Meeting the needs of children with medical complexity and their families: an investigation into health care service delivery for children with 22q11.2 Deletion Syndrome (22q11DS) and their families

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Statement of Originality

The thesis contains no material which has been accepted for the award of any other degree or diploma in any university or other tertiary institution and, to the best of my knowledge and belief, contains no material previously published or written by another person, except where due reference has been made in the text. I give consent to the final version of my thesis being made available worldwide when deposited in the University's Digital Repository, subject to the provisions of the Copyright Act 1968.

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The University of Newcastle Human Research Ethics and Hunter New England Human Ethics Committee approved the conduct of this research:

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Abstract

Introduction: over the past few decades there has been a rapid rise in the number of children with medical complexity (Kuo, Cohen et al. 2011), that is children with chronic condition(s), functional limitations, high health care needs and high health care utilisation (Cohen, Kuo et al. 2011), putting strain on health care systems and services. As a result, children with medical complexity and their families are at high risk of receiving insufficient health care and having unmet health care needs.

Aims: overall, the aim of this research project is to identify ways to reduce unmet health care needs and improve health care satisfaction for children with 22q11DS and their families. The specific objective of this research project is to develop an evidenced based model of service delivery that can inform further research into health care service delivery for children 22q11DS and potentially provide insight into strategies that may improve health care service delivery for children with complex medical conditions and their families.

Methods: a mixed method concurrent design including three studies was undertaken. Study one investigated the needs of children with 22q11DS and their families. This study utilised a survey of parents of children with 22q11DS, comprising the Family Needs Questionnaire (FNQ), and parent specific focus groups. Study two examined parents of children with 22q11DS experiences and satisfaction with health care service, using the Patient Satisfaction Questionnaire III (PSQ-III) and one-to-one interviews. Study three explored health care professionals' experiences and perspectives on the design and delivery of health care services for children with 22q11DS and their families using one-to-one interviews and focus groups. Data analysis for the quantitative component of the research included proportional analysis and one sample *t*-test, while the qualitative component utilised thematic analysis, descriptive phenomenology and a grounded theory approach.

Results: after collecting the data concurrently, as per the concurrent mixed methods design, the results were reported separately (Creswell, Klassen et al. 2010). Study one comprised of 49 parents of children with 22q11DS. Results showed that the top three unmet health care service needs, identified by parents, related to financial support, health care professionals showing

parents respect and health care professionals agreeing on the best way to treat their child. The qualitative component of this study included 24 parents of children with 22q11DS. The results identified five key needs: *care coordination, health care professionals of excellence, efficient health care, continuity and comprehensive care and support for carers.* Study two comprised of 50 parents of children with 22q11DS. The findings indicated that, parents of children with 22q11DS were significantly less satisfied with the delivery of health care than a comparative population. Parents of children with 22q11DS were most dissatisfied with the interpersonal aspects of their child's health care. The qualitative component of this study consisted of six participants, the results showed ten key transformed meaning units that described the parents' collective experience of health care services: (1) *Prenatal Complications* (2) *Early Post Natal Complications* (3) *Mothers Instinct ("The Observant Mum")* (4) *Before The Diagnosis* (5) *Potential Diagnosis And Information Gathering* (6) *Diagnosis* (7) *Post Diagnosis* (8) *The Genetic Counsellor* (9) *Appointments – Tests & Specialists* (10) *How To Get The 'Right' Care* (11) *Hospitals – Which One Delivers?* (12) *Qualities Of A Good Health Care Professional* (13) *Clinics* (14) *Coping With 22q11DS*. The final study used a grounded theory approach. This study included 38 health care professionals from six different countries. The results showed that the components of a potential theory of optimal health care service delivery for children with 22q11DS should include (1) a*n Expert Approach* (2) *Holistic Care* (3) *Integrated Care* (4) *Multidisciplinary Team* and (5) *Resource Efficiency*.

Discussion: the results were merged and compared side-by-side (Creswell, Klassen et al. 2010) showing the importance of addressing the models of health care for children with 22q11DS and their families. To address unmet health care needs and improve health care satisfaction, a model of service delivery for children with 22q11DS requires an integrated care approach. This will reduce siloing and fragmentation of health care, build skill, knowledge and experience in primary care and engage and equip parents to be an active part of the health care team.

Conclusion: this model of integrated care has the potential to raise the quality of health care service delivery for children with 22q11DS and their families as it is driven by their needs and experiences. It is hoped that the

proposed model of service delivery leads to greater satisfaction, better quality care for children with 22q11DS and more equitable distribution of expertise across professionals and setting.