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# Managing misaligned paternity findings in research including sickle cell disease screening in Kenya: 'Consulting communities' to inform policy☆

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ABSTRACT

The management of misaligned paternity findings raises important controversy worldwide. It has mainly, however, been discussed in the context of high-income countries. Genetic and genomics research, with the potential to show misaligned paternity, are becoming increasingly common in Africa. During a genomics study in Kenya, a dilemma arose over testing and sharing information on paternal sickle cell disease status. This dilemma may be paradigmatic of challenges in sharing misaligned paternity findings in many research and health care settings. Using a deliberative approach to community consultation to inform research practice, we explored residents' views on paternal testing and sharing misaligned paternity information. Between December 2009 and November 2010, 63 residents in Kilifi County were engaged in informed deliberative small group discussions, structured to support normative reflection within the groups, with purposive selection to explore diversity. Analysis was based on a modified framework analysis approach, drawing on relevant social science and bioethics literature.

The methods generated in-depth individual and group reflection on morally important issues and uncovered wide diversity in views and values. Fundamental and conflicting values emerged around the importance of family interests and openness, underpinned by disagreement on the moral implications of marital infidelity and withholding truth. Wider consideration of ethical issues emerging in these debates supports locally-held reasoning that paternal sickle cell testing should not be undertaken in this context, in contrast to views that testing should be done with or without the disclosure of misaligned paternity information. The findings highlight the importance of facilitating wider testing of family members of affected children, contingent on the development and implementation of national policies for the management of this inherited disorder. Their richness also illustrates the potential for the approach adopted in this study to strengthen community consultation.



## Introduction

The benefits and harms of sharing incidental findings on misaligned paternity during biomedical activities have been raised as an ethical issue in the literature from many parts of the world (Lucassen & Parker, 2001; Ross, 1996; Turney, 2005; Young et al., 2009). Given the concentration in high-income countries of biomedical activities likely to show this type of genetic information, guidelines, commentaries and empirical research on sharing incidental misaligned paternity findings have largely focused on those settings. The consensus of guidelines is that incidental misaligned paternity information should generally not be shared with parents, albeit with some controversy, reflecting reasoning that genetic testing should not be used in ways that disrupt families (Lucassen & Parker, 2001). In large-scale international surveys of attitudes to disclosing misaligned paternity findings in genetic testing, a majority of professionals expressed this attitude, although many



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women surveyed in the USA held a different view (Wertz, Fletcher, & Mulvihill, 1990; Wertz & Fletcher, 2004).

Recently, the question of potential for benefits and harms from sharing misaligned paternity information arose during a genomics study at an international biomedical research programme in Kilifi in coastal Kenya. The study, addressing genetic susceptibility and resistance to common serious childhood illnesses, included screening for sickle cell (SC) disease in a population of around 15,000 healthy infants (Marsh, Kamuya, Mlamba, Williams, & Molyneux, 2010). SC disease, a serious genetic disorder, occurs in just under 1% of infants in this area but is not well recognised within the community as a biomedical condition (Marsh, Kamuya, & Molyneux, 2011a). Parents of children found to have SC disease in the genomics study were informed of this result, and referred to a dedicated clinic at the district hospital - run collaboratively between researchers and government health providers – for counselling and long term care (Marsh et al., 2010).

Given the autosomal recessive inheritance of this condition, both parents of an affected child must be carriers of at least one sickle cell gene; a status referred to as having 'sickle cell trait'. As a corollary, where the social father of an affected child does not have either SC trait or SC disease, he cannot be the biological parent of that child. One unexpected outcome of sharing SC disease information in affected children during the genomics study, reported in detail elsewhere, was the emergence of several requests for paternal testing for SC trait, related to paternal denial of genetic responsibility for their child's condition (Marsh et al., 2011a). Some degree of paternal denial was described as part of a wider cultural tendency for mothers to be seen as mainly responsible for health problems in children in this setting. Paradoxically, this risk of maternal blaming was seen as potentially both *reduced* or *increased* by researchers disclosing information on the genetic roles of parents in SC disease, depending largely on influences within the family and at wider structural levels. Where fathers understood and accepted information on their genetic role in SC disease, paternal denial could be countered. Where explanations were interpreted differently, or not accepted, fathers might continue to deny their role. In others, shared understanding of the inheritance of SC disease might still be associated with paternal denial through doubts about biological fatherhood. Requests for paternal SC carrier testing were seen as particularly likely in families where fathers denied their role in their child's condition. In this way, researchers in Kilifi seemed to be presented with a moral dilemma in deciding how to respond to requests for paternal SC testing.

This paper reports on a study set up to consult a range of residents in Kilifi on the way researchers should respond to requests for paternal SC testing, including whether findings showing misaligned paternity should be shared. The consultation aimed to support the development of local policy on this potentially sensitive issue, as part of a wider research activity to explore residents' perceptions of SC disease and views on sharing information on the condition. Consulting people who will be affected by research policies in this way is widely recognised as morally and practically important, particularly where there may be significant differences between researchers and those who participate in studies, for example, in their technical knowhow, wealth, culture and language (Emanuel, Wendler, Killen, & Grady, 2004). In relation to sharing genetic findings, research ethics guidelines and commentaries highlight the importance of taking account of grounded views on the nature of possible harms and benefits in making decisions about disclosure, including how ethical challenges related to community interests should be met (Knoppers, Joly, Simard, & Durocher, 2006; Ravitsky & Wilfond, 2006).

There are many methodological and theoretical challenges in the literature on undertaking 'community consultation' to strengthen ethical practice, including how 'communities' are identified and represented (Kamuya, Marsh, Kombe, Geissler, & Molyneux, 2013; Tindana et al., 2007); how views are elicited, particularly around unfamiliar topics (Parker et al., 2009); and how these views should be fairly taken forwards to inform practice, as a normative rather than descriptive process (Dunn, Sheehan, Hope, & Parker, 2012; Ives, 2013). The consultation methods described in this paper draw upon principles of deliberative ethics in which public discussion is seen as central to the identification and analysis of ethical issues, as a substantive and pluralist model (Parker, 2007). Through this description, we make a contribution to the methodological literature on empirical ethics, although it is beyond the scope of the paper to describe the place of this study within current epistemological debates (Dunn et al., 2012; Ives, 2013). Rather, we show that a rich account of informed ethical reflection *by people affected by a specific moral dilemma* can be achieved through qualitative methods based on a structured and deliberative type of dialogue; and that the outputs are highly relevant to an overall process of normative analysis.

## Methods

### *Study site*

The Kenya Medical Research Institute (KEMRI) Wellcome Trust Research Programme, and the setting of its main centre in Kilifi County on the coast of Kenya, have been described in detail elsewhere (Marsh et al., 2010). In summary, Kilifi County includes rural and semi-urban populations of around 1 million; subsistence farming is the primary livelihood and between 55% and 65% households live below the poverty line (Virtual Kenya, 2011). The study was conducted within the population of 260, 000 people included within the research programme's Health and Demographic Surveillance System (KHDSS) that accounts for around 60% of admissions to the district hospital (Scott et al., 2012). This population constitutes the 'community' referenced throughout this paper. The majority of residents are Mijikenda (Parkin, 1991); 47% describe Christianity, 13% Islam and 24% traditional beliefs as their faith system. 45% adults reported an inability to read a newspaper or letter during randomised household surveys in 2005.

### *Study population, sampling and data collection*

Between December 2009 and November 2010, 63 Kilifi residents in the KHDSS area were engaged in a series of consultation activities to explore their views on the way researchers should respond to requests for paternal SC disease testing and sharing findings showing misaligned paternity. Drawing on experience in community engagement at the research programme over many years (Marsh, Kamuya, Parker, & Molyneux, 2011b; Marsh, Kamuya, Rowa, Gikonyo, & Molyneux, 2008) the consultation was planned as a series of small group discussions (9 groups) each with 3–6 people and held in two stages, and individual interviews (8).

Table 1 gives a summary description of participants. To inform practice, the consultation aimed to take account of the range of views likely to be encountered within the area, and to include those of mothers with an affected child. *A priori* purposive sampling was based on exploring diversity, using criteria of role, gender and rural/urban geographic residence, and all groups included participants of different ages, religion and educational status. Types of residents included: i) those working full time within the research programme (20), including Community Facilitators, Field Workers (front-line staff supporting studies in informed consent processes, interviews and sample-taking), Data Entry Clerks and a Scientist in training; ii) District Health Managers (4); iii) Administrative leaders, Chiefs and Assistant Chiefs (18); iv) KEMRI Community Representatives (KCRs)