. Single-parent family status was reported in 13%. There was no statistically significant difference in the number of siblings at subject age 5–6 years compared with national data (P = 0.62).

**Mental health**

Data on current maternal mental health were available in 100% of subjects’ records at age 0–1 years, 95% at 2–3 years and 94% age 4–5 years. The prevalence of reported maternal mental illness in children’s medical notes was highest in the first year of life (10/75, 13.3%), was predominantly affective and stress-related disorders, and decreased with increasing child age (4/55 (7.3%) and 2/33 (6.1%) at year-of-life time points 2–3 and 4–5, respectively). The estimated prevalence of maternal post-natal depression/anxiety in Australia is 16%.

The prevalence of fathers’ mental illness in the child’s records was lower during child’s 0–1 years: 5/75 (6.7%). An active referral to Tertiary Psychological Medicine was recorded in 24/75 (32%) children at any time during the first 6 years of life, almost exclusively to address issues with the parent-child relationship and child behaviour in the first 3 years of life.

**Health-care utilisation in the first 6 years of life**

Most (83.3%) children attended a scheduled CF outpatient clinic four or more times/year. Outpatient clinic attendance was most frequent in the first year of life: mean (standard deviation, SD) 8.34 (7.9)/year, decreasing slightly with age to 5.45 (2.43) visits/year at age 5–6 years (P = 0.233). There were 53 presentations to the emergency department with CF-related issues in 28/75 (37%) subjects over the first 6 years: mean (SD) visits 7.17 (8.12)/year. Emergency visits were more frequent in 0–1 years (20/75, 27%), 63/75 (84%) children were admitted one or more times to hospital for CF-related illness (Table 4). Maternal mental illness and an active referral of the child to tertiary psychological services were associated with increased inpatient days due to a CF-related illness. Decile scores in any of the examined SEIFA indices did not predict health-care utilisation.

**Discussion**

This is the first Australian study to describe psychosocial characteristics of families of young children with CF and to identify psychosocial factors in early life associated with hospitalisation, CF outpatient, and emergency department attendance. SEIFA indices indicate families of young children with CF in WA are at greater risk of economic, education and employment disadvantage compared with the general population. This study demonstrates CF families have significantly reduced labour supplies and increased residential mobility, with trends towards increased marital breakdown: factors with implications for child health and well-being. Importantly, this study identifies marital breakdown and maternal and family psychological distress, rather than SES per se, as significant predictors of health-care use in early life in CF, after adjusting for markers of disease severity. The role of family-psychological factors as indicators and drivers of increased health-care use, together with development of family-based psychosocial intervention strategies needs further research in larger populations, and may reduce hospitalisations and emergency presentations in young children. Similar to findings in other larger studies in CF populations, SEIFA indices did not emerge as independent predictors of health-care use in young children with CF in WA when controlled for disease severity. This might be explained by universal access to public CF health care in Australia.

Reduced household labour supply in this cohort may relate to fewer mothers returning to the work force following the birth of their child with CF, perhaps reflecting the burden associated with caring for a child with CF and hampering opportunities for flexible employment and income generation. Lower family income is associated with poorer lung function and nutritional outcomes in adolescents and adults with CF. A clearer understanding of maternal employment behaviour following CF diagnosis and the implications for family wealth and child health in CF is needed. These data can inform government policies surrounding flexible employment opportunities for families of children with chronic disease.

### Table 3 Home ownership and housing characteristics

<table>
<thead>
<tr>
<th>Age (years)</th>
<th>Homeowner</th>
<th>Private rental</th>
<th>Housing authority</th>
<th>Extended family</th>
<th>Missing data</th>
<th>Total</th>
<th>P value†</th>
</tr>
</thead>
<tbody>
<tr>
<td>0–1</td>
<td>42 (56.0)</td>
<td>8 (10.7)</td>
<td>4 (5.3)</td>
<td>7 (9.0)</td>
<td>14 (18.7)</td>
<td>75 (100)</td>
<td>0.26</td>
</tr>
<tr>
<td>2–3</td>
<td>28 (50.9)</td>
<td>7 (12.7)</td>
<td>2 (3.6)</td>
<td>4 (5.3)</td>
<td>14 (25.5)</td>
<td>55 (100)</td>
<td>0.65</td>
</tr>
<tr>
<td>4–5</td>
<td>14 (42.4)</td>
<td>3 (9.1)</td>
<td>2 (6.1)</td>
<td>2 (3.0)</td>
<td>12 (36.4)</td>
<td>33 (100)</td>
<td>0.37</td>
</tr>
</tbody>
</table>

†Homeownership proportions compared with mean Australian homeownership data.

All data presented as frequency (%).
While a social gradient in the health of CF patients exists, CF is genetically inherited and therefore disease prevalence is not socially determined per se. This study suggests that the birth of a child with CF to families in WA may precipitate a move down the ‘socio-economic ladder’, whether because of diminished labour supply or other direct and indirect costs of chronic disease that hamper social opportunity and the ability to accumulate wealth. The concept of chronic disease leading to social disadvantage through reduced economic and social assets is widely recognised, yet the socio-economic impact of CF on disadvantage through reduced economic and social assets is not fully understood.

Table 4 Psychosocial predictors of annual CF-related hospitalisation, outpatient and emergency department attendance

<table>
<thead>
<tr>
<th>Psychosocial predictors</th>
<th>&gt;4 CF outpatient attendances per year</th>
<th>Emergency department visits per year</th>
<th>Hospital inpatient days per year</th>
</tr>
</thead>
<tbody>
<tr>
<td>Primary caregiver relationship status</td>
<td>Reference</td>
<td>Reference</td>
<td>Reference</td>
</tr>
<tr>
<td>Intact</td>
<td>Reference</td>
<td>Reference</td>
<td>Reference</td>
</tr>
<tr>
<td>Separated</td>
<td>1.13 (1.05, 1.22); ( P = 0.001 )</td>
<td>4.23 (1.97, 9.08); ( P &lt; 0.001 )</td>
<td>1.36 (0.94, 1.97); ( P = 0.106 )</td>
</tr>
<tr>
<td>New partner</td>
<td>1.12 (1.02, 1.24); ( P = 0.018 )</td>
<td>2.94 (1.04, 8.33); ( P = 0.041 )</td>
<td>1.46 (1.03, 2.07); ( P = 0.032 )</td>
</tr>
<tr>
<td>Single parent</td>
<td>1.26 (1.10, 1.44); ( P = 0.001 )</td>
<td>4.08 (1.69, 9.87); ( P = 0.002 )</td>
<td>0.67 (0.29, 1.53); ( P = 0.338 )</td>
</tr>
<tr>
<td>Self-reported relationship dissatisfaction</td>
<td>1.16 (1.06, 1.27); ( P = 0.001 )</td>
<td>0.35 (0.16, 0.73); ( P = 0.006 )</td>
<td>1.40 (1.01, 1.94); ( P = 0.046 )</td>
</tr>
<tr>
<td>Reported maternal mental illness</td>
<td>1.05 (0.90, 1.22); ( P = 0.564 )</td>
<td>1.02 (0.40, 2.59); ( P = 0.970 )</td>
<td>1.78 (1.28, 2.47); ( P = 0.001 )</td>
</tr>
<tr>
<td>Active referral to paediatric psychological services</td>
<td>1.01 (0.99, 1.04); ( P = 0.287 )</td>
<td>1.33 (0.44, 4.08); ( P = 0.612 )</td>
<td>1.64 (1.22, 2.21); ( P = 0.001 )</td>
</tr>
<tr>
<td>IndRelSEAdDis</td>
<td>0.99 (0.94, 1.04); ( P = 0.617 )</td>
<td>1.09 (0.88, 1.35); ( P = 0.434 )</td>
<td>0.97 (0.88, 1.07); ( P = 0.524 )</td>
</tr>
<tr>
<td>IndRelSEDIs</td>
<td>1.02 (0.99, 1.05); ( P = 0.172 )</td>
<td>1.07 (0.91, 1.27); ( P = 0.414 )</td>
<td>0.97 (0.90, 1.05); ( P = 0.492 )</td>
</tr>
<tr>
<td>IndEconResource</td>
<td>1.01 (0.99, 1.04); ( P = 0.240 )</td>
<td>1.17 (0.99, 1.39); ( P = 0.060 )</td>
<td>0.98 (0.90, 1.06); ( P = 0.618 )</td>
</tr>
<tr>
<td>IndEdOcc</td>
<td>1.02 (0.99, 1.04); ( P = 0.147 )</td>
<td>1.03 (0.88, 1.21); ( P = 0.724 )</td>
<td>0.99 (0.92, 1.06); ( P = 0.742 )</td>
</tr>
</tbody>
</table>

All data presented as risk ratio (95% confidence interval); \( P \) value and adjusted for markers of cystic fibrosis (CF) severity. IndRelSEAdDis, Index of Relative Socioeconomic Advantage and Disadvantage; IndRelSEDIs, Index of Relative Socioeconomic Disadvantage; IndEconResource, index of Economic Resources; IndEdOcc, index of Education and Occupation.

Conclusion

Families of young children with CF in WA are socio-economically disadvantaged, are more residentially mobile and may experience significant marital and family-psychological stress. These exposures may have important implications for the future health and wellbeing of the child with CF. Psychological and relationship distress within a family is a predictor of health care use. This study cannot comment on causality and while it is plausible that parental psychological distress may precipitate ill health and hospitalisation in children with CF, it is equally possible that increased health-care use resulted from heightened clinical surveillance and intervention in response to parental and family distress.

A limitation is the relatively small sample size and potential for significant differences and associations to be missed. Reliance on documentation in notes rather than specific measures designed to capture psychosocial factors within the environment of the young child were limitations. Health-care use was restricted to hospitalisation, emergency and outpatient clinic attendance and did not capture community health-care use. Results may be biased towards families under stress where documentation of psychosocial data is more comprehensive. Further research is warranted to determine the findings’ generalizability to other CF populations and to other populations of families of children with chronic disease.

While a social gradient in the health of CF patients exists, CF is genetically inherited and therefore disease prevalence is not socially determined per se. This study suggests that the birth of a child with CF to families in WA may precipitate a move down the ‘socio-economic ladder’, whether because of diminished labour supply or other direct and indirect costs of chronic disease that hamper social opportunity and the ability to accumulate wealth. The concept of chronic disease leading to social disadvantage through reduced economic and social assets is widely recognised, yet the socio-economic impact of CF on families of young children is less understood. This merits further research to inform government social policy and local psychosocial screening and intervention strategies. It is not likely that these findings are confined to CF and other chronic diseases of childhood should be examined.

Descent in SES increases the risk of exposure of diagnosed children to health hazards associated with social disadvantage, including infection, food and housing insecurity and adverse health behaviour, in addition to morbidity and increased mortality conferred by CF. A social gradient in health exists within adult and paediatric CF populations, with lower SES among CF patients linked to worse pulmonary function, nutrition and increased mortality. To the authors’ knowledge, these are the first Australian data to suggest a social gradient may exist for families of young children with CF compared with the general population living in a relatively affluent state with moderate social gradients of health.

Residential mobility and marital breakdown were more common in this study cohort than in Australian population data and are independent risk factors for poorer child health and educational capital. Children who move frequently in early life (independent of SES) are at increased risk of inferior physical health, behavioural problems and lower educational achievement. Single parenthood may increase the risk of lower lung function and suboptimal nutrition in children with CF. Whether CF families experience lower rates of homeownership and insecure housing requires further study in larger populations.

In this cohort, one-third of preschool children were accessing psychological services and levels of maternal mental illness paralleled Australian population data. Significant levels of psychological distress within maternal–child and marital relationships may exist in early life in CF and have the potential to drive health-care use. This study cannot comment on causality and while it is plausible that parental psychological distress may precipitate ill health and hospitalisation in children with CF, it is equally possible that increased health-care use resulted from heightened clinical surveillance and intervention in response to parental and family distress.

A limitation is the relatively small sample size and potential for significant differences and associations to be missed. Reliance on documentation in notes rather than specific measures designed to capture psychosocial factors within the environment of the young child were limitations. Health-care use was restricted to hospitalisation, emergency and outpatient clinic attendance and did not capture community health-care use. Results may be biased towards families under stress where documentation of psychosocial data is more comprehensive. Further research is warranted to determine the findings’ generalizability to other CF populations and to other populations of families of children with chronic disease.
increased health-care use in early life. Further research is required to test these observations in larger populations (CF and other chronic disease) using detailed measures of family systems, parental functioning and economic resources, to elucidate the relationships between psychosocial factors, health-care use and health outcomes. These studies should be designed to account for causality and consider the biological mechanisms that link psychosocial distress to health outcomes. Intervention studies that focus on family-based psychosocial interventions in early life may reduce emergency presentations and hospitalisations in young children with CF, with significant health economic benefits.

Acknowledgements

We acknowledge the support and intellectual contribution of Ms Jenny Mace in the design of the study. This study was supported by the National Health and Medical Research Council of Australia CRE grant 513730.

References