

**B Cell antigen D8/17 as a marker of susceptibility to
rheumatic fever in Australians**

and

The sharp end of the needle:

**Rheumatic fever prophylaxis and concepts of care for
Yolngu patients**

A thesis in two parts

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Statement of work undertaken

In part 1, the project was conceptualised by Prof. Bart Currie and Prof. Jonathan Carapetis. I principally performed the data and sample collection, the statistical analysis and interpretation of the D8/17 results. I undertook the D8/17 staining of the community samples, with assistance from Mr Michael Harrington. Kumar Visvanathan and Narelle Skinner stained the samples in the laboratory. The D8/17 flow cytometry was performed by Dr Kumar Visvanathan.

In part 2, I conceptualised and planned this study, with the assistance of my supervisor, David Thomas. My co-researcher, Joy Bulkanhawuy and I developed the interview structure and carried out the interviews in a bilingual fashion. Joy translated the interviews, and then participated in the analysis of the data. I performed the bulk of the analysis and write-up of the data. Joy assisted with the feedback to the community, and the review of the final conclusions.

Declaration

I certify that this thesis does not incorporate without acknowledgement any material previously submitted for a degree or diploma in any university; and that to the best of my knowledge and belief it does not contain any material previously published or written by another person except where due reference is made in the text.

Abstract

Aboriginal Australians have some of the world's highest rates of rheumatic fever. Two approaches to reducing the burden of rheumatic fever are discussed in this thesis. The B cell antigen D8/17 has a strong association with rheumatic heart disease and may be a universal marker of inherited susceptibility to rheumatic fever. Identifying a population at increased risk of rheumatic fever provides an opportunity to focus primary prevention measures. **In part one of the thesis** I evaluate the accuracy of D8/17 as a marker of past rheumatic fever amongst Australians from the Northern Territory. D8/17 levels were measured and compared in patients with acute rheumatic fever, rheumatic heart disease or past rheumatic fever, first-degree relatives and healthy, unrelated controls. The mean percentage of B cells positive for D8/17 was 83.7%, 38.9%, 20.2% and 11.6% respectively. The difference between the groups was significant ($p < 0.0001$). A receiver operator curve analysis indicated that 22.1% of B cells positive for D8/17 was the most accurate cut-off to distinguish patients with acute or past rheumatic fever from healthy subjects. These results indicated that the B cell antigen D8/17 is an accurate marker of past rheumatic fever in Aboriginal Australians and could be a helpful addition to the Jones Criteria for strengthening or excluding a diagnosis of acute rheumatic fever. The intermediate levels of D8/17 expression in the relatives of index cases supports the hypothesis that D8/17 is a marker of an inherited susceptibility to rheumatic fever, although prospective trials are required to provide conclusive proof of this hypothesis.

Non-compliance with secondary prophylaxis was suspected to be the cause of increasing rates of rheumatic fever in the Top End. **In part two of the thesis** I discuss the 'problem of compliance' with respect to Aboriginal patients, and investigate the

factors that affected the delivery and uptake of prophylaxis for rheumatic fever in an Aboriginal community. Patients, relatives and health practitioners were interviewed on the topic of the care of patients with rheumatic heart disease. The data were analysed using the principles of grounded theory.

The main finding was the desire for more personalised care and support for patients with rheumatic heart disease from the community clinic, rather than simple medical care. These ideas crystallised through two Yolngu terms to describe care: *djaka* (to physically care for) and *gunga'yun* (to encourage). Thus even from the outset there was divergence in the focus of the 'consumer' - holistic care - and that of the health-care professional/ researcher – improving the rate of secondary prophylaxis coverage.

With regards to service provision, a significant reason for failure to receive secondary prophylaxis was the differing approaches of urban and community health services, patient mobility, and a differing understanding of the responsibilities of patients and health service providers in the different settings. Other factors pertaining to service provision, such as staff motivation, administrative issues and program coordination affected the uptake of secondary prophylaxis to a lesser extent. With regards to treatment uptake, individual patient factors inhibiting uptake of treatment were apparent in some cases, but treatment refusal was rare. Pain was not found to be a deterrent. No simple relationship was found between treatment compliance and biomedical knowledge of the disease. There was no simple relationship between patient passivity and sense of responsibility that guaranteed compliance.

This study demonstrated that the failure to achieve good uptake of prophylaxis for rheumatic fever related as much to factors of service provision as patient factors and

that providing holistic care within a familiar and supportive framework is important to Yolngu patients. However, there are real difficulties for health services as they are currently structured to meet the expectations of patients and families.

Abbreviations

| | |
|------|---|
| ALPA | Arnhem Land Progress Association |
| ARDS | Aboriginal Resource and Development Society |
| JB | Joy Bulkanhawuy |
| NT | Northern Territory |
| PBMC | Purified blood mononuclear cells |
| RF | Rheumatic fever |
| RHD | Rheumatic heart disease |
| ROC | Receiver operator characteristics |
| SD | Standard deviation |
| WHO | World Health Organisation |
| ZH | Zinta Harrington |

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PART 1

**B Cell antigen D8/17 as a marker of susceptibility to
rheumatic fever in Australians**

1 Introduction

1.1 Background

Rheumatic fever (RF) is a shifting shadow: each breakthrough in the management of this disease has been matched by an unresolved problem. Said to have “vanished” in the industrialised world, its effects are increasingly recognised in developing countries. Although RF was recognised in the 19th century and diagnostic criteria were defined in 1944, accurate diagnosis remains elusive and there is still no definitive diagnostic test. The familial tendency of RF has been noted for more than a hundred years, but the nature of any inherited susceptibility factor is unknown.

Understood for half a century to be a post-streptococcal autoimmune phenomenon, the pathogenetic pathway between infection and valve damage is still not understood. The prevention of recurrent RF with penicillin was a breakthrough in patient management - dramatically reducing morbidity and mortality in many countries - yet human and environmental factors have prevented the control of this disease in poor countries and amongst disadvantaged sub-groups in wealthy countries.

RF is an active problem in Indigenous communities in Australia, unlike in the majority of the developed world where it has become exceedingly rare. This spectre hovers over Aboriginal children, affecting between two and five in every thousand each year (Carapetis, Currie et al. 2000). Young adults are learning to live with painful injections, chronic rheumatic heart disease (RHD), cardiac surgery and complicated pregnancies. Some die, and they die young. Despite improvements in access to medical services the burden of disease has not reduced. RHD registers, concerted primary and secondary prophylaxis programmes, and efforts to improve

sanitation, living conditions and reduce skin sepsis have all failed to produce a sustained benefit so far. New ways to reduce the burden of disease are being sought, addressing both primary attacks of rheumatic fever (RF) as well as recurrent RF.

Epidemiologic, genetic and laboratory evidence support the existence of an inherited susceptibility to RF. The World Health Organisation has identified the search for a genetic marker for susceptibility to RF as a worthy area for research (Anonymous 1995b). The most encouraging relevant discovery to date is the B cell antigen D8/17 that was present at an elevated level in 100% of RHD patients in the US and only 15% of healthy controls (Khanna, Buskirk et al. 1989). This marker was expressed in intermediate levels in family members of affected cases, suggesting that it represented an inherited trait. D8/17 has been evaluated in a number of other populations around the world with similar results (Gibofsky, Khanna et al. 1991; Herdy, Zabriskie et al. 1992; Harel, Zeharia et al. 2002). As a screening test, D8/17 may identify a population susceptible to RF, which could be targeted for primary prevention strategies. The World Health Organisation (WHO) recognises the value of further research to identify a population at higher risk for developing RF, as efforts at primary prevention of RF could be directed at this group (Anonymous 1988; Anonymous 1995b). This is particularly desirable in the event of the development of an effective Group A streptococcal vaccine.

Furthermore, it has been proposed that if D8/17 is an accurate marker of susceptibility to RF, it may augment the accuracy of the Jones criteria in the diagnosis of RF. When the diagnosis of RF is uncertain, patients need to be closely followed up for a number of years. The risk of missing a diagnosis includes heart damage and premature death;

over-diagnosis may lead to years of unnecessary painful injections and a considerable outlay of scarce resources.

1.2 The aim

There are two basic aims for this project. The first aim is to evaluate the accuracy of D8/17 as a marker of susceptibility to RF in Aboriginal Australians by examining its frequency in patients with a past history of RF, first-degree relatives and controls. This will add to the international observations regarding antigen D8/17 and RHD susceptibility.

The second aim of this project is to adapt the D8/17 assay to the clinical scenario of the greatest relevance in Australia: the testing of Aboriginal people living in remote settings. The effect of time-delay on staining of blood for D8/17 assay as well as the accuracy of the D8/17 assay on frozen and stored specimens will be evaluated.

1.3 The scope

In the literature review, the evidence for an inherited susceptibility to RF will be evaluated. However, it is beyond the scope of this study to ascertain the genetic basis of the inheritance of D8/17. Nor will the study identify the means by which the protein is involved in the pathophysiology of the disease.

Study subjects were recruited predominantly from one Aboriginal community and one tertiary referral hospital. The results from the two centres and those gathered elsewhere were compared in order to ascertain the generalisability of any conclusions.

2 The evidence for a genetic susceptibility to rheumatic fever

2.1 Introduction

The familial tendency of RF has been recognised for more than a century. Whether this is due to an inherited susceptibility, or a common environmental influence was not clear for some time. The debate has been characterised thus:

...the inherited factor may not be an increased susceptibility to rheumatic fever per se but rather an altered host response to repeated infection, the latter being enhanced or becoming recurrent within the family circle when poor housing and crowding in the home are present. (Gray, Quinn et al. 1952)

There has been much work to clarify the respective input of the environment versus host factors in the causation of RF. In this chapter I present the evidence for an inherited susceptibility to RF.

2.2 Epidemiology

RHD is known to cluster in families (Pickles 1943; Beers 1948). The relative risk of RF in patients with a family history of the disease has been estimated using clinical observation, a cohort study and a twin study, thereby providing indirect evidence of an inherited susceptibility for RF. In his 1889 treatise on rheumatism in childhood, Cheadle examined the family histories of 492 children admitted to the Hospital for Sick Children “for diseases of all kinds, both medical and surgical”. One hundred and

seventy three children had evidence of a “clear history of acute rheumatism in blood relatives”. In this group, 38 children developed RF. In the remaining 319 children without a family history of RF, only 15 developed RF. Cheadle concluded “that with a family history of acute rheumatism in immediate blood relatives the chance of an individual with such hereditary tendency contracting acute articular rheumatism is nearly five times as great as that of an individual who has no such hereditary taint” (see table 1) (Cheadle 1889). Cheadle admitted that his data lacked accuracy as he had difficulty confirming the diagnosis of rheumatism in family members. Whilst there is insufficient detail to assess the methods of this investigation, issues of recall bias may well have affected the likelihood of patients with rheumatism admitting to a family history of the condition more than the children who presented with other conditions. Thus the odds ratio calculated by Cheadle may be an overestimate.

Table 1 - Risk of developing RF with a family history of ‘acute rheumatism’ in a first-degree relative, case-control study

| | Family history of RF | No family history of RF | Total |
|-------------------------|-----------------------------|--------------------------------|--------------|
| Child with RF | 38 (22%) | 15 (5%) | 53 |
| Child without RF | 135 (78%) | 304 (95%) | 439 |
| Total | 173 | 319 | 492 |

Odds ratio 4.67; 95% confidence interval 2.64 to 8.24, calculated from the data provided. Source: (Cheadle 1889)

More convincing evidence emerged from a cohort study in an Israeli Kibbutz, where children of a similar age were raised in communal homes separate from their parents. Over a one-year period of follow-up, children with a parental history of RHD were

nearly three times as likely to develop RF as children without a family history. Affected children were also more likely to develop streptococcal infections than their peers who had identical exposure. Therefore it is not clear if the susceptibility was not to RF *per se*, but instead to the development of streptococcal infection when exposed. From the data provided, the risk difference of developing RF given a history of RF in either parent was 36% (see table 2). It should be noted that the study described an incredibly high incidence of RHD (Davies and Lazarov 1960).

Table 2 - The risk of developing RF with a history in either parent, cohort study

| | One or both parents with RHD | Neither parent with RHD | Total |
|-------------------------|-------------------------------------|--------------------------------|--------------|
| Child without RF | 12 (44%) | 47 (81%) | 58 |
| Total | 27 | 58 | 85 |

Relative risk 2.93, 95% Confidence intervals 1.56 to 5.50, calculated from the data provided. Source: (Davies and Lazarov 1960)

This contrasts with a report of 56 sets of twins that revealed 19% concordance of RF in monozygotic twins and only 5% concordance in dizygotic twins (Taranta, Torosdag et al. 1959). In this study monozygotic twins were nearly four times more likely than dizygotic twins to have concordance for RF but the result was not statistically significant (see table 3). This compares with the relative concordance rates for multiple sclerosis: 25.9% concordance in monozygotic twins and 2.3% in dizygotic twins (Hawkes 1997). However as the selection process for the twin pairs was not stated, it is possible that selection bias may have affected the result. Also, twin studies rely on the assumption that the environment is shared for each set of twins, or that zygosity does not affect the incidence of disease. An exception to this rule is where

dizygous twins of differing sexes may be raised quite differently, whilst monozygous twins will not be affected by the social gender differentiation. While recognising these methodological concerns, this study shows that the concordance for RF in monozygous twins is only 19%, suggesting that genetic factors, whilst they play a significant role, are not the only determinants of disease.

Table 3 - The concordance of RF in twins

| | Monozygous twins | Dizygous twins | Total |
|--------------------------|-------------------------|-----------------------|--------------|
| Concordant for RF | 3 (19%) | 2 (5%) | 5 |
| Discordant for RF | 13 (81%) | 38 (95%) | 51 |
| Total | 16 | 40 | 56 |

Relative risk 3.75, 95% Confidence intervals 0.69 to 20.38, calculated from the data provided. Source: (Taranta, Torosdag et al. 1959)

It appears that generally a similar proportion people infected by Group A streptococci develop RF in various populations, suggesting a uniform prevalence of susceptibility to RF. Amongst recruits in military barracks in the United States during the Second World War three percent of patients with streptococcal pharyngitis went on to develop RF, independent of variation in streptococcal strains (DiSciascio and Taranta 1980). In the Top End of Australia where there is a high rate of streptococcal infection and transmission, the lifetime cumulative incidence of RF is between 2.7 and 5.7%, which may represent the entire susceptible population (Carapetis, Currie et al. 2000). An exhaustive review of the epidemiology of RF, including historical and contemporary surveys, found the maximum prevalence of RHD across a variety of countries to be generally less than 6% and mostly less than 3% (Quinn 1989). If we assume that only

half of the people who contract RF develop RHD, then the potential lifetime cumulative incidence of RF in most populations would be approximately 6%. As comprehensive as this review may be, it is limited by the availability of quality data from developing countries. Occasional reports of extremely high prevalences of RHD - 10% in Brazil and 7% in Vietnam (Kaplan 1993)– suggest that there may be some populations with a higher prevalence of RF susceptibility.

2.3 Genetics

The mode of inheritance of the susceptibility to RF remains uncertain. Autosomal recessive (Wilson, Schweitzer et al. 1943; Wilson and Schweitzer 1954; Hafez, Chakravarti et al. 1985; Khanna, Buskirk et al. 1989), autosomal dominant with incomplete penetrance (Pickles 1943; Beers 1948; Gerbase-DeLima, Scala et al. 1994), sex-linked (Draper and Seegal 1923) and indeterminate (Gray, Quinn et al. 1952; Stevenson and Cheeseman 1953; Uchida 1953) modes of inheritance have all been proposed. Diallelic inheritance of the susceptibility to RF has also been demonstrated (Zabriskie, Lavenchy et al. 1985).

A study showing that siblings were more likely to have similar clinical presentations of RF than unrelated persons (particularly relating to chorea) suggested the influence of heredity on the clinical manifestations of RF (Spagnuolo and Taranta 1968). However the authors were not able to rule out the role of environmental confounders, or selection bias. Furthermore, as argued in a subsequent letter to the editor, the RF cases were defined by reference to the Jones criteria, yet carditis and polyarthritis were the primary outcomes measured indicating circularity in the research design (Rammelkamp and Rutstein 1968). The data provided by Taranta *et al*, consisting of

six twin pairs(Taranta, Torosdag et al. 1961) are insufficient to support DiSciascio's assertion that the clinical manifestations of RF in monozygotic twins are more concordant than in dizygotic twins (DiSciascio and Taranta 1980).

2.4 Blood groups, secretor status and rheumatic fever susceptibility

Persons with blood groups other than O and those who don't secrete ABO (H) substances in their saliva were found to have an increased risk of a history of RF although the relationship was not strong (Clarke, McConnell et al. 1960; Buckwalter, Neifeh et al. 1962; Edwards 1962). Lewis antibodies have also been implicated – a small but significantly increased risk of RF in Lewis positive secretors has been noted (Glynn and Holborow 1961). However the attributable risk from these blood group features was small and no convincing link with pathogenesis has been made.

2.5 Rheumatic fever and HLA associations

With the discovery of human leukocyte antigens (HLA) and their relationship to connective tissue diseases and autoimmunity, a search began for an association between HLA and RF (Falk, Fleischman et al. 1973; Caughey, Douglas et al. 1975; Murray, Montiel et al. 1978; Hafez, Chakravarti et al. 1985; Anastasiou-Nana, Anderson et al. 1986; Taneja, Mehra et al. 1989; Guilherme, Weidebach et al. 1991; Gerbase-DeLima, Scala et al. 1994; Weidebach, Goldberg et al. 1994; Guedez, Kotby et al. 1999; Visentainer, Pereira et al. 2000). The great diversity of results is demonstrated in table 4. The HLA associations described are not consistent across the different ethnic groups studied, although many studies describe a relationship with an HLA-DR allele and RF. In some cases, HLA antigen subsets have been found to be

reduced in the rheumatic group, for example HLA-A3 (Falk, Fleischman et al. 1973) and HLA-DR6 (Anastasiou-Nana, Anderson et al. 1986) implying a protective role for these HLA subsets. It seems plausible that a hitherto unidentified rheumatogenic gene may be in linkage disequilibrium with certain HLA subtypes, or that HLA may act as a cofactor in disease causation, somewhat like the rheumatic epitope in rheumatoid arthritis (Stastny 1978). Furthermore, the problem of significance shrouds most of the studies looking at relationships between HLA alleles and RF. In many studies multiple HLA types were tested against the incidence of RF, without acknowledgement of the increasing possibility of a chance false positive result with the increasing number of tests performed.

Different techniques for HLA ascertainment have produced different results even within the same population. Guedez argues that most of the studies investigating HLA associations in RF are unreliable because of technical issues relating to the serological typing reagents that are unable to differentiate between HLA allelic splits and subdivisions (Guedez, Kotby et al. 1999). Molecular techniques to investigate DNA may yield more reproducible results. Furthermore, according to Guedez, researchers have failed to clinically define a homogenous group for study. Thus, there is room for further work to investigate HLA associations in RF.

Table 4 - Relationship between HLA type and RF

| Study | Lab technique | Population | HLA protective for RF | HLA and risk of RF | OR (EF) |
|---------------------------------------|--------------------|-----------------------------|------------------------|------------------------|----------------------------|
| Falk, Fleischman et al. 1973 | Serology | USA | A3 | - | - |
| Caughey, Douglas et al. 1975 | Serology | New Zealand, indigenous | A10 | A3 A8 | Infinity* 9* |
| | | New Zealand, non-indigenous | A28 | A17 | 39* |
| Murray, Montiel et al. 1978 | MLCA | Mexican-Americans | Nil found | Nil found | - |
| Hafez, Chakravarti et al. 1985 | MLCA | Egypt | - | B5 | 2.5** |
| Anastasiou-Nana, Anderson et al. 1986 | MLCA | USA | DR6 | DR4 | 2.3 (0.3) |
| Ayoub, Barrett et al. 1986 | MLCA | USA, blacks | DR5 | DR2 DR4 | 3.86 (0.40) 2.02 (0.07) |
| | | USA, Caucasians | | DR4 DR9 | 3.55 (0.45) 18.8 (0.16) |
| Guilherme, Weidebach et al. 1991 | MLCA | Brazil | - | DRw53 DR7 | 4.2 (0.43) 3.8 (0.56) |
| Weidebach, Goldberg et al. 1994 | Molecular genetics | Brazil | - | DR53 DR16 | 2.5* |
| Guedez, Kotby et al. 1999 | Molecular genetics | Egypt | DQA1*0103 DQB1*0603 | DRB1*0701 DQA1*0201 | 2.5 1.8* |
| Visentainer, Pereira et al. 2000 | MLCA | Brazil | | DR7 | 2.4 (0.003) |

MLCA= microlymphocytotoxicity assay, using HLA antisera; OR= odds ratio; EF= aetiologic fraction – the fraction of the overall risk attributable to a single cause

*Calculated from data provided

**Result not significant

2.6 Evidence for an inherited altered immune response in rheumatic fever

Ayoub presents evidence of an inherited abnormality in the immune response in RF patients. Immune hyper-responsiveness after Brucella vaccination was shown to be a risk factor for subsequently developing RF (Ayoub 1984). The antibody response to streptococcal products in RF patients is variable; no hyper-responsiveness to streptolysin O or streptokinase has been demonstrated, but there is a significant elevation of antibody to Group A streptococcal carbohydrate at 3 years in patients with rheumatic carditis when compared with patients with Sydenham's chorea or post-streptococcal glomerulonephritis. The elevation of the Group A streptococcal carbohydrate antibody appeared to persist only in patients with ongoing cardiac lesions, but not in those in whom the carditis was transient – 90% of patients with persistent mitral insufficiency versus 20 % with transient or no carditis had elevated levels of anti Group A streptococcal carbohydrate antibody at 8 years after RF.

A few animal studies suggest that immune response to streptococcal antigens is under genetic control. A study of the response of inbred mice to immunisation with Group C streptococcal carbohydrate showed that genetically related mice were more likely than unrelated mice to produce identical antibodies (in terms of the Fab portion, variable region) to streptococcal antigens (Eichman and Kindt 1971). Based on this data

Ayoub conjectured that there exists a genetically determined, inherited abnormality in the immune response to Group A streptococcal carbohydrate that predisposes to RF with cardiac involvement. Furthermore, extrapolating from the animal studies, the genetic locus of the anti-carbohydrate response may be located near the HLA-DR locus. Whilst there is some appeal to this hypothesis, there is insufficient evidence to prove the pathogenicity of the anti-carbohydrate antibody, and some evidence that it may simply be an epiphenomenon. There are no human studies showing that the response to carbohydrate is genetically controlled. Ayoub's theory also does not account for the HLA associations found in other populations with high incidences of rheumatic disease, and finally it makes no attempt to explain the pattern of chorea and its inheritance.

Another hypothesis of inheritance arises from evidence that shows that rheumatogenic streptococci are more adherent to the epithelial pharyngeal cells from patients with RHD than normal controls in vitro (Selinger, Julie et al. 1978). The mechanism of adherence was not identified, but the authors conjecture that a cell surface component distinct from the HLA and blood group systems may be involved. Hafez confirmed this finding and further showed that the increased adherence of rheumatogenic streptococci to pharyngeal cells is "probably inherited" in autosomal dominant fashion, closely linked to HLA (Hafez, Abdalla et al. 1990). Pharyngeal cells from children with RF show increased adherence for rheumatogenic streptococci when compared with their own siblings and also unaffected controls (Haffejee 1992).

Cell-mediated immunity is also modified in patients with RF. There is evidence of increased leucocyte migration inhibition, increased natural killer cell cytotoxicity,

exaggerated cytotoxic cell activation by streptococcal proteins, increased tumour necrosis factor (TNF) and interleukin 2 (IL-2) (Haffejee 1992). To what extent these findings are the result of RF rather than the cause of the disease is unclear. Another study showed increased killer T cells (CD8+) and decreased IL-2 in both RF patients and their unaffected siblings (Alarcon-Riquelme, Alarcon-Segovia et al. 1990). These conflicting results have not been reconciled. Thus both in humoral immunity and cell-mediated immunity there exists some evidence of an abnormality that distinguishes RF patients from healthy controls. This abnormality may be inherited.

2.7 Conclusion

In the interplay between the host, the organism and the environment, the role of host factors achieves particular significance in RF due to the autoimmune pathophysiology of the disease. There is epidemiologic, genetic and immunologic evidence for an inherited susceptibility to RF, although the results are heterogenous and at times conflicting. The nature of this susceptibility and its mode of inheritance remain controversial. The most consistent evidence of an inherited susceptibility to RF to date relates to B cell antigens, which will be discussed in the next chapter.

3 B cell antigens and rheumatic fever

3.1 Preliminary discoveries – B cell antigens 883 and 256

Hypothesising that non-HLA B cell antigens might be markers of inherited abnormalities in the immune system, Patarroyo's group used the sera from multiparous women to screen the B cells of patients with RF from the US and Colombia. (Women generate alloantibodies to the B cell antigens of their partners during pregnancy.) Sera with specificity for B cells were selected for testing against patients with documented RHD. One serum showed a strong association with RF: serum 883 reacted with the B cells of 71-75% of rheumatics, but only 16-17% of healthy controls. The relative risk of 883 for RHD was 12.94 and the difference between the groups was significant ($p < 0.001$). The pattern of reaction of 883 in other diseases such as post-streptococcal glomerulonephritis, systemic lupus erythematosus, rheumatoid arthritis, multiple sclerosis, tuberculosis and leprosy was described as unremarkable. No association was found with the HLA-A, B or D antigens known at that time (Patarroyo, RJ et al. 1979; Patarroyo, Winchester et al. 1980).

As serum 883 was human derived and exhaustible, it became imperative to develop a laboratory clone. Mice were immunised with B cells derived from patients with RHD, including some who had not reacted to serum 883. Splenocytes from these mice were fused with myeloma cells. One of the resulting supernatants - 83S19.23 - reacted with rheumatic and non-rheumatic B cells in an identical way to serum 883 (Yeadon, Buskirk et al. 1984; Zabriskie, Lavenchy et al. 1985).

Sera 883 and 83S19.23 were evaluated in a number of ethnically and geographically distinct populations (Figure 1). The results in many locations were reasonably similar, although the antigen had less predictive value in Egypt or New Zealand than in other populations. Whether this was a true finding of ethnic difference in B cell antigen associations with RF or due to technical errors or differences is not known.

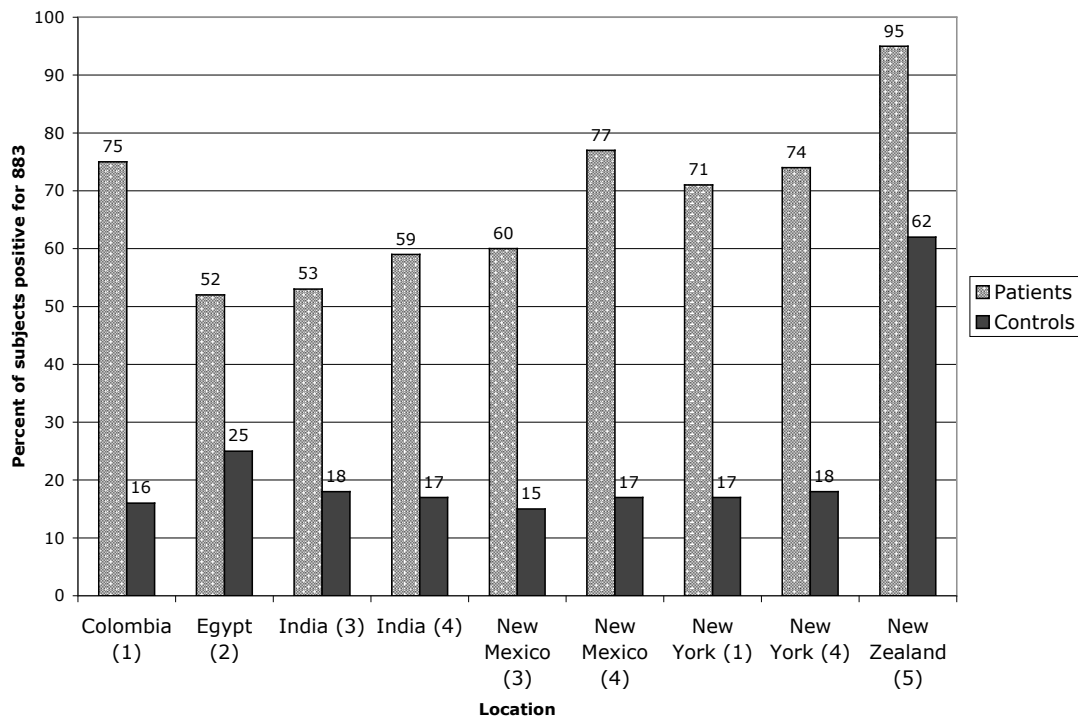


Figure 1 - Serum 883 (or its clone 83S19.23) as a marker of past RF or RHD

Sources: (1)(Patarroyo, Winchester et al. 1980) (2)(Gray, Regelman et al. 1984) (3)(Williams, Raizada et al. 1985) (4)(Yeadon, Buskirk et al. 1984) (5)(Regelman, Talbot et al. 1989)

In a New Zealand study that included Maori, Polynesian and Caucasian subjects, rheumatics, unaffected family members and controls were tested for reactivity to the 83S19.23 serum. The means of the percentage of B cells reactive to 83S19.23 in the three groups were 29.6%, 20.7% and 10.0% respectively, and the difference between

the groups was significant ($p=0.0003$). The intermediate expression of the marker in unaffected family members supported the hypothesis that the marker identified by 83S19.23 was inherited. The antigen identified by serum 83S19.23 was shown to have non-Mendelian inheritance via a single dominant locus (Rich, Gray et al. 1988).

Further antibodies were developed: antibody 256S10 identified five of seven patients with RHD who were negative for 883. The combination of 83S19.23 and 256S10 identified 92% of a small group of RF patients from the US and India. A di-allelic susceptibility to RF was proposed, with the 256S10 antibody indicating a milder form of the disease (Zabriskie, Lavenchy et al. 1985). However only 56% of patients in Egypt were correctly identified by the combination of markers (Gray, Regelman et al. 1984). Serum D8103 was also evaluated but found to have less specificity for RHD than the other sera discussed above (Rich, Gray et al. 1988; Regelman, Talbot et al. 1989).

An understanding of the role of the 883 and 256S10 antigens in the disease process was sought. The 883 antigen was shown to be spatially associated with the group A streptococcal binding sites on B cells. However, 883 antibody did not block the group A streptococcal binding sites on B cells, thereby indicating the functional distinction between 883 and the streptococcal binding sites. Streptococcal blastogen stimulation of blood and tonsillar tissue did not result in the up-regulation of 883 or 256S10 positive cells, refuting the hypothesis that antigen expression is simply a consequence of streptococcal infection (Williams, Raizada et al. 1985).

Despite larger numbers of 883 and 256 positive cells in peripheral blood, RF patients have fewer 883/256 positive lymphocytes in tonsillar tissue – the proposed site of streptococcal binding in the pathogenesis of the disease – when compared with healthy controls (Gray, Regelman et al. 1984). The implications of this finding are not fully understood, although it may further support the existence of an immune abnormality in patients with RF.

3.2 Anti D8/17 antibody

Further anti-rheumatic sera were prepared by repeatedly vaccinating a mouse with B cells from a patient with past RF and then fusing the mouse spleen cells with mouse myeloma cells (Khanna, Buskirk et al. 1989). The multiple antibodies produced were screened against 883-positive and negative RF patients. Serum D8/17 was selected for further study and was tested prospectively against 84 patients with past RF and 76 controls in the USA and West Indies. The mean level of expression of D8/17 was significantly different for patients with a past history of RHD and healthy controls, 33.5% and 5-7% of B cells respectively, $p < 0.0001$. There was intermediate expression of D8/17 by first-degree relatives of the index case (13-14.6% of B cells), suggesting that D8/17 expression was inherited. The results were also expressed as a categorical variable: subjects were defined as D8/17 positive if the B cell percentage was greater than one standard deviation above the mean for controls. When analysed in this way D8/17 identified 99% of subjects with past RF, with a false positive rate in controls of 14%. Importantly, post-streptococcal glomerulonephritis and uncomplicated streptococcal tonsillitis were not associated with increased expression of D8/17 (Khanna, Buskirk et al. 1989; Kemeny, Husby et al. 1994).

The anti-D8/17 antibody has been tested internationally with consistent results in most studies (see table 5 and figure 2). Further small studies in the US and Canada supported Khanna's findings (Herdy, Zabriskie et al. 1992; Feldman, Zabriskie et al. 1993). In Russia, Mexico and Chile, more than 90% of patients with RF, and less than 16% of controls, were positive for D8/17 (Gibofsky, Khanna et al. 1991). However, the specific methodology and cut-off points used to define D8/17 positivity for these assays were not stated. In Israel, over 90% of rheumatics and no healthy controls had more than 7.55% of B cells positive for D8/17 (Harel, Zeharia et al. 2002). It seemed at this stage that a universal marker for RF susceptibility had been found. However, three groups in India found significantly less correlation of D8/17 with rheumatic fever, even though RF patients still expressed higher levels of D8/17 when compared with controls (Taneja, Mehra et al. 1989; Ganguly, Anand et al. 1992; Kaur, Kumar et al. 1998). Whilst different from the international results, there was consistency between the Indian studies. D8/17 could no longer be assumed to have accuracy in populations of differing ethnic origins.

Table 5 - Comparison of studies relating D8/17 expression and RF or RHD

| Study | RHD or RF/ controls (number) | Substrate Measurement technique | Cut-off (% D8/17 positive B cells) | Patients positive for D8/17 (%) | Controls positive for D8/17 (%) |
|---|------------------------------------|---|---|---------------------------------------|---------------------------------------|
| (1) Khanna, Buskirk et al. 1989, USA and West Indies | 84/ 76 | PBMCs Fluorescence microscopy | >12 | 99 | 14 |
| (2) Herdy, Zabriskie et al.1992, USA | 10/ 14 | Whole blood Fluorescence microscopy | >12 | 90 | 0 |
| (3) Gibofsky, Khanna et al. 1991, Russia | 82/ 78 | Not described** | 1SD above mean for controls | 90-96 | 5 |
| (3) Gibofsky, Khanna et al. 1991, Mexico | 39/ 72 | Not described** | 1SD above mean for controls | 89 | 8 |
| (3) Gibofsky, Khanna et al. 1991, Chile | 50/ 50 | Not described** | 1SD above mean for controls | 90 | 16 |
| (4) Feldman, Zabriskie et al. 1993, Canada | 4†/ - | PBMCs Fluorescence microscopy | >20 | 100 | - |
| (5) Harel, Zeharia et al. 2002, Israel | 22/ 9 | Whole blood Flow cytometry | >7.55 | 90.2 | 0 |
| (6) Taneja, Mehra et al. 1989, India | 54/ 32 | B cells Fluorescence microscopy | Not specified | 63 | 13 |
| (7) Ganguly, Anand et al. 1992, India | 90/ 50 | B cells Fluorescence microscopy | >40 | 66 | 14 |
| (8) Kaur, Kumar et al., 1998 India | 24/ - | B cells Flow cytometry | >10 | 71 | - |
| | 140/ 50 | Whole blood Fluorescence microscopy | >10 | 65* | 14 |

PBMC = peripheral blood mono-nuclear cell, SD = Standard deviation

*Calculated as a mean value between RF and chronic RHD patients.

**These results described in Gibofsky's article are not referenced and presumably not published.

†This includes 2 patients with chorea and 2 with past RF (no details given)

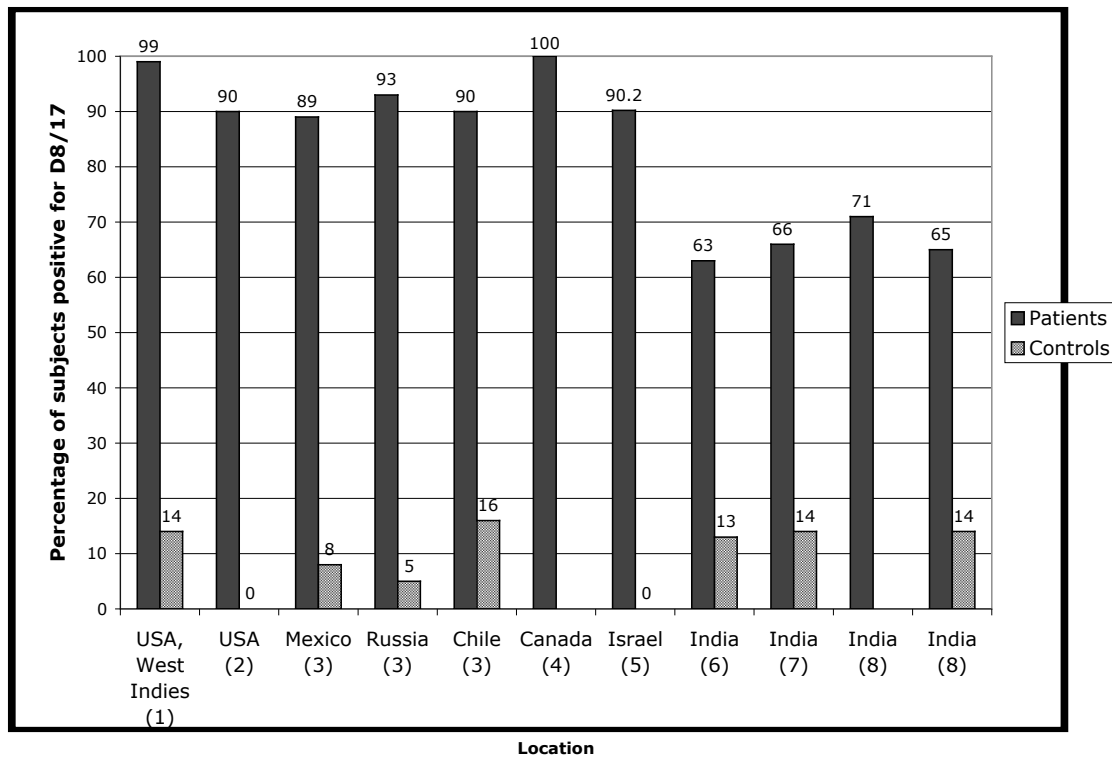


Figure 2 - D8/17 as a marker of past RF or RHD

Sources (1)(Khanna, Buskirk et al. 1989)(2)(Herdy, Zabriskie et al.

1992)(3)(Gibofsky, Khanna et al. 1991)(4)(Feldman, Zabriskie et al. 1993)(5)(Harel,

Zeharia et al. 2002)(6)(Taneja, Mehra et al. 1989)(7)(Ganguly, Anand et al.

1992)(8)(Kaur, Kumar et al. 1998)

In India monoclonal antibodies PG-12A, PG-12A and PG-20A produced by injecting mice with B cells of Indian RHD patients and fusing the spleen cells with myeloma cells proved to be more sensitive and specific than D8/17 at detecting RF and past RHD (see figure 3) (Kaur, Kumar et al. 1998; Kumar, Kaur et al. 1998; Kumar, Kaul

et al. 2001). There was variable expression of the antibodies within individual subjects, suggesting that the tests could be combined to identify an increased proportion of susceptible individuals (Kumar, Kaur et al. 2000). Furthermore, a dot-ELISA test was developed as an effective, cheap and simple test for susceptibility to RHD (Kumar, Kaur et al. 1998).

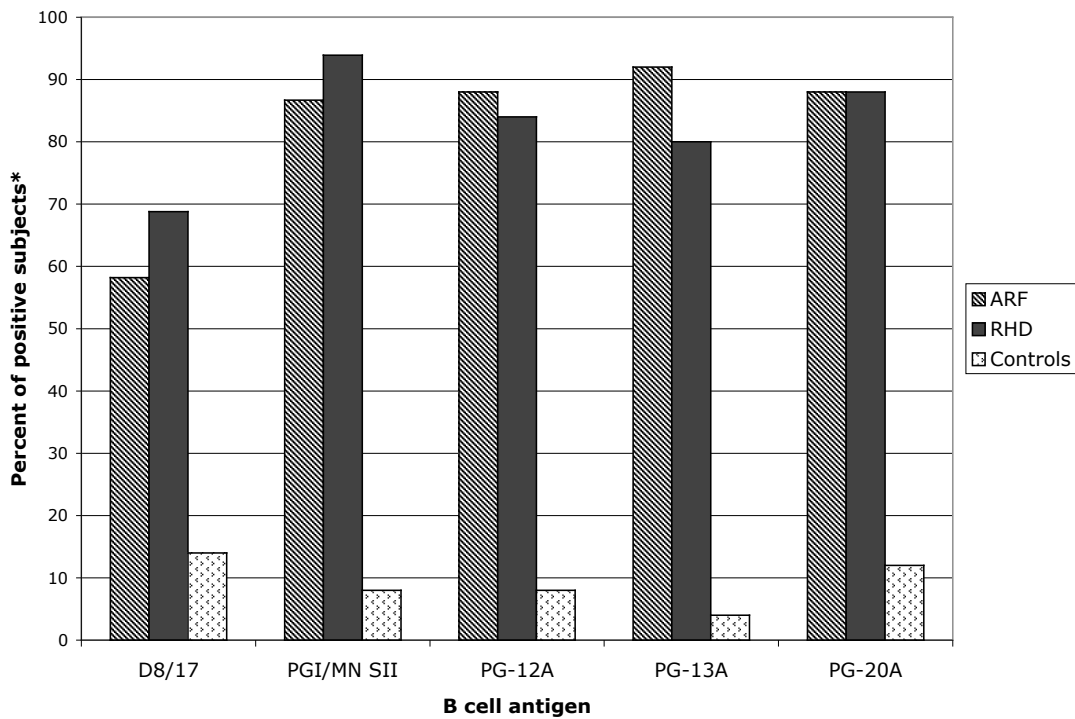


Figure 3 – Comparison of D8/17 with Indian antibodies for identifying RF/RHD

Sources: (Kaur, Kumar et al. 1998; Kumar, Kaur et al. 1998; Kumar, Kaul et al. 2001)

* Positive patients were defined as having >10% of cells staining for each marker.

3.3 Chorea and D8/17

Most of the studies discussed above did not include patients with non-cardiac presentations. One exception documented the D8/17 results of two families that included members with isolated chorea as well as past RF (see table 6) (Feldman,

Zabriskie et al. 1993). The authors suggested that D8/17 might prove to be as equally discriminating in chorea as it had in RHD, but the sample was small and included no controls, making it difficult to draw any conclusions.

Table 6 - Comparison of studies relating D8/17 expression and RF or RHD

| | Positive | Intermediate | Total |
|-------------------------------------|-----------------|---------------------|--------------|
| Patients with chorea | 2 | 0 | 2 |
| Relatives with history of RF | 2 | 0 | 2 |
| Unaffected family members | 2 | 17 | 19 |

Positive defined as >20% cells staining for D8/17.

Intermediate defined as 6.8-20% cells staining for D8/17

Another study looked at D8/17 levels in Sydenham's chorea as well as paediatric patients with tics and healthy controls. Only 46.2% of Sydenham's chorea cases tested positive for D8/17, although how positivity was defined was not stated, nor was the rate of D8/17 positivity in healthy controls provided (Hamilton, Garvey et al. 2003).

3.4 D8/17 expression - inherited trait or a reactive state?

There are a number of possible interpretations of the increased expression of D8/17 in RHD: D8/17 may represent an inherited trait predisposing a person to RF, it may be the result of the repeated streptococcal infections that are believed to cause RF, or it may simply be an epiphenomenon in the pathophysiologic process of RF. The evidence to support the role of D8/17 as an inherited marker of predisposition to RF is

presented in this section, with a detailed discussion of the results presented by Khanna et al. (Khanna, Buskirk et al. 1989).

In the studies described above, D8/17 is expressed as a categorical variable (either positive or negative), rather than a continuous variable, implying that D8/17 expression is a trait inherited in a Mendelian fashion. If this were true, we would expect a modal distribution of D8/17 values, in clearly distinguishable groups. In the study performed by Khanna et al. the distribution of the results for healthy controls was described as skewed: the majority of healthy controls had a low value of D8/17 positive cells (5-7%) but a small number of apparently normal individuals exhibited higher results. By setting a cut-off value to create a categorical result for D8/17, a number of healthy controls and relatives were shown to be “positive” for D8/17. We could speculate that “positive” individuals, whether healthy controls or relatives of the index case, are at risk of developing RF following Group A streptococcal infection, but this remains to be proven. There is a single published account of a person contracting RF subsequent to testing positive for the B cell marker, however this involved the antigen 83S19.23 (the clone of 883) (Regelmann, Talbot et al. 1989).

The strongest evidence that D8/17 expression represents an inherited state comes from work showing significantly different levels of D8/17 in patients, family members and healthy controls, with the values for family members being intermediate between patients and controls (Khanna, Buskirk et al. 1989; Herdy, Zabriskie et al. 1992; Feldman, Zabriskie et al. 1993) (see table 7). Other B cell antigens have also been shown to have intermediate expression in family members (Rich, Gray et al. 1988; Kaur, Kumar et al. 1998; Kumar, Kaur et al. 2000).

Table 7 - Percentage of B cells positive for D8/17 according to category

| | Mean percentage of B cells expressing D8/17 (number of subjects) | | | | |
|----------------|---|--|---------------|---------------|-----------------|
| Study | RHD patients | Siblings | Mother | Father | Controls |
| Khanna | 33.5 (14) | 14.6 (33) | 14.6 (14) | 12.0 (14) | 6.1* (61) |
| Herdy | 38.5 (10) | 21.1 (7) | 12.8 (12) | 12.6 (11) | 4.6 (8) |
| Feldman | 22.1 (4) | First and second degree relatives: 6.8 to 23% | | | Not done |

*Calculated from the data provided

Sources: (Khanna, Buskirk et al. 1989; Herdy, Zabriskie et al. 1992; Feldman, Zabriskie et al. 1993)

Based on the assumption that D8/17 is an inherited trait, Khanna presented the segregation pattern of D8/17 phenotypes in twenty families with an index case of RF. The results suggested an autosomal recessive pattern of inheritance, as 66% of the children of parents negative for D8/17 phenotype had a positive phenotype. This is considerably higher than the expected for an autosomal recessive condition (25% positive siblings of a positive case), but even more unexpected for an autosomal dominant condition where D8/17 negative parents should have no positive children, except in the case of non-paternity. Furthermore, the high proportion of dual D8/17 positive parents (ten of twenty families) is uncommon in autosomal dominant conditions where only a single positive parent is required to produce a positive phenotype in a child.

Khanna also provides evidence that genetics are not the sole determinant of D8/17 expression. A family is described in which there were two sets of identical twins, with

one twin being an index case of RF. In this family the identical twins had no more concordance in D8/17 levels than their non-identical siblings, and the case of RF had a significantly elevated level of expression of D8/17. This implies that the process of RF, or the infections that cause it, may augment D8/17 expression. Yet the authors reject the explanation that D8/17 is simply a non-specific B cell activation factor with the following arguments: the levels of expression of known markers for B cell activation (Tac, 4F2 and CD23) did not correlate with D8/17 levels. Also, the elevation in D8/17 persisted for up to twenty years, which would be unusual in an acute reaction. Finally, D8/17 was not elevated in patients with post-streptococcal glomerulonephritis, or their families.

Whilst maintaining that inheritance of D8/17 was the best explanation to fit the data, Khanna did not rule out the possibility that “the throat infection responsible for RF induces a B cell marker that is unique for this disease and is not seen in streptococcal infections causing nephritis” or that D8/17 expression results “from the expansion of a normal subset of B lymphocytes under genetic control” (Khanna, Buskirk et al. 1989). A longitudinal study looking at premorbid levels of D8/17 and its response to streptococcal infection and the long-term profile has not been conducted. Such a study would quantify the predictive accuracy of D8/17 as an indicator of susceptibility to RF, but also to add evidence to explain its possible role in the aetiology of RHD.

Other influences on D8/17 levels have been documented. Corticosteroids have been reported to affect D8/17 levels (Murphy and Goodman 2002). A non-linear relationship between D8/17 expression and age has been described: 20-50 year olds have been reported as having the highest expression of D8/17 (Ganguly, Anand et al.

1992) and newborns as not showing any D8/17 positive cells (Murphy and Goodman 2002).

In summary, the mode of inheritance of D8/17 is not straightforward. D8/17 is present in all people to some extent, in a distinctive familial pattern: relatives of a person with RHD exhibit an intermediate level of D8/17, between the levels seen in affected persons and healthy controls. Through the analysis of D8/17 phenotypes in rheumatic families it has been proposed that it is inherited in a recessive Mendelian pattern. Additional evidence suggests that the acute disease process further influences the expression of D8/17.

3.5 D8/17 and other disease processes: post-streptococcal reactive arthritis and neuropsychiatric disorders

Post-streptococcal reactive arthritis is associated with elevated levels of D8/17, such that D8/17 expression did not conclusively differentiate between RF and post streptococcal reactive arthritis in one small series (Zemel, Hakonarson et al. 1992). Whether this is because the two conditions share a susceptibility factor or a common pathogenetic pathway is not known. This may be evidence that the two diseases are not distinct, but represent a spectrum of post streptococcal rheumatic disorders. This contrasts with the lack of expression of D8/17 in post-streptococcal glomerulonephritis (Khanna, Buskirk et al. 1989).

Additionally, D8/17 has been shown to correlate with certain neuropsychiatric disorders. The theory that some paediatric neuropsychiatric disorders may be due to autoimmune phenomena secondary to streptococcal infections (known as PANDAS)

was based on observations of similarities with Sydenham's chorea. This led to the investigation of D8/17 antigen positivity in childhood onset obsessive-compulsive disorder, Tourette's syndrome and tic disorders (Murphy, Goodman et al. 1997; Chapman, Visvanathan et al. 1998; Eisen, Leonard et al. 2001; Hoekstra, Bijzet et al. 2001; Murphy, Benson et al. 2001; Hamilton, Garvey et al. 2003), anorexia and autism (Hollander, DelGiudice-Asch et al. 1999; Sokol, Ward et al. 2002) with some significant correlations (Murphy and Goodman 2002). There was a strong *linear* relationship between D8/17 value and compulsion score in a study of autistic subjects, suggesting that D8/17 could just as well be treated as a continuous variable rather than a categorical variable (Hollander, DelGiudice-Asch et al. 1999). However, a prospective study of a group of children with tic disorders and obsessive-compulsive disorder showed no association between indices of recent streptococcal infection and neuropsychiatric symptoms – calling into question the whole concept of paediatric autoimmune post-streptococcal neuropsychiatric disorders (Luo, Leckman et al. 2004).

3.6 D8/17 – characterisation, function, role in disease process

Soon after it was discovered, D8/17 was fully characterised as a helical coil-coiled cell surface protein. It did not match any known protein; in particular, it did not correspond to any known HLA (Feldman, Zabriskie et al. 1993; Carreno-Manjarrez, Viteri-Jackson et al. 1998). The antibody to D8/17 has been shown to cross-react with cardiac muscle, skeletal muscle, smooth muscle cells of blood vessels and recombinant M6 streptococcal protein. Adsorption testing showed that B cells from a RHD patient inhibited the binding of the D8/17 antibody to cardiac muscle. It was suggested on this basis that the D8/17 antigen may act as a streptococcal binding site

on the B cells, and consequently become up-regulated following an infection, with B cells acting as antigen presenting cells and influencing T cell specific cytotoxicity to heart and brain cells (Kemeny, Husby et al. 1994). How this relates to the clinical manifestations of RF is not known.

A different pathogenetic link between D8/17 and RHD was suggested through the demonstration of a correlation between Na/H exchange and D8/17. This idea came from the chance observation that, like the plasma membrane Na/H exchanger, D8/17 expression varied by up to a dozen times in the normal population. This work suggested that either the D8/17 antigen and the Na/H exchanger were the same protein, or that the two were genetically linked. This study further proposed how activation of this membrane protein could be the cause of both the lymphocyte activation in response to streptococcal infection and the cardiac muscle degradation (Koren, Koldanov et al. 1996).

3.7 Conclusion

The identification of B cell antigen D8/17 is a promising development in the search for a marker of genetic susceptibility for RF. Many international studies have found that D8/17 distinguished accurately between patients with RHD and healthy controls, with the exception of work carried out in Northern India. The similarity in the results obtained with D8/17 and the earlier identified marker 883 raises the possibility that these are the same antigen. Evidence that D8/17 is an inherited trait has been presented alongside results that indicate that D8/17 expression also has a reactive component.

4 Diagnostic difficulties with rheumatic fever

4.1 Introduction

RF is diagnosed using a set of clinical and laboratory criteria developed empirically but inadequately evaluated. However there is evidence of under-diagnosis in populations with high incidence, with unfortunate consequences, while over-diagnosis in affluent countries, although less disastrous, is also undesirable. A diagnostic tool with greater accuracy than the Jones criteria is required.

4.2 Limitations of the Jones criteria for the diagnosis of rheumatic fever

The features of arthritis, heart disease, chorea, subcutaneous nodules and erythema marginatum were linked together as “the rheumatic state” by Walter Cheadle in 1889, whose wife and son suffered from the disease (Cheadle 1889; DiSciascio and Taranta 1980). The Jones criteria were published in 1944 as a tool for the diagnosis of RF (Jones 1944). They have been modified (Rutstein, Bauer et al. 1955), revised (Stollerman, Markowitz et al. 1965), edited (Shulman, Kaplan et al. 1984), updated (Dajani, Ayoub et al. 1992) and reaffirmed (Ferrieri and Group 2002) since their initial development. The current criteria are summarised in table 8. The diagnosis of RF requires two major manifestations, or one major and two minor manifestations as well as evidence of a recent Group A streptococcal infection. The criteria are applied less strictly in the diagnosis of recurrent RF where one major or two minor criteria may be sufficient to diagnose RF in a patient with established RHD. As chorea can present after a considerable latent period, recent Group A streptococcal infection does

not need to be demonstrated if there is no other reasonable diagnosis. Whilst they remain the working tool for most clinicians to diagnose RF, the criteria are only a guideline - they “represent recommendations to supplement practitioners in the exercise of their clinical diagnosis and are not a substitute for clinical judgement” (Dajani, Ayoub et al. 1992).

Table 8 - The Jones criteria

| Major manifestations | Minor manifestations |
|---|-----------------------------|
| Carditis | Arthralgia |
| Polyarthritits | Fever |
| Chorea | Raised ESR or CRP |
| Erythema marginatum | Prolonged PR interval |
| Subcutaneous nodules | |
| PLUS | |
| SUPPORTING EVIDENCE OF ANTECEDENT GROUP A STREPTOCOCCAL INFECTION: | |
| Positive throat culture or rapid streptococcal antigen test | |
| Elevated or rising streptococcal antibody titre | |

Source: (Dajani, Ayoub et al. 1992)

Despite the absence of a gold standard test with which to compare the Jones criteria, some attempts have been made to evaluate their accuracy. In one study only 58% of patients on a RHD registry list and 72% of patients hospitalised with the diagnosis of RF in India satisfied the Jones criteria (Padmavati and Gupta 1988). Likewise, only 60% of RF cases reported in Minnesota in 1978 fulfilled the Jones criteria (DiSciascio and Taranta 1980). A review of published South African data concluded that the sensitivity of the Jones criteria was between 51 and 77% and the specificity between

84 and 99% (Ralph, 2002). This suggests either that clinicians are making incorrect diagnoses, or have used their clinical judgement to make diagnoses that do not satisfy the Jones criteria. In the absence of a “tissue diagnosis” (i.e. surgical or post-mortem specimen) the clinician and researcher must commit to prolonged follow-up and clinical review of all patients in whom the possibility of diagnosis of RF is raised.

The positive predictive power of a test depends not only on the accuracy of the test, but also on the prevalence or incidence of the disease being studied in the background population. In industrialised countries RF is now a rare diagnosis, and the clinical presentation may have altered since the time the criteria were published (Williamson, Bowness et al. 2000). In developing countries and in specific populations within developed countries where the prevalence of RF is high, the condition is being under-diagnosed. Currently in the Top End of Australia’s Northern Territory, where the prevalence of RHD is amongst the highest in the world, nearly half of all cases are diagnosed with established chronic rheumatic valvular disease, suggesting that the earlier cases of RF in these people were missed (Carapetis, Wolff et al. 1996). One of the reasons for poor case ascertainment for RF may be that the Jones criteria may be insufficiently sensitive for this population.

In keeping with this observation, those studying RF in developing countries and indigenous populations within developed countries have argued for specific modifications of the criteria based on regional patterns to increase the sensitivity of the criteria (Markowitz 1988). For example, Carapetis suggested upgrading monoarthritis to a major manifestation in the Northern Territory (Carapetis and Currie 2001). Similar arguments have been put forward in India (Bhattacharya and Tandon

1986). Others argue that there is little real difference in the presenting features of RF around the world, that apparent differences are the result of errors inherent in retrospective studies (Sanyal, Thapar et al. 1974). In particular, the American establishment are reluctant to accept any modifications of the Jones criteria that may reduce the specificity of the diagnostic criteria, but accept that clinicians in high incidence areas may need to modify the criteria according to their local situation (Ferrieri and Group 2002).

There are many possible differential diagnoses for RF, listed in table 9. A particular need for disease specificity of the Jones criteria is highlighted by the recent description of post-streptococcal reactive arthritis without carditis. It is still debated to what extent this may be a separate entity from RF and whether prophylaxis should be recommended to prevent carditis (Jansen, Janssen et al. 1999). Even experienced clinicians are having difficulty achieving agreement in the diagnosis and management of this entity (Birdi, Hosking et al. 2001). Thus Shulman wrote in 1999, that “unless a highly sensitive and specific diagnostic test for RF that obviates reliance on clinical criteria is developed, it is highly probable that additional changes in the Jones criteria will be considered appropriate in future decades” (Shulman 1999).

Table 9 - Common differential diagnoses of acute rheumatic fever

| Category | Diagnosis |
|--|--|
| Infections | Arbovirus infections (in Australia – Ross River Virus and Barmah Forest Virus) |
| | Septic arthritis |
| | Gonococcal arthritis |
| | Osteomyelitis |
| | Viral carditis/ pericarditis |
| | Bacterial endocarditis |
| | Uncomplicated streptococcal pharyngitis |
| | Bacteraemia |
| | Tuberculous pericarditis |
| | Connective tissue disorders |
| Reactive arthritis, including post-streptococcal | |
| Rheumatoid arthritis | |
| Cardiac disease | Mitral valve prolapse |
| | Congenital heart disease |
| | Atrial fibrillation in established RHD |
| Neurologic | Epilepsy |
| | Wilson’s disease |
| | Chorea of pregnancy |

4.3 Towards a diagnostic test for acute rheumatic fever

There have been attempts to develop a diagnostic laboratory test for RF. One proposal came from India where the injection of autologous blood caused subcutaneous nodules in patients with RF but not in healthy controls. Three criteria - subcutaneous

nodules following autologous blood injection, elevated IgA and elevated C3 - in combination produced a composite laboratory test with a sensitivity of 84% and specificity of 100% for RF when compared with the Jones criteria (Bhattacharya, Reddy et al. 1987).

In Australia, Norton described a serological test based on reactivity to a peptide sequence derived from the streptococcal M24 protein that was discriminatory in RF and RHD cases and controls amongst Aboriginal people, but not in non-Aboriginals (Norton, Heuzenroeder et al. 1996). The likely explanation for this is that the non-Aboriginals were tested many years after their last episode of RF attack and didn't maintain a high antibody level. The calculated sensitivity for RF of 100% and specificity of 91% amongst Aboriginals suggests that this may indeed be a useful diagnostic test in this population and deserves further prospective evaluation.

A third candidate for a diagnostic test for RF is the B cell marker D8/17. According to current conceptualisation, D8/17 represents an inherited susceptibility trait, which may be induced or up-regulated in the acute phase of the disease after exposure to Group A streptococci. As D8/17 elevation indicates *susceptibility* to RHD, it cannot be used in isolation to diagnose the disease. However, the suggestion that there may be a significant further elevation during the acute phase suggests the possibility of identifying a threshold level of D8/17 diagnostic for RF. Of concern, however, is the observation that D8/17 was least accurate in identifying RHD in the ten to nineteen year age group in India, a period of high risk for RF (Ganguly, Anand et al. 1992). Furthermore, the use of D8/17 to identify cases with chorea has not been sufficiently

investigated. Given the high sensitivity of the test, a low or “negative” D8/17 in a low-risk population should have good negative predictive value.

4.4 Public health implications of a screening and diagnostic test for acute rheumatic fever

To date, few attempts at reducing the incidence of first attacks of RF have been successful or sustainable (Hanna and O'Rourke 1993; Bach, Chalons et al. 1996). Strategies used have consisted of increasing community and health professional awareness of the disease and strict application of the rules of clinical testing and treatment of streptococcal pharyngitis. An overall goal remains the improvement in living conditions, which has been linked to the reduction in RF incidence in the developed world (Quinn 1989). However, a test for susceptibility for RF extends the possibilities of primary prevention of this disease: people identified as susceptible to RF could be targeted in a prevention programme that might include surveillance for streptococcal infections, prophylactic anti-streptococcal antibiotics and eventually, a streptococcal vaccine. As the peak onset of disease, disability and excess mortality is in children and young adults, the benefits of primary prevention in terms of productive-life-years is great.

King analysed the cost-benefit ratio of a test for susceptibility to RF for the US and found that there would be an overall economic advantage if such a test existed (King, Fischler et al. 2002). Amongst the Aboriginal people of Australia the incidence of RF is high and thus the likely benefits of prevention are even greater than in the US.

Location specific factors such as geographical isolation, cultural and language difference could potentially increase the cost of service provision, thus a local cost-

benefit analysis is necessary to confirm the hypothesis that a test for susceptibility to RF would yield economic savings in Australia.

A diagnostic test for RF would have varied applications depending on the disease prevalence and the socio-economic setting. A highly sensitive but poorly specific test has a good negative predictive value. This would be especially desirable in remote or under-serviced locations such as in developing countries where clinical resources may be limited, thereby acting as a screening test in order to determine which patients are referred for definitive investigation. A highly specific test is more useful for confirming a suspected diagnosis in low prevalence areas or within tertiary referral centres where an accurate result prevents years of unnecessary treatments, inconvenience to patients and cost to health providers.

4.5 Conclusion

The Jones criteria, whilst assisting clinicians over half a century in making the diagnosis of RF have a number of limitations. In populations with a high prevalence of RF the Jones criteria may lead to under-diagnosis, due to their lack of sensitivity. Laboratory tests with greater accuracy for RF have been proposed, including the B cell marker D8/17. This research aims to answer the questions about the usefulness of D8/17 as a marker for susceptibility for RF in aboriginal Australians, and its potential as a diagnostic test for RF.

5 The setting of the study

5.1 Social geography

The focus of this study was the problem of the high prevalence of RHD amongst the Aboriginal people of the Top End of the Northern Territory. The Top End is a culturally and geographically distinct region of Australia. It encompasses the tropical northern segment of the Northern Territory, and excludes the desert region in the south. The Top End consists of four statistical regions: Darwin, Darwin region, East Arnhem region and Katherine region. Some basic demographics of the area according to the 2001 census are summarised in table 10. The poor health of the Aboriginal people of the NT can be surmised from their greatly reduced life expectancy – see table 11.

Table 10 - Top End basic demographics

| | Darwin | Darwin region | East Arnhem | Katherine |
|------------------------------------|---------------|----------------------|--------------------|------------------|
| Area (Sq km) | 10 356 | 134 466 | 40 376 | 337 363 |
| Population (no.) | 108 715 | 12 816 | 13 944 | 17 431 |
| Indigenous population (no.) | 10 148 | 8 864 | 7 940 | 7 521 |
| Median age (y) | 31.2 | 23.9 | 25.9 | 27.3 |

Source: (Anonymous, 2004c)

Table 11 - Median age at death of Australians in 2002 (years)

| Population | NT Indigenous | NT | Australian |
|-------------------|----------------------|-----------|-------------------|
| Male | 47.1 | 68.1 | 76.2 |
| Female | 50.0 | 73.6 | 82.2 |

Source: (Anonymous, 2004c)

5.2 Epidemiology of acute rheumatic fever and rheumatic heart disease in the Top End

Comprehensive epidemiological data regarding RF and RHD in the Top End since 1989 are available. The incidence of RF and prevalence of RHD are amongst the highest in the world (figures 4 and 5). The reasons given for this extraordinarily high incidence of disease include poverty, overcrowding, a high rate of streptococcal carriage (skin, but not throat carriage, paradoxically) and a lack of access to medical services (Currie and Brewster 2002). In 2003, the reported number of cases of RF in the Top End, Barkly Districts and Alice Springs districts fell considerably, from 83 to 43 cases (Anonymous 2004; Anonymous 2004). Whether this is a sustained trend remains to be seen. In urban Australia RF is rare; RHD is most often seen in migrants from high prevalence areas, or in elderly people who developed the disease prior to the 1960s.

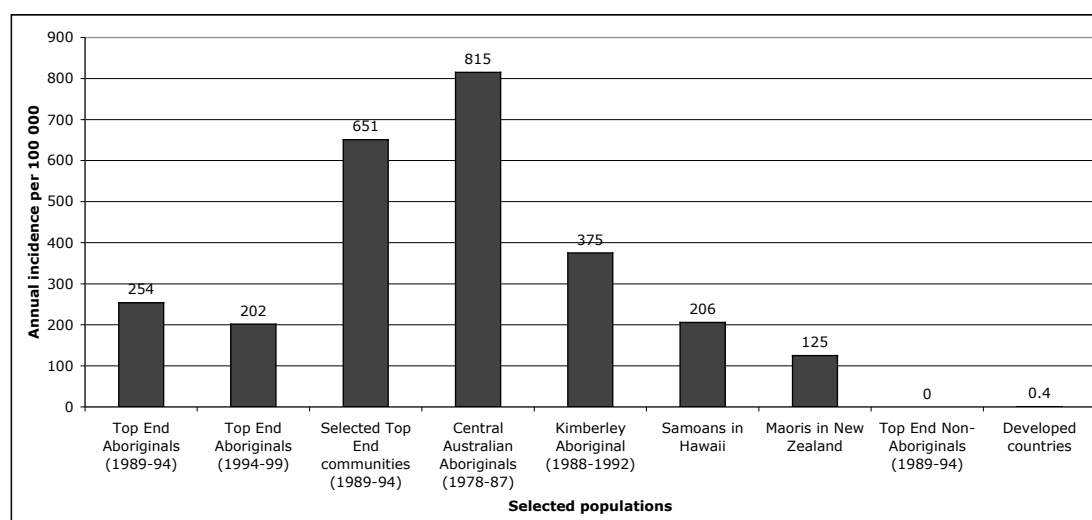


Figure 4 - Annual incidence of RF in selected populations

Sources: (Carapetis, Wolff et al. 1996; Richmond and Harris 1998; Edmond, Noonan et al. 2001)

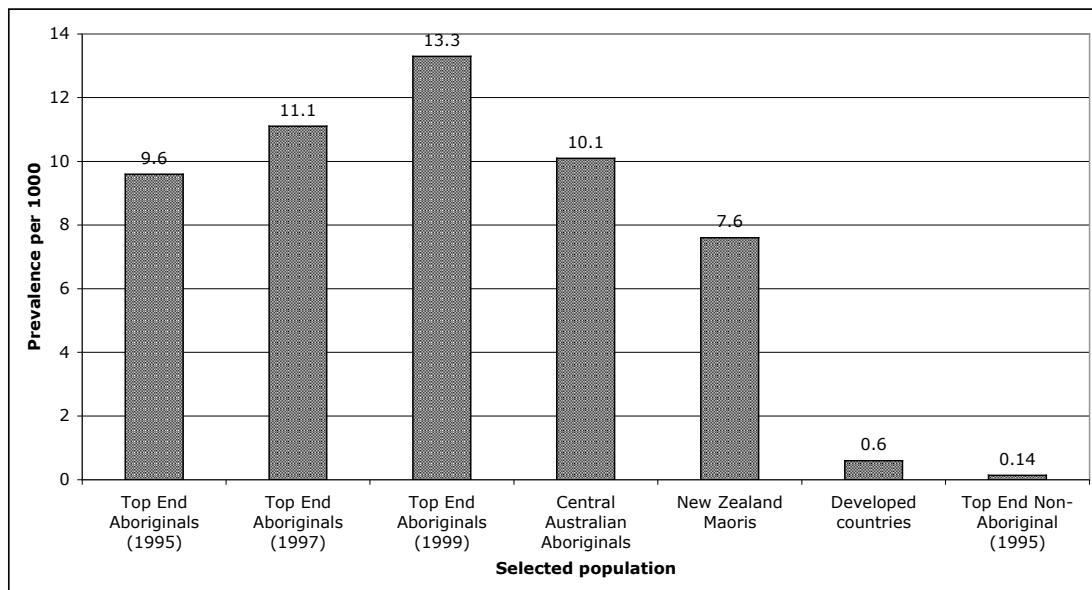


Figure 5 - Point prevalence of RHD in selected populations

Sources: (Carapetis, Wolff et al. 1996) and (Edmond, Noonan et al. 2001)

The mortality due to RF and RHD for Aboriginal people in the Northern Territory at the end of the twentieth century was 26 times the mortality in the non-indigenous population, four times the published rate in some developing countries, and greater than the crude death rates in industrialised countries a century ago (Carapetis and Currie 1999). A key observation is that a large proportion of this excess disease is preventable; much of the RF recorded is recurrent RF, preventable through prophylactic benzathine penicillin treatment. (For a more detailed discussion see part 2 of this thesis.)

6 Methods

6.1 Overview of the research design

This study was designed to answer one main question: whether the B cell antigen D8/17 is a susceptibility marker for RF in Australians. A secondary aim was to review some aspects of the methodology of the D8/17 assay in order to make the test more applicable in a setting where many of the patients live remotely and there are delays in receiving blood samples in the laboratory, as well as lack of trained staff to process the assay. For an overview of the research design, see figure 6.

In order to evaluate D8/17 as marker of susceptibility for RF, patients with established RHD or evidence of past RF, relatives of RF/RHD patients and healthy controls were recruited. This occurred in three centres: an Aboriginal community in the Northern Territory, a Northern Territory tertiary referral hospital and a major teaching hospital in Victoria. The primary endpoint was the evaluation of D8/17 levels in all three groups (discussed below in detail).

In addition to this we evaluated two methods of performing the D8/17 assay. In a subset of subjects we compared the D8/17 assay on whole blood with an assay performed on purified blood mononuclear cells (PBMCs) that had been frozen and stored. We also investigated the effect of time-delay on the staining of cells for D8/17: a subset of specimens were stained on site in the remote community, and these results were compared with the conventional process of staining the blood upon arrival in the laboratory in the tertiary referral hospital.

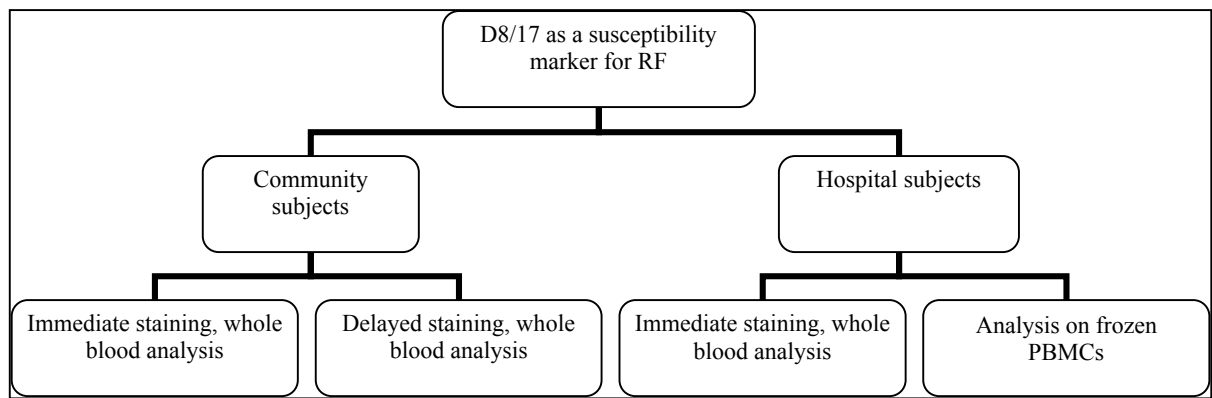


Figure 6- Research design

6.2 Community selection and subject recruitment

Galiwin'ku community on Elcho Island was selected as the community for study as it has a high prevalence of RHD with more than 30 people being treated regularly through the health centre. Support for the project was obtained from the Galiwin'ku community council and Ngalkanbuy health centre. Ethics approval was obtained from the human research ethics committee of Territory Health services and Menzies School of Health Research. General information relating to the disease as well as the study was translated into Djambarrpuyngu, the local language. This was done in conjunction with *Yalu Marngithinyaraw*, a locally constituted group that mediates between non-local researchers and the community.

Patients with RHD were identified from a register held at the health centre. Health staff approached these patients and invited them to participate in the study. Unrelated, healthy controls were enrolled through the health centre and also during the discussions with the rheumatic families in the field. For this study, relatives included parents, grandparents, siblings, aunts and uncles and first cousins (those who shared a

single grandparent). Making clear distinctions between first-degree relatives (parents, siblings and children) and second-degree relatives was difficult, not only because of language differences, but also due to differences in kinship structure. The research team (a doctor, nurse and Aboriginal health worker) explained the purpose and process of the study in the local language with the aid of the developed materials to all the interested parties.

Hospital based patients were identified via the hospital database, or referred by their treating physician. Control subjects were selected from willing in-patients to match the patients' age, sex and ethnicity.

6.3 Collection of data, specimens and transportation

After consent was obtained, information was sought to ascertain whether the participant had a history of RF or was related to a patient with a history of RF. This was difficult to verify in some cases as the medical records of long deceased people had been destroyed. Documentation of Jones criteria was often incomplete, so evidence of established rheumatic valvular disease on echocardiogram was often required to identify an index case. In cases that had chorea as the presenting symptom, alternative diagnoses had to have been excluded. The absence of a history of RF/RHD in relatives and controls was documented by interview with the individual, review of medical notes and auscultation.

Blood was collected in lithium heparin vacutainers for the D8/17 B cell antigen assay. Transportation via light aircraft involved delays of up to 8 hours as well as temperatures in excess of 30 degrees Celsius on occasion. Specimens were discarded

when they showed evidence of heat or time delay artefact, with distortion of the white cells. In addition, a number of specimens were stained on site for comparison with delayed staining in the laboratory.

6.4 Preparation of purified blood mononuclear cells

Purified blood mononuclear cells (PBMCs) were isolated using the Ficoll extraction method. Whole blood in lithium heparin vacutainers was diluted with phosphate buffered solution (PBS) was poured over Ficoll-Paque Plus (Pharmacia Biotech), then centrifuged. The buffy layer was harvested and washed with PBS. The resulting PBMCs were resuspended in foetal calf serum with 10% dimethylsulfoxide (DMSO), and then slowly frozen to minus 80 degrees centigrade.

6.5 D8/17 assay

All assays were performed within one hour of being received in the laboratory. Laboratory staff were blinded to the clinical category of the sample. To three hundred microlitres of whole blood, either 20 microlitres of mouse anti-D8/17 monoclonal antibody (kindly provided by Prof J. Zabriskie, Rockefeller University, New York, USA) or the isotype control of 5 microlitres of mouse IgM was added. After incubation for 40 minutes at 4°C the samples were washed with phosphate buffered saline (PBS). Five microlitres of goat anti-mouse antibody labelled with fluorescein isothiocyanate (FITC) and 5 microlitres of anti CD19 labelled with phycoerythrin (PE) were added. After incubation at room temperature in the dark for 20 minutes red cells were lysed with an ammonium chloride lysis solution for 20 minutes. The samples were centrifuged, washed and resuspended in PBS (see figure 7).

These samples were analysed by flow cytometry. Lymphocytes were gated according to their forward and side scatter profile. To eliminate background fluorescence, values were set for the quadrants by defining the control samples (no D8/17 antibody) as 99% negative. The fluorescence was measured and the percentage of D8/17 positive cells of all CD19 positive cells was calculated using Flo Jo (Flow Jo, USA) software. The flow cytometry method used in this research has been formally evaluated against the standard immunofluorescence microscopy (Chapman, Visvanathan et al. 1998).

Frozen PBMCs were thawed and incubated with anti D8/17, mouse IgM, goat anti-mouse and anti CD19 antibodies as described above and analysed by flow cytometry.

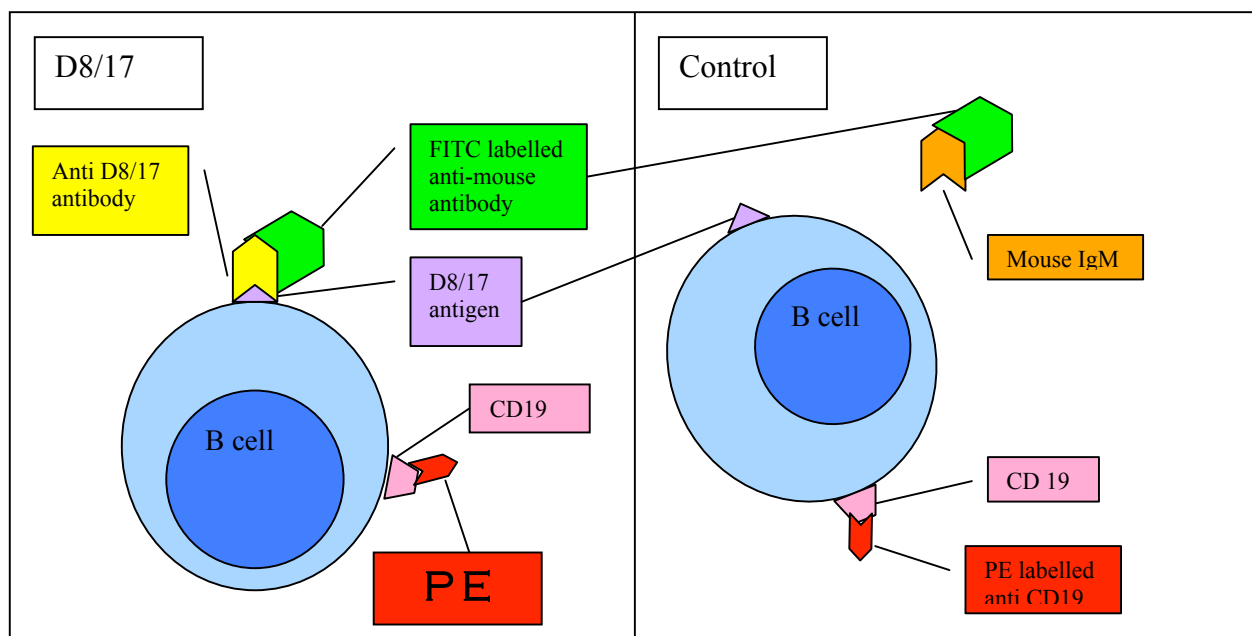


Figure 7 – Method of staining for D8/17 assay

6.6 Statistical analysis

The required sample size was calculated using the results for RHD patients and controls published by Khanna as outlined in table 7 (Khanna, Buskirk et al. 1989). Only two cases and controls were required to identify a significant difference between the percentage of D8/17 positive B cells with a power of 90% and alpha of 0.05. However, to detect a statistically significant difference between family members and controls, we would need 35 family members and 18 controls, using a ratio of 2 to 1. Consequently we aimed to enrol 20 patients, 40 family members and 20 healthy controls.

The mean and standard deviation of percentage of B cells reactive to the D8/17 antibody was calculated for each group of subjects. ANOVA was performed to assess the significance of the difference between the means. The differences between the means of each of the categories and the 95% confidence interval of these differences were calculated. The relationship between first degree and second-degree relatives were analysed using the student t-test.

A receiver operator characteristics (ROC) curve was constructed using the results from the positive cases (RHD and RF cases) and controls. The sensitivity and specificity of D8/17 as an indicator of disease (acute and past RF) were calculated for each defined cut-off point. The percentages of accurate results and the likelihood ratios given positive and negative results were derived from these.

The correlation coefficient comparing the results on the PBMCs and whole blood was calculated, as was the correlation between the D8/17 results for early and late staining of the specimens.

Statistical calculations were performed using Stata 7 statistical package (Stata Corporation, College station, Texas, USA). Graphs were produced using Excel spreadsheets (Office OS X for Macintosh, Microsoft Corporation, Seattle, USA).

7 Results

7.1 Demographics

The basic demographics of the patients enrolled in the study are described in table 12.

Table 12 - Research subjects

| Demographic | Subgroup | Category | | | | | Total |
|----------------------------|----------------|--------------|-------------------|---------------------------------------|---------------------------------------|--------------|-------|
| | | RF | RHD or past RF | 1 st degree relative | 2 nd degree relative | Control | |
| Ethnicity | Aboriginal | 3 | 34 | 7 | 8 | 35 | 87 |
| | Non-Aboriginal | 0 | 4 | 5 | 0 | 10 | 19 |
| Sex | Male | 1 | 16 | 3 | 0 | 22 | 42 |
| | Female | 2 | 22 | 9 | 8 | 23 | 64 |
| Elcho cohort | - | 1 | 14 | 7 | 6 | 12 | 40 |
| Mean age, (range) years | | 10 (8-14) | 36 (19-52) | 37 (11-58) | 34 (19-68) | 39 (4-63) | - |
| Total | | 3 | 38 | 12 | 8 | 45 | 106 |

7.2 Results of the D8/17 assay

The results of the D8/17 assay according to demographic group are presented in table 13 and figure 8.

Table 13 – Mean (SD) percentage of B cells positive for D8/17 according to category

| | RF | RHD or past RF | Relative | Control | p value ANOVA |
|---|-------------|---------------------------|-----------------|----------------|--------------------------|
| All subjects | 83.7 (10.1) | 39.3 (11.8) | 20.2 (7.2) | 11.6 (7.3) | <0.0001 |
| Aboriginal subjects | 83.7 (10.1) | 38.3 (11.9) | 19.0 (7.2) | 11.8 (7.4) | <0.0001 |
| Elcho Aboriginal subjects | 78.1 | 39.2 (7.7) | 20.2 (6.9) | 12.7 (5.1) | <0.0001 |
| All subjects (first degree relatives only) | 83.7 (10.1) | 39.3 (11.8) | 22.5 (5.2) | 11.6 (7.3) | <0.0001 |

The mean D8/17 level for first-degree relatives was 22.5% (SD 5.2), compared with 16.8% (SD 8.7) for second-degree relatives. The difference between these two groups approached significance (t test, $p=0.08$), thus only first-degree relatives were included in the remainder of the analysis.

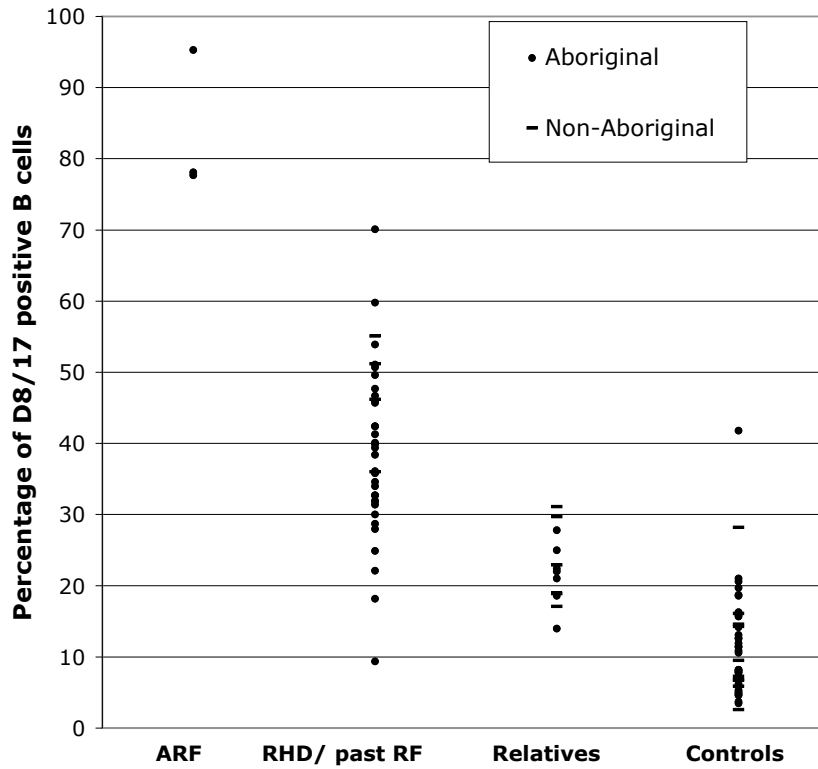


Figure 8 - % B cells staining positive analysed by clinical category

The difference of means was calculated for each clinically important comparison (see Table 14). Significant and sizeable differences were found between all groups.

Table 14 - Difference of the mean of B cells positive for D8/17, by category

| Categories compared | Difference of the mean of B cells positive for D8/17 (95% Confidence intervals) |
|---------------------------|---|
| RF to controls | 72.0 (63.1-81.0) |
| RF to RHD/ past RF | 44.4 (30.2-58.7) |
| RHD/ past RF to controls | 27.6 (23.4-31.8) |
| RHD/ past RF to relatives | 16.7 (9.6-23.8) |
| Relatives to controls | 10.9 (6.4-15.4) |

Levels of D8/17 according to clinical presentation could not be accurately assessed as only two patients had presented with chorea and only one with neither chorea nor carditis. However, it can be noted that the D8/17 results for these patients were within two standard deviations of the mean of all patients with RHD (30.0, 53.9 and 35.9% of B cells respectively).

7.3 Receiver operator curve analysis of D8/17 as a test for current or past rheumatic fever

In the receiver operator curve (ROC) analysis, patients with either acute RF or a history of past RF or RHD were defined as positive cases and were compared with healthy controls. The results from the ROC analysis in graph and table form are presented in figure 9 and table 15. Based on this analysis, a cut-off of greater than or equal to 22.1% of B cells positive for D8/17 yielded the greatest percentage of accurate results (95.4%). The likelihood ratio of a positive test at this cut-off is 21.4, thus the patient's likelihood of having the disease given a positive test result is 21.4 times whatever the pre-test probability is estimated to be. If a negative result is obtained, the pre-test probability is multiplied by 0.05. Using this cut-off 50% of first-degree relatives are 'positive' for D8/17.

The values for the cut-offs used in the literature were also calculated. A cut-off of 11.9% B cells yielded an accuracy of 76.7%, whilst the mean for controls plus one standard deviation (18.7%) yielded an accuracy of 90.7%.

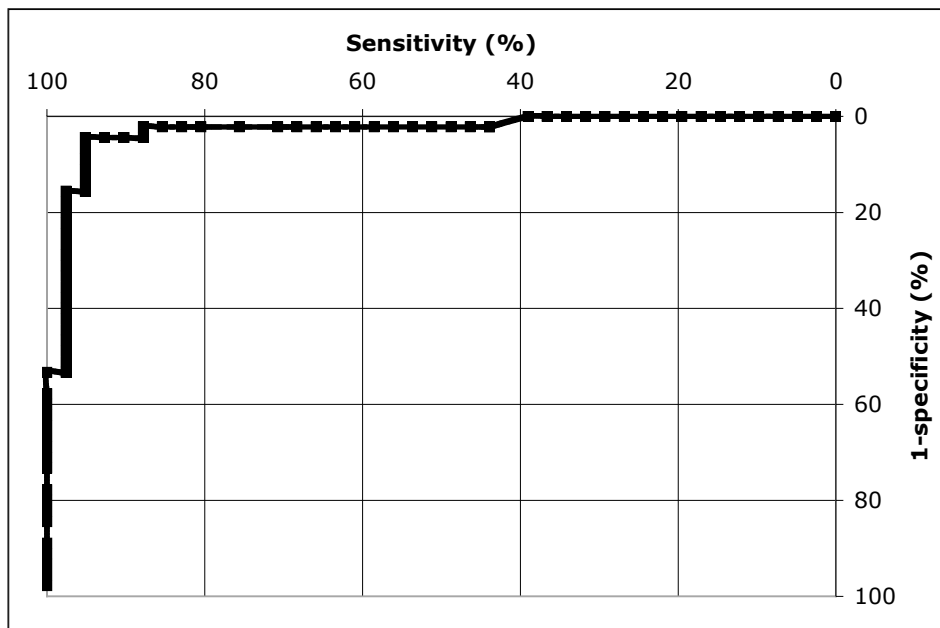


Figure 9 - ROC curve for D8/17 as a test for RF or past RF

Area under ROC curve =0.970

Table 15 - Sensitivity and specificity of D8/17 for past or current RF at various cut points

| Cut point (% B cells expressing D8/17) | Sensitivity (%) | Specificity (%) | Correctly classified (%) | Likelihood ratio (positive) | Likelihood ratio (negative) |
|---|----------------------------|----------------------------|---|--|--|
| 3.5 | 100 | 2.22 | 48.8 | 1.02 | 0.00 |
| 8 | 100 | 40.0 | 68.6 | 1.67 | 0.00 |
| 11.9* | 97.6 | 57.8 | 76.7 | 2.31 | 0.04 |
| 18.7† | 95.1 | 86.7 | 90.7 | 7.13 | 0.06 |
| 21 | 95.1 | 93.3 | 94.1 | 14.26 | 0.05 |
| 22.1 | 95.1 | 95.6 | 95.4 | 21.4 | 0.05 |
| 24.9 | 92.7 | 95.6 | 94.2 | 20.85 | 0.08 |
| 30 | 85.4 | 97.8 | 91.9 | 38.4 | 0.15 |
| 36 | 61.0 | 97.8 | 80.2 | 27.4 | 0.40 |
| 40.1 | 48.8 | 97.8 | 74.4 | 21.95 | 0.52 |

* Cut-off point used by Khanna et al. (Khanna, Buskirk et al. 1989)

† Approximately equal to the mean for controls plus one standard deviation

7.4 Technical aspects of the D8/17 assay

There was good correlation found between the results for the D8/17 analysis on blood stained immediately compared with blood stained with a delay of half a day (mean 0.5 days, SD 0.2) on 38 sample pairs ($R=0.94$), see figure 10. There was poor correlation between the results of the D8/17 assay performed on fresh whole blood compared with frozen PBMCs, analysing 19 sample pairs ($R=0.32$), see figure 11.

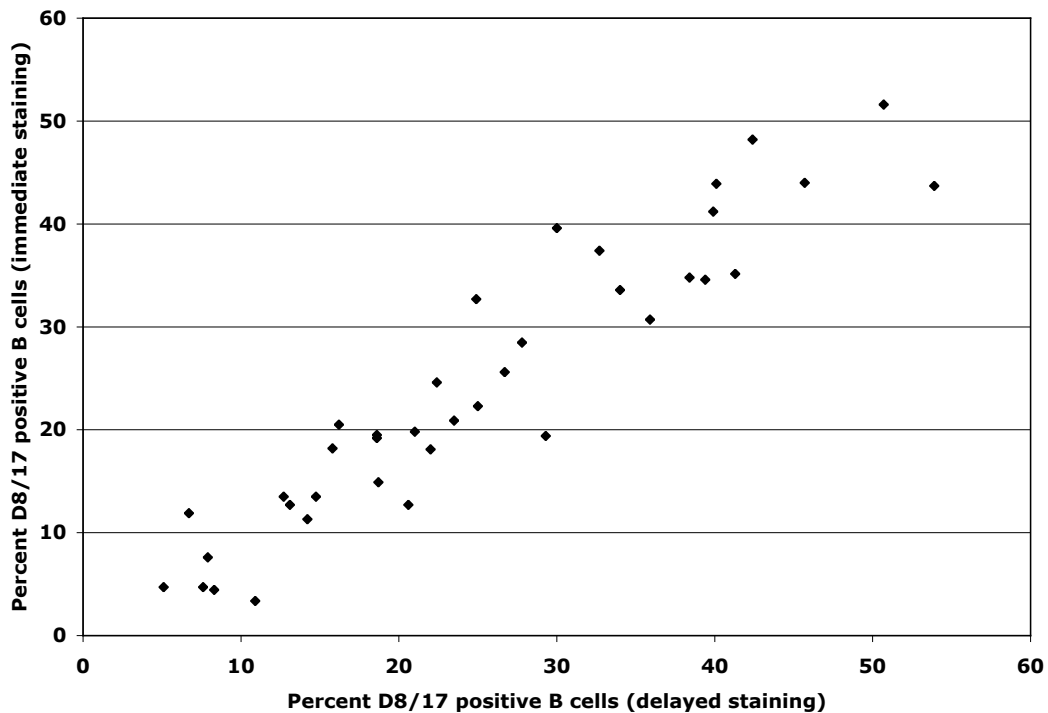


Figure 10 - Comparison of immediate versus delayed staining for D8/17 analysis

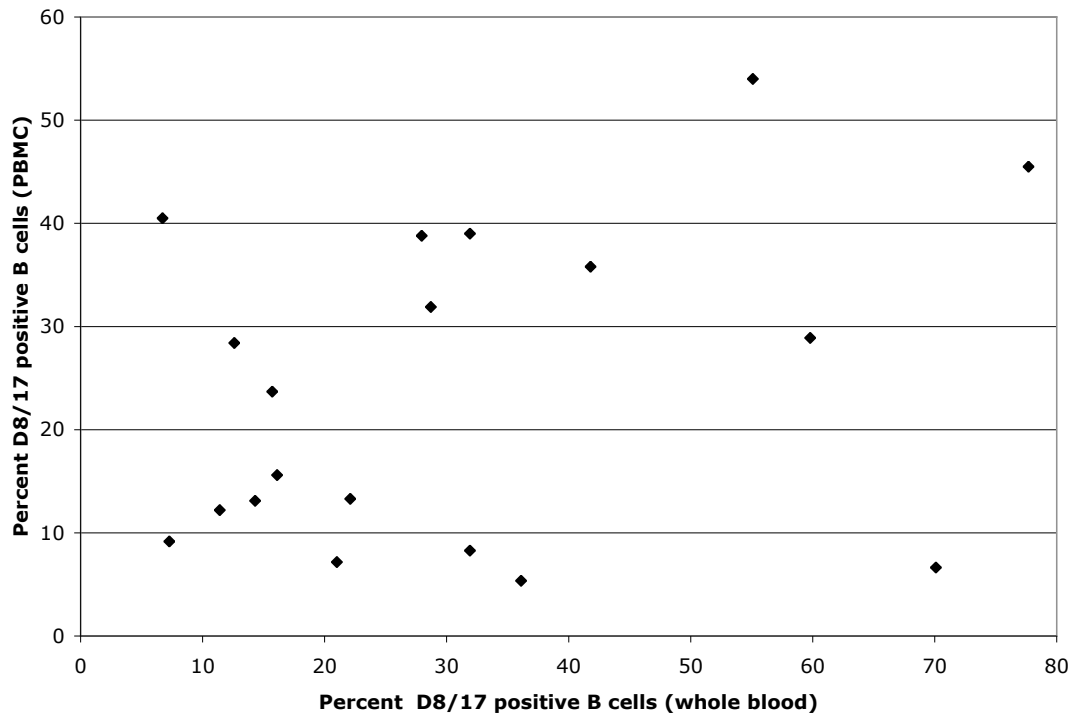


Figure 11 - Comparison of D8/17 analysis on fresh whole blood versus frozen PBMCs

8 Discussion

8.1 Significance of the results

The B cell antigen known as D8/17 is a sensitive and specific marker of past RF in Australians. The results obtained in this study support the findings of researchers in the US, Mexico, Russia, Chile and Israel (Khanna, Buskirk et al. 1989; Gibofsky, Khanna et al. 1991; Herdy, Zabriskie et al. 1992; Feldman, Zabriskie et al. 1993; Harel, Zeharia et al. 2002). This adds to the growing evidence that D8/17 may be a universal marker of susceptibility to RF in disparate populations. The B cell antigen identified by serum 883 was also an accurate predictor of past RF (Patarroyo, RJ et al. 1979; Gray, Regelman et al. 1984; Yeadon, Buskirk et al. 1984; Williams, Raizada et al. 1985; Regelman, Talbot et al. 1989). As serum 883 has been exhausted it will not be possible to determine the relationship between them. Anti-D8/17 and 883 may identify the same protein.

India remains the exception to the rule: fewer patients in northern India are positive for D8/17 compared with controls (Taneja, Mehra et al. 1989; Ganguly, Anand et al. 1992; Kaur, Kumar et al. 1998). There are a number of possible explanations for this discrepancy. Firstly, the gene responsible for susceptibility to RHD may be in linkage disequilibrium with D8/17, and the closeness of the association may vary between populations of different origins. However, the diversity of populations in which D8/17 is an informative test makes this implausible; there is little evidence that would suggest that Australian Aborigines have more in common with the people of the USA, Mexico, Russia, Chile and Israel, than do Indians. Secondly, as genetic analysis does not consistently point to a single gene responsible for RHD there is potential for more

than one gene to be identified. Thirdly, differences in technique (for example immuno-fluorescence versus flow cytometry) or experimental error may account for some of the discrepancy.

In our study the absolute values of the percentage of B cells positive for D8/17 in patients with a past history of RF or RHD, first-degree relatives and healthy controls replicates the results published by Khanna and increases their reliability and generalisability (Khanna, Buskirk et al. 1989). A cut-off point of 22.1% cells positive for D8/17 was the most accurate break-point for a past or current episode of RF with 95% sensitivity and 96% specificity. If we were to use the cut-off used by other published authors (either 11.8%, or the mean plus one standard deviation for controls, in our case 18.9%) the specificity but not the sensitivity of the test would have suffered. Clearly, a positive test result is not diagnostic of RF, as 50% of unaffected first-degree relatives were found to be positive for D8/17. Rather, D8/17 may be seen as a marker of susceptibility to RF.

The intermediate levels of expression of D8/17 in unaffected first-degree relatives supports, but does not prove, the conjecture that the susceptibility is inherited, rather than acquired. These values are strikingly similar to the results obtained with serum 883 (or its clone 83S19.23) (Rich, Gray et al. 1988). The consistency of the elevation of D8/17 in first-degree relatives but not controls in this region where streptococcal infection is endemic suggests that inheritance, not environment or exposure to streptococci, is the cause of the observed difference. Conclusive proof that D8/17 is inherited rather than acquired could be obtained through a prospective study of levels

of D8/17 in a high-risk population. Such a study would also determine the relative risk of developing RF given a positive D8/17 result.

If D8/17 proves to be an inherited marker of susceptibility to RF then it has a number of potential useful clinical applications. One clinical application would be to select a cut-off point that could 'rule out' past or current RF. When patients present to remote clinics with symptoms that raise the differential diagnosis of RF, a negative result for D8/17 could be used to support the decision not to refer the patient to a tertiary referral centre for further investigation.

It is not currently possible to 'rule-in' a diagnosis of RF with a positive D8/17 result, as D8/17 levels are elevated in people with past RF, healthy relatives of index cases as well as some healthy controls. In these situations it is plausible that a positive result could represent susceptibility to RF rather than the actual disease state. Thus in areas of high incidence of RF, such as amongst Aboriginal people in the Northern Territory, the level of expression of D8/17 could be used as a screening test to identify a group at increased risk of developing RF. GROUP A STREPTOCOCCAL

If D8/17 indeed accurately defines a population at increased risk of developing rheumatic fever then the WHO recommendations for early disease detection can be fulfilled (Wilson and Jungner 1968; Whitby 1974). The WHO recommends that screening tests be used in diseases with a natural history that involves a latent or early stage when an intervention could change the outcome of the disease. Primary prevention measures have been shown to be modestly effective and with a favourable cost-benefit ratio in a population with a high incidence of RF (Bach, Chalons et al.

1996) Further refining the target population should improve the effectiveness of the intervention, in this case an educational programme directed towards the general population as well as health care workers that encouraged the active diagnosis and treatment of streptococcal pharyngeal infections with penicillin. Other potential interventions could include vaccination with the long-awaited Group A streptococcal vaccine (Carapetis and Currie 1998).

The elevation of D8/17 in RF beyond the levels observed in patients with RHD has been noted elsewhere (Khanna, Buskirk et al. 1989). Our results support D8/17 being significantly elevated in patients with RF when compared with patients with a history of past RF. This suggests that D8/17 could be incorporated into the Jones criteria for the diagnosis of RF, either as an additional major manifestation, or as an obligatory feature in all diagnoses of RF. Improving the accuracy of the diagnosis for RF is a worthwhile goal not only for the patient who either suffers years of painful prophylactic injections or the serious consequences of a missed diagnosis, but also for the health care provider in ensuring the appropriate allocation of resources. D8/17 levels could also usefully distinguish between chronic valvular disease of rheumatic versus non-rheumatic origin. This would be clinically useful in younger patients who would stand to benefit from RF prophylaxis.

For clinical applicability, a diagnostic test must be readily available and cost effective. We looked at one way of reducing the overheads of this technically sophisticated test: storing specimens in the form of frozen PBMCs so that they could be tested in batches. Short delays would be unlikely to have significant clinical impact, and could potentially lead to laboratory cost-saving by concentrating the need for equipment and

staff expertise. However, our results show that flow cytometry for D8/17 was not reliable when performed on frozen PBMCs stored at minus 80 degrees. The D8/17 assay performed on fresh whole blood remains the preferred standard for testing.

We also demonstrated that a short delay, such as that required to transport whole blood from a remote community to a central laboratory (up to eight hours), has no significant effect on the D8/17 analysis. This is of relevance in the Top End of the Northern Territory, as it would be in other areas of high incidence of RF such as in Central Australia and the Kimberley region of Western Australia, with patients living in very remote communities with infrequent transportation services.

8.2 Technical difficulties and the human response

A major difficulty for the research team was the coordination of skilled laboratory staffing and the collection of specimens. The skilled staff required to perform the flow cytometry on freshly stained whole blood were recruited from a distant urban centre. In the initial project design we collected blood samples over a limited time for immediate analysis as well as storage of PBMCs for later comparison. However, the test run of the D8/17 analysis on whole blood - more than seventy samples - failed due to human error (unfamiliarity with the Cell Quest software). This was not apparent for some months, so in the meantime, another thirty specimens had been collected and stored as frozen PBMCs for a parallel project: the comparison of D8/17 levels in RF with other acute conditions. These, along with the frozen PBMC specimens that were stored from the initial recruits, were discarded when it was found that PBMCs did not produce reliable results with the D8/17 assay. In this way the results from more than one hundred samples were lost.

Furthermore, as noted above, our PBMCs were stored at minus eighty degrees centigrade, rather than the standard minus 130 degrees centigrade, another human error. Whether the D8/17 assay is reliable when performed on PBMCs stored at minus 130 degrees centigrade is unknown.

These difficulties highlight the importance of the appropriate training of laboratory staff. Local expertise would have been highly desirable in this case, and would have circumvented some of the issues we faced in the storage and subsequent wastage of samples, in particular, samples taken during acute illness. As researchers, we have an obligation to maintain a high standard of work with minimal wastage of samples to maintain the goodwill of the community within which we work. We were honest about our early failings with the community that participated in this study. To our surprise, instead of anger and disappointment, the response was of sympathy and ongoing willingness to participate. This enabled further sampling for the definitive study producing the results as presented. This cooperation was in a large part due to our excellent local co-workers in the community who communicated the complex issues in the local language. Thanks to their assistance, the project eventually achieved an excellent outcome.

8.3 Future directions

The results from this research need to be taken further to achieve their full clinical potential. Firstly, D8/17 should be evaluated as a diagnostic test for RF by comparing D8/17 levels in patients who fulfil the Jones criteria, patients who do not fulfil the criteria initially, but are subsequently found to have RHD, and a broad spectrum of

patients with the differential diagnoses of RF such as infective arthritides, systemic lupus erythematosus, other post-streptococcal auto-immune diseases and valvular heart diseases. We have already shown that RF and RHD can be confidently differentiated using D8/17 analysis with very small numbers of samples. RF now has to be differentiated from other acute conditions that pose diagnostic difficulties to clinicians.

Secondly, it is important to confirm that D8/17 is a marker of susceptibility to RF rather than an epiphenomenon accompanying it. A cohort of D8/17 positive and negative people should be followed prospectively in order to assess the relative risk of developing RF according to D8/17 status.

Finally, the aetiological role of D8/17 and the other B cell markers identified should be sought. The sequence of events between infection with Group A streptococci and tissue damage is not yet fully understood. An understanding of the disease process may bring hope for the eventual development of a targeted intervention to prevent organ damage, such as carditis.

PART 2

The sharp end of the needle:

**Rheumatic fever prophylaxis and concepts of care for
Yolngu patients**

9 Introduction

The primary prevention of rheumatic fever (RF) is a long-term goal for the control of the disease in Indigenous Australians. However this will require considerable resources and is likely to require not only a disease-directed strategy, such as the development of a vaccine against group A streptococci, but also an overall improvement in living conditions for Aboriginal people. The secondary prevention of RF seems a more readily achievable goal: monthly injections of penicillin greatly reduce the risk of recurrent RF. Yet the rate of recurrent RF in the Top End of the Northern Territory has been rising: in 2002 recurrent RF represented 48% of all cases of RF, implying that penicillin prophylaxis coverage was inadequate (Edmond, Noonan et al. 2001). This had occurred despite the establishment of a rheumatic heart disease (RHD) control program in 1997. Following WHO recommendations, a centralised register of patients with a diagnosis of RF was instituted to oversee the delivery of secondary prophylaxis. In addition to this, educational resources were created with the aim of improving the knowledge of the disease by the predominantly Indigenous patients. The lack of clear improvement in response to the initiatives, despite their enthusiastic implementation, suggests the need for a new approach.

In the contemporary debate over treatment compliance, the doctor is no longer regarded as the source of objective truth and the patient as the sole source of the 'problem'. In the cross-cultural setting there has long been an awareness of the impact of cultural, language, socio-economic factors and the power differential on treatment adherence. In response to this, there has been a call to investigate the structural issues impeding therapeutic outcomes for patients, as well as to shift the power balance so

that patients and health service providers might become partners seeking improved health outcomes together (Humphery and Weeramanthri 2001). Furthermore, recent research has highlighted the profound miscommunication occurring in interactions between Aboriginal patients and non-Aboriginal health staff and raised the question of how a shared understanding of disease might relate to compliance with prescribed treatments for Aboriginal patients (Cass, Lowell et al. 2002). These ideas have informed this study, which is an exploration of the reality of people's lives, in an Aboriginal community in the Top End with a high incidence of RF.

The purpose of this study was to ascertain what factors affected the uptake of prophylaxis for RF. This led to related questions. What was the relationship between biomedical knowledge of the disease and treatment compliance? Who was taking responsibility for the health of RHD patients? I investigated what constituted good care for Aboriginal patients with RHD, and what were the roles of the different parties involved in caring for patients with RHD. A secondary aim was to describe the patients' experiences and explanations for the cause of acute rheumatic fever/ rheumatic heart disease, as a prerequisite for achieving a *shared* understanding of the disease. The answers to these questions were sought through the use of qualitative research methods, focussing on the experiences of patients, their families, and the staff looking after them in a remote Aboriginal community. This research falls in line with the National Heart Foundation's redirection of focus away from the basic sciences and towards public health and social research, especially in reference to Aboriginal people in rural and remote populations. Research priorities identified by the National Heart Foundation include "strategies to enhance community control and

participation in ... health services” as well as “building shared cultural understanding”
(Field and Wakerman 2002).

10 Preventing recurrent rheumatic fever

10.1 The evidence for penicillin prophylaxis for acute rheumatic fever

The primary attack of RF may result in chronic valvular disease, but the risk of significant cardiac involvement increases with recurrences. Patients with a past history of RF have a 50% chance of developing RF following a Group A streptococcal infection, which is up to 500 times the risk for an unaffected person (Manyemba and Mayosi 2002). Prompt treatment of streptococcal throat infections is not an adequate preventive measure, as infections can be subclinical, and thus undetected.

A Cochrane systematic review confirmed that prophylaxis with regular penicillin is highly effective in reducing the incidence of recurrent RF. Penicillin, when compared with placebo, reduces the rate of recurrent RF by 55%. Intramuscular penicillin is far superior to oral penicillin, reducing recurrent RF by between 87% and 96%.

Increasing the frequency of the injections from four weekly to two weekly is associated with further clinical benefit. The risk of recurrences of RF despite receiving regular prophylaxis is reported as up to 2% of patients per year (Manyemba and Mayosi 2002). However, in New Zealand the rate of recurrences in patients receiving four weekly penicillin prophylaxis was only 0.14% per patient year (Lennon 2003).

A simple analysis of the cost-benefit ratio of secondary prophylaxis for RF showed that the reduction in the length and frequency of hospital stays associated with good

coverage with penicillin outweighed the cost of the antibiotics (Strasser 1985). However, the prolongation of life and the subsequent need for high level medical services as well as the years of productive life saved were not accounted for and would likely contribute to the benefits achieved by prophylaxis. But similarly, the costs to patients and the health services of distributing the penicillin do not seem to be included.

10.2 Rheumatic fever control programs

In 1980 WHO made recommendations for the control of recurrent RF: it suggested that each country should establish a RF control program with a centralised register of RF cases, that there should be an organised method of distributing benzathine penicillin and that non-compliance with prophylaxis be actively addressed (Anonymous, 1980b). Evidence that a register-based system is effective in controlling RF can be found in the New Zealand experience. Prior to the establishment of a formal program for RHD, the loss to follow-up of patients being cared for with ‘standard medical resources’ was high: for patients cared for through hospital outpatient departments 31% were lost to follow up, compared with 77% of those cared for by general practitioners. Not surprisingly, recurrence rates were high: 40% for non-Maoris and 22% for Maoris (Neutze 1988). A register-based program was successful in reducing recurrent RF as a percentage of total hospital admissions from 22% to 6% (the difference for Maoris and non-Maoris was not described) (Lennon, Trotman et al. 1995). The mechanism by which the register led to a reduction in admissions with RF is a matter for conjecture, but noting that 30 out of 51 recurrences in this series were in patients who had never been on prophylaxis implies that

suboptimal prescribing practices may have been identified and rectified. This was noted to be a larger problem than patient non-compliance.

10.3 Rates of prophylaxis for patients with acute rheumatic fever

WHO quantified the rates of coverage of patients with RF with penicillin prophylaxis in seven developing countries in the 1970s. Out of a total of 5500 patient-years, 60% of patients achieved full prophylaxis (10-12 monthly injections per year), 20% achieved irregular prophylaxis (6-9 injections) and 20% achieved only occasional or no prophylaxis (Strasser 1985). In Soweto in the early 1980s only 17% of children with a diagnosis of RF received more than 75% of their monthly prophylactic injections. A community-based program raised the coverage to 38%: a disappointing result (Edginton and Gear 1982).

In 1986 WHO commenced a multinational program for the prevention of RF; a total of 12 000 patient-years were evaluated in sixteen developing countries. The average rate of coverage of secondary prophylaxis was 63.2% overall, but varied from 47.2% in the Americas, to 90.2% in the Western Pacific (see figure 12) (Nordet 1992).

National RF control programs in Thailand (Vongprateep, Dharmasakti et al. 1988), Taipei (Vongprateep, Dharmasakti et al. 1988), India (Iyengar, Grover et al. 1991) report prophylaxis coverage rates of 54-84%, 90% and 85-95% respectively.

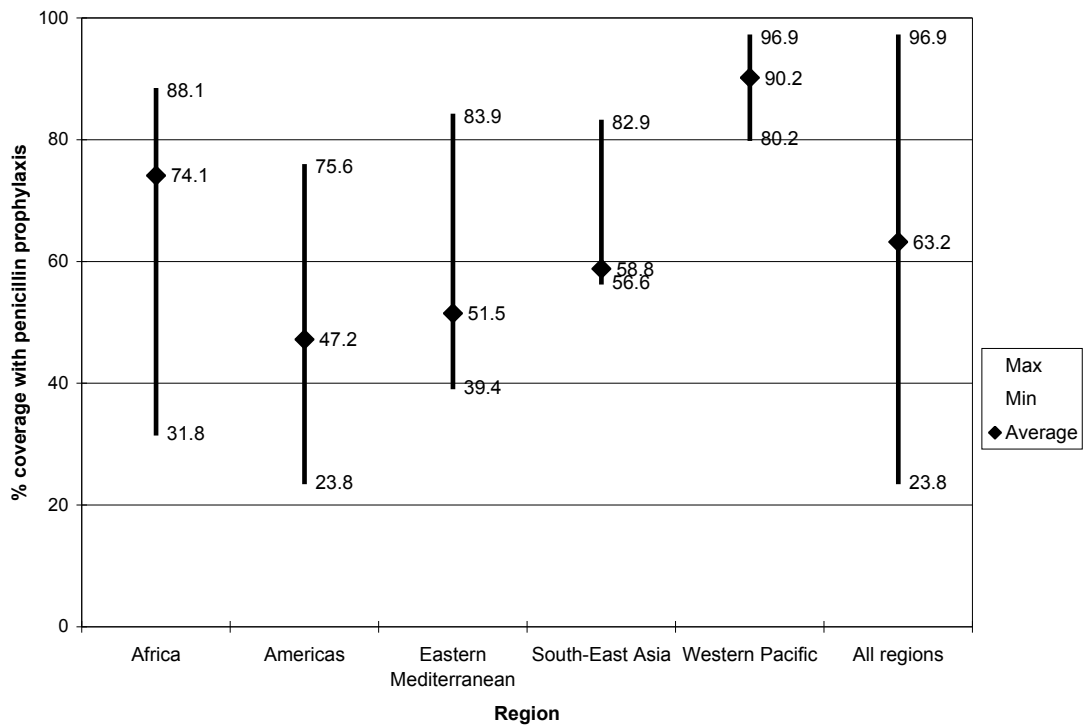


Figure 12 - WHO project for the control of RF: ranges and means of penicillin prophylaxis by world region

Source: (Nordet 1992)

In Australia, the average rate of penicillin coverage of patients with RF in twelve Top End communities during the mid 1990s was 66.1%. On average, 28.4% of patients received only occasional prophylaxis (less than 50% of scheduled injections). The rates differed greatly between communities, from one community where only 3.3% of patients had inadequate coverage, to another where all patients had inadequate coverage with penicillin (Carapetis 1998). In Central Australia four remote communities were selected as sentinel sites to assess the rates of penicillin coverage. The rate of coverage in 2000 was 55% and in 2002 it rose to 68% per patient-year (Brown, Purton et al. 2003). In Western Australia, only 5% of patients with RF received all (100%) of their prophylaxis (Ramsay 2000). In an audit of 78 patients from the Kimberley prescribed benzathine penicillin, median compliance was 67% of the prescribed injections, but less than 20% of injections were delivered on time. This

estimate does not take into account the results for a further 27 patients who were prescribed oral penicillin for whom compliance data could not be elicited. Of note, for 40 patients with RHD no prophylaxis was prescribed (Mincham, Mak et al. 2002).

10.4 RHD control program in the Northern Territory

In response to the high rates of RF and RHD identified in the Top End, a RF control program was established in 1997. The program's objectives reflected the recommendations made by WHO (Noonan, Edmond et al. 2001). Educational materials designed for use with Aboriginal patients and families were created (Angeles, Bengner et al. 1996; Angeles, Bengner et al. 1996). However data on rates of penicillin coverage were incomplete and thus the efficacy of efforts to improve adherence to secondary prophylaxis by community education and discussions could not be assessed. The mainstay of the program was a centralised register of patients to facilitate the coordination of secondary prophylaxis for RF. Regular communication between the register and the primary health care providers promoted the clinical care of patients and the maintenance of clinical standards.

The program aimed to reduce the incidence of recurrent RF, which constituted 39% of RF episodes prior to the establishment of the register (Carapetis and Currie 2001). Rates of recurrent RF initially decreased, but after 1999, they rose (figure 13, figure 14) (Carapetis and Currie 2001; Edmond, Noonan et al. 2001; Kelly 2003).¹ This contrasted with the improvement in the situation in Central Australia (Brown, Purton et al. 2003) and was an indication of the failure to achieve adequate coverage of

patients with previous RF with secondary prophylaxis. The situation in the Kimberley (North-West Western Australia) for the years 1988-1992 was equally grave. Rates of recurrent RF ranged from 17%-52% and showed no signs of diminishing (Richmond and Harris 1998).

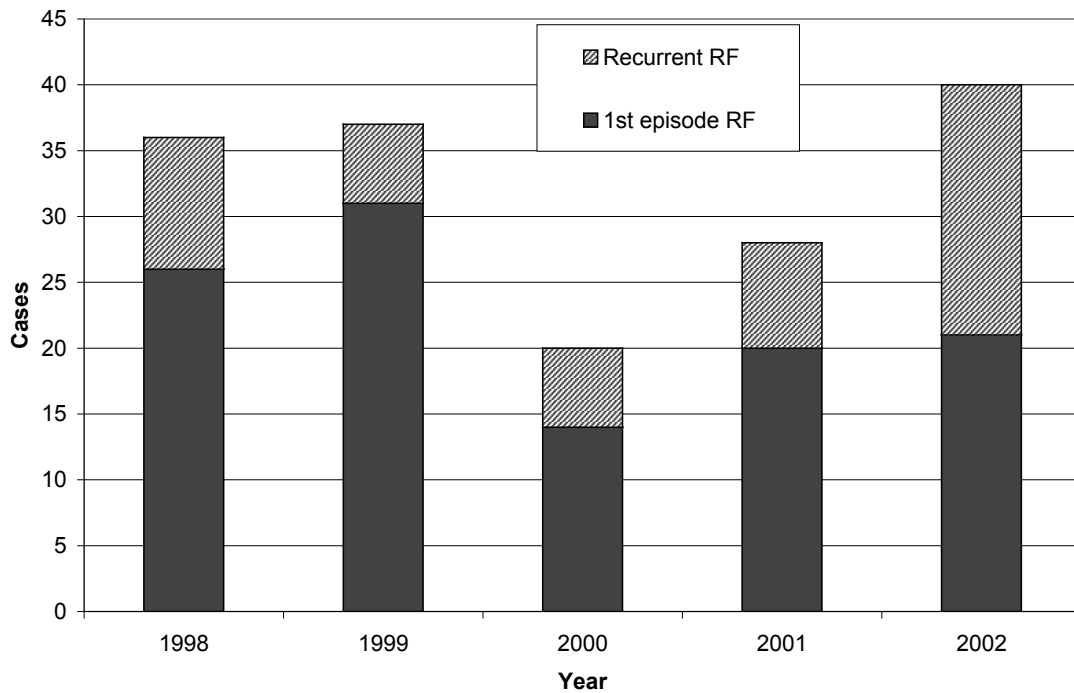


Figure 13 - Cases of acute RF in the Top End

Sources: (Carapetis and Currie 2001; Edmond, Noonan et al. 2001; Kelly 2003)

¹ The slump in the incidence of acute and recurrent ARF in 1999/ 2000 may be real or artefactual due to reduced case ascertainment: in the years 1999-2000 there was no resident cardiologist in the Top End.

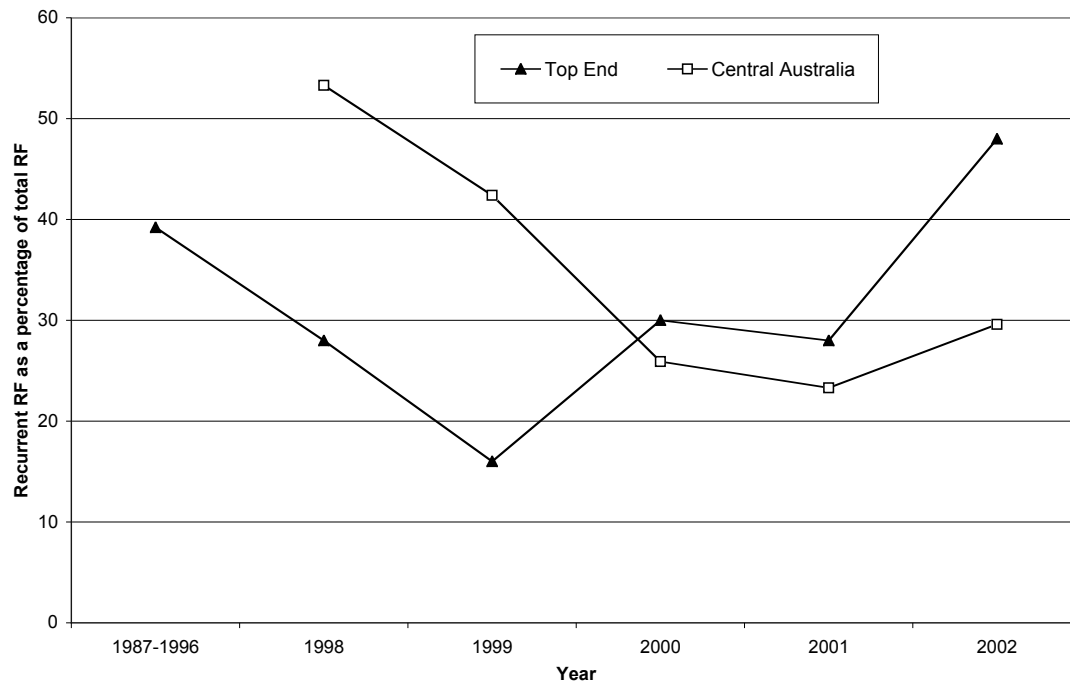


Figure 14 - Recurrent RF as a percentage of RF

Sources: (Carapetis and Currie 2001; Edmond, Noonan et al. 2001; Brown, Purton et al. 2003; Kelly 2003)

In response to the failure to achieve a reduction of recurrences, primary care centres were reminded of best practice management: four-weekly benzathine penicillin injections. Some centres had been using oral penicillin, and this was associated with a higher rate of recurrences (Currie, personal communication). Educational resources were developed for health care providers which included suggestions as to how to increase the uptake of secondary prophylaxis, but it seems that these have had limited circulation. Finally, it was recommended that every community health centre should have an active recall system with a designated local RF program coordinator (Kelly 2003). Thus it appears that approaches to improving the coverage of patients with RF have focussed on health service delivery measures, rather than tackling the controversial issue of patient compliance.

10.5 Summary

Monthly injections with benzathine penicillin are proven to be effective in controlling recurrent RF. The rate of recurrent RF in the Top End was rising despite the implementation of a rheumatic fever control program and RHD register. Whilst a small number of recurrences may occur even with full penicillin coverage, recurrences generally indicate failure to achieve adequate secondary prophylaxis. It is not known whether this is due to factors in health service delivery or patient factors, such as compliance. This situation is part of a more general picture where Aboriginal and Torres Strait Islander people are up to eleven times more likely than non-Indigenous people to be hospitalised for a preventable condition (Stamp, Duckett et al. 1998). The predicament in the Top End is not unique. Data from Western Australia and many other world regions give a similar impression of failure to achieve comprehensive coverage of penicillin for RHD patients.

11 An overview of 'compliance'

11.1 Rethinking or 'forgetting' compliance

Patient compliance has been seen as of paramount importance in achieving good health outcomes. That doctors have the authority, based on knowledge of medical science, to make treatment decisions for patients is implicit. Non-compliance has been attributed to errant patient behaviours, negative aspects of the therapeutic regime, service delivery factors or difficulties with the clinical interaction between health care providers and patients (for example, the 'doctor-patient relationship'). There was little investigation of the broader social and cultural contexts for compliance, although a relationship between compliance and socio-economic background, ethnicity and language skills was found (Humphery and Weeramanthri 2001). The traditional approach to solving the problem of compliance had health professionals systematically, if not quasi-scientifically, measuring and overcoming the obstacles to compliance (Becker, Maiman et al. 1979). Strategies to improve compliance largely concentrated on patient educational and behavioural modification, although the doctor-patient relationship has also received some attention (Humphery and Weeramanthri 2001).

Thus compliance has been traditionally defined as a predominantly clinical problem. Yet as early as 1979 researchers looked to the related disciplines of management, epidemiology and social psychology to understand compliance (Sackett 1979). The contemporary debate on compliance, in post-modern fashion, redefines the problem itself. Humphery et al. suggest that the problem is not so much the *lack* of patient compliance, but the *notion of compliance* itself. They argue that medical practitioners

and patients hold valid but different perspectives on health issues. Even when a common goal is pursued, the strategies of doctors and patients may differ, thus the purpose of the clinical interaction should be an informed decision by the patient, rather than treatment compliance *per se*. A similar argument is put forward by Langer who states that the most important step in becoming “culturally competent professionals” is to recognise the relativism of the expectations of the health care provider and the validity of the patient’s experiences and choices. The health care provider should acknowledge how the ‘sociocultural location’ of the patient will determine their actions (although Langer fails to comment on the effect of this ‘sociocultural location’ on the health care provider). Yet in the final analysis Langer reverts to outcome measures such as patient ‘performance’ and compliance (Langer 1999).

To relinquish a therapeutic goal is difficult for many health professionals. When they perceive choices as equivalent, or with few consequences, health professionals often encourage patients to make health decisions appropriate to their personal situation, such as in the choice between contraception strategies. In socially-condoned self-destructive behaviours, such as smoking, many health professionals will leave it “up to the patient” (to make the decision to quit), but few could comfortably support patients’ refusal of life-saving therapy. This is both a moral issue, as well as increasingly a legal one, bound up in the “duty of care”. The discomfort with treatment refusal is particularly apparent in hospitals in the antagonistic way we deal with the common situation of a patient requesting to self-discharge.

In their analysis, Humphery et al. recognise that health professionals will be reluctant to abandon compliance, and give up on ideas of public health and the responsible allocation of health resources. Yet they insist that compliance does not need to be 'solved', rather that the assumption of the authority of doctors needs to be challenged. They suggest two strategies: firstly, looking 'before' compliance, at the relativism of the beliefs and perceptions of health professionals and, secondly, looking 'beyond' the clinic walls to acknowledge the impact of the social and cultural context of the medical interaction. Thus on the one hand the authors demand that compliance be 'forgotten', yet they suggest new and constructive ways of addressing the issue.

[In discussing non-compliance] we are talking neither about [a] 'problem' nor simply 'patient behaviours' but about an issue more usefully and constructively framed as *the material consequences of particular models and practices of health service provision, undertaken within particular institutional, political, social and cultural contexts*. By adopting this view, we can both jettison the pejorative implications of the language of 'problem patients' or indeed of 'problem groups' while continuing to address the concrete reality that services and treatments being offered within a Northern Territory context are often not being taken up, to the detriment of people's health. We can suggest also that this is a product of a dynamic between provision, on the one hand, and the broad social and economic context of Aboriginal ill-health (and individual patient health behaviours) on the other. (Humphery and Weeramanthri 2001) (Original italics)

Humphery et al. suggest the need to act on three levels to bring about change in the dynamic of the provision and uptake of health services. These are the biomedical level (the locus of most of the current attention), the level of health service delivery and its institutional structures, as well as a broad social, cultural and economic level. Thus, as a researcher investigating ‘compliance’ or ‘treatment uptake’ in the setting of RHD in an Aboriginal community, I will attempt to be aware of the contexts of colonialism, the power differential between health care providers and their patients, and the crisis in communication demonstrated between health service providers and Aboriginal people in the NT. As a doctor, I will be looking for ways to improve patient care, reduce morbidity and prolong life in a population that has an unfair burden of disease whilst recognising that my strategies may not be those of my patients and their families, and even that our priorities may differ.

11.2 Compliance and Aboriginal Australians

Most of the research relating to compliance of Aboriginal patients is conceptually conservative – focussing on quantification of the ‘problem’ and achieving clinical ‘solutions’ (Burns 1992; Kemp, Nienhuys et al. 1994; Kruske, Ruben et al. 1999). Kemp et al. attempted to identify causes and solutions to non-compliance with antibiotic treatment of children with ear infections in a remote community. Patient factors were believed to predominate and included low motivation, forgetfulness, and the poor health of the carer. The researchers also identified environmental aspects, such as ants eating the medication, and socio-economic factors, such as the lack of refrigerators, as having an impact on patient compliance. Yet the recommended solutions were educational and behavioural strategies to promote treatment uptake; addressing the environmental issues was seen as counterproductive. The researchers

described their approach to the patient as “one of mutual cooperation wherein, ideally, both parties have recognised the benefits of ... therapy” but they provided little evidence of a collaboration towards a therapeutic alliance (Kemp, Nienhuys et al. 1994).

In their investigation of the attitudes of NT health practitioners to the compliance of Aboriginal patients, Humphery et al. found that most health practitioners interviewed saw treatment compliance as a major issue. The reasons for non-compliance were mostly attributed to patient factors such as communication difficulties, a lack of a biomedical understanding of the disease, problems with the practitioner-patient relationship, conflicting priorities, the lack of family support, apathy, lack of perception of ill health. Features of the treatment regime and health service delivery were also mentioned, such as the impracticality of the treatment and the lack of a culturally appropriate hospital service. Socio-economic factors included lack of education, social mobility and the separation from family that the treatment required. Few practitioners acknowledged the larger issues of the institutional and socio-economic contexts of health service delivery and treatment uptake. It was on the basis of the narrowness of this construction of the problem of compliance that the recommendation for a more culturally and politically informed approach to compliance should be developed (Humphery and Weeramanthri 2001).

Devitt and McMasters investigated the experiences of Aboriginal people on renal replacement therapy in Central Australia. These authors highlighted the impact of social and political issues such as poverty, domestic overcrowding and relationship difficulties on patients' interactions with health services. The presence or absence of

family support was nominated as the single most influential factor affecting patients' compliance with treatment. Patients' lack of perception of declining health was also identified as a factor reducing compliance. Finally, patients and health practitioners were shown to have different perceptions of the requirement for pedantic adherence to a treatment regime, leading to dissonant assessments of patient compliance (Devitt and McMasters 1998).

With his experience in primary health care in an East Arnhem Aboriginal community, Bryce proclaimed the principles of developing credibility as a healer within the community, gaining the trust of the patient and overcoming “the considerable language and world view barriers that exist to good communication”. These strategies are equivalent to establishing oneself as a source of authority, improving the doctor-patient relationship, and successfully transferring a Western biomedical explanation of the disease to improve patient compliance. Bryce departed from this conservative opening to present a more radical, reflexive and power-shifting challenge to health professionals. He suggested that non-Indigenous people “need to listen more and lecture less” and advised health professionals not to “fall into the trap of ‘blaming’ indigenous patients for not ‘fitting in’ with their own medical model”. At a social level he described barriers to adherence including alcoholism, domestic violence and the ‘welfare mentality’ that originated in the history of colonisation. Finally, on a political level, he suggested that the community control of the health service was likely to create a more culturally sensitive and effective health service (Bryce 2002).

11.3 Compliance and patient education

The quality of communication is commonly believed to affect patients' decisions to follow medical advice. The literature pertaining to communication tends to focus on the effective transfer of a Western biomedical understanding of disease from the health practitioner to the patient; the practitioner's understanding of the patient's perspective holds less importance. A commonly cited cause for non-compliance by Aboriginal patients is a lack of a Western biomedical understanding of health, disease and treatments (Humphery and Weeramanthri 2001) (p42). Is there evidence to substantiate this perception?

Early research did not find that educational strategies improved compliance (Haynes 1976). In a recent "scientific review" communication strategies such as 'information', 'counselling' and 'reinforcement' were listed amongst other methods of enhancing patient adherence to prescription medication (McDonald, Garg et al. 2002). The accompanying article providing advice to practicing clinicians recommended "counselling about the importance of adherence", "counselling about the regimen" and conducting "support group sessions" (Haynes, McDonald et al. 2002).

Communication on a deeper level either about the disease or the patient's priorities was not included - perhaps due to a lack of evidence relating to efficacy let alone cost-effectiveness. Even so, Haynes concluded that "negotiating priorities with the patient" was of use in establishing compliance, touching on the role of reciprocal communication.

Research that is informed by sociological theory is more supportive of efforts to improve communication between health practitioners and their patients. There is a

small amount of literature pertaining specifically to the role of communication as a method of improving compliance for Aboriginal patients. No direct link was found between knowing the cause of the disease in biomedical terms and adhering to treatments in a qualitative study of patients with end stage renal disease in Central Australia (Devitt and McMasters 1998). By contrast, both Bryce and Trudgen working in East Arnhem Land argue that educating the patient in the Western model of disease causation as crucial to ensuring treatment uptake. Both described cases where sensitively transmitted information in the patient's preferred language had instant and remarkable results, apparently often after many years of ignorance and miscommunication (Trudgen 2000; Bryce 2002). The evidence provided is anecdotal and personal, therefore neither scientific nor objective. A qualitative study investigating the views of patients with rheumatic heart disease in the Kimberley found that patients desired more information about their disease (Mincham, Toussaint et al. 2003). Without further substantiation, the authors concluded that "though a sound understanding of the disease and disease process is not the only determinant of compliance and effective management, it must still be considered an essential requirement for individuals undertaking long-term management" (Mincham, Toussaint et al. 2003). Ethically justified, though it is, no evidence was provided to suggest better clinical outcomes for patients who 'understood' their disease.

Good cross-cultural communication requires an understanding of the patient's world-view, an attention to the detail of language and the development of appropriate metaphors to construct a shared understanding of physiology and disease. Poor communication affects the health care provider's ability to inform the patient about his/her condition, and deliver general health education. One of the few authors to

comment on the importance of patient-doctor communication, Trudgen pointed out that poor communication may also affect a doctor's ability to make a diagnosis. More profoundly, it would impair the ability to "diagnose the overall problem", referring to the underlying social preconditions for illness (Trudgen 2000).

Gross miscommunication between non-Aboriginal health professionals and Aboriginal patients was identified as a likely cause for poor treatment uptake amongst patients in a renal dialysis unit in the Top End according to Cass et al. In their careful study of clinical interactions, "miscommunication reduced the ability to actively engage (the patient) and his family in controlling his blood pressure, in retarding progression of his renal disease and in planning for future dialysis" (Cass, Lowell et al. 2002). The authors of this study, like Trudgen, claimed that effective communication was not simply about using the appropriate language, but also acknowledging the social, cultural and political contexts of communication.

11.4 Compliance with rheumatic fever prophylaxis

11.4.1 Patient factors

Researchers attempting to identify risk factors for non-compliance with RF prophylaxis have generally focussed on patient demographics. A case series in India showed that compliance related to occupation and the number of medications prescribed, but not age, sex, marital status, distance of residence from the hospital, symptoms and the number of previous admissions to hospital (Rolston, Brahmadathan et al. 1981). By contrast, a South African case-control study showed that race, sex, age, the distance of residence from the hospital and the frequency of appointments

were significantly related to treatment uptake (Walker, Human et al. 1987). In Thailand an association between increasing age and reduced uptake of penicillin prophylaxis was described (Vongprateep, Dharmasakti et al. 1988). The WHO program of RF control in sixteen developing countries showed variable rates of uptake in different world regions. The reason for the variations are said to include logistical problems such as inadequate supplies of penicillin, and problems with reporting, but also low patient compliance (Nordet 1992). This point was not elaborated, but raises the question of possible national or ethnic differences in treatment compliance.

Less quantifiable factors relating to treatment uptake such as patient beliefs, motivations and priorities have been difficult to study. Referring to compliance with prophylaxis for RF in India, Rolston highlighted the unknown factor of patients' personal perspectives on treatment compliance in RF:

... inconvenience to the patient, expense, pain and discomfort , the patients' emotional make-up, social circumstances, illness of others in the family and the quality of the relationship between the physician and patient would also influence compliance in any program. Some of these factors are difficult to quantify and only an intelligent guess can be made on the impact they may have on drug compliance. (Rolston, Brahmadathan et al. 1981)

11.4.2 Service provision

Factors relating to service provision have been recognised to affect treatment uptake. At a policy level, RF control must be made a priority, which can be difficult in

developing countries where health services struggle to deal with the acute crises that occur with regularity (Anonymous, 1980a). A centralised register of patients with RF is effective in improving the overall coverage of rheumatic patients with secondary prophylaxis (Carapetis and Currie 1998). Targeted education leading to the harnessing of community and health professional support may be required for good outcomes. No specific mechanism was suggested, other than the “organised and concerted efforts of physicians, parents, teachers and health personnel” (Lue, Wu et al. 1988; Bach, Chalons et al. 1996).

In a paper unique for its recognition of the social and economic factors affecting treatment adherence, Walker et al recommended a number of structural changes to service provision for RHD patients in South Africa: providing better transport, keeping appointments to a minimum, strengthening the role of local clinics, and ensuring the availability of affordable health care (Walker, Human et al. 1987). At an individual and clinical level, the particular needs of adolescent patients have been noted but not elaborated on (Walker, Human et al. 1987; Haffejee 1992). Directed patient education and support by allied health staff were seen as essential (Walker, 1987; Anonymous 1980b). The reason for the success of an Indian educational program was put down to concerted patient education, based on real-life cautionary tales. This rather negative measure was supplemented by practices of active recall and reminders (Iyengar, Grover et al. 1991).

11.4.3 The local situation

In the Top End of the NT, Central Australia and the Kimberley a number of authors have supplied reasons for imperfect adherence rates to RF prophylaxis, but these are

largely unsubstantiated by evidence (Richmond and Harris 1998; Noonan, Edmond et al. 2001; Brown, Purton et al. 2003). In these papers, patient factors reported as interfering with the uptake of treatment include “community activities and priorities” and the mobility of the population. Service delivery factors were also implicated, and included a poorly maintained register in the Kimberley, high staff turnover throughout the three regions, difficulties in service provision associated with remoteness and insufficient resources, the lack of communication between clinics and the (presumably negative) attitudes of health staff to active follow-up.

One paper, however, describes the results of qualitative in-depth interviews of seven patients or parents of patients with RF or RHD in the Kimberley (Mincham, Toussaint et al. 2003). This study attempts to directly address the problem of poor compliance with RF prophylaxis in the Kimberley. A close relationship between the patient (or parent) and the health service providers and the support of the family for treatment were identified as important factors favouring patient compliance. Patient dissatisfaction with health services was recurrently implicated in poor compliance. Patients complained about inefficiency in urban health systems, about the intermittent provision of services, the transience of staff and the lack of choice for rural patients. Interestingly, one patient was concerned about a lack of confidentiality (an *excess* of familiarity) amongst community health staff. A passive resistance to health staff was implicated in one patient declining to be reminded of the timing of his monthly injections, whilst having no reliable system of his own to remember them.

11.5 Summary

Convincingly argued by Humphery *et al.*, ‘compliance’ is a pejorative concept that needs to be reworked. The relativism of health beliefs is commonly unrecognised, leading to a paternalistic approach by health professionals. Problems relating to health service delivery are commonly overlooked. Furthermore, the broader contexts of the patients’ economic, social and political situation remain unaddressed. In the cross-cultural context found in Aboriginal Australia these issues are pertinent, as communication issues and differences in health beliefs could be expected to affect treatment uptake. Yet treatment uptake - especially treatments perceived as having significant benefits - remains a pivotal issue for clinicians.

Health service providers have conjectured as to the reasons for the failure of uptake of prophylaxis for RF, but a comprehensive study has not been performed. In a more respectful and informed approach to health service delivery there is a role for understanding the perspective of the patient, their understanding of their disease, and their attitudes to treatment. At the same time, the attitudes and motivations of the health service providers should be assessed and analysed. This is the purpose of this study.

12 Setting the scene

12.1 A brief historical and contemporary account

Galiwin'ku is an Aboriginal community in Northeast Arnhem Land, on Elcho Island. Elcho Island is located approximately 600km east of Darwin, only a few kilometres off the coast of mainland Australia. The Island is nearly 60km long and less than 8km wide. Despite the monsoonal climate, the island's airstrip and roads remain mostly open throughout the year.

The settlement was founded by Methodist missionaries in 1942, and many Galiwin'ku residents remember mission times fondly. The Reverend Shepherdson was a formidable man who flew his own plane and oversaw the establishment of timber and fishing industries, a productive market garden and a school. In 1973 a town council was established in Galiwin'ku in a move towards Aboriginal self-determination initiated by the Commonwealth Government. Limited self-government was transferred to the council in 1977, with a reduced missionary presence on the island. Currently the community council has an Indigenous chairperson and a council board with broad community representation. Since the 1970s, the homelands movement has seen the foundation of a number of 'outstations', whereby Aboriginal people returned to their traditional lands to live (Djandilnga and Barlow 1997). The main community and the outstations have separate health services and municipal services providers.

Yolngu is the term commonly used to identify the local people, and non-Aboriginal Australians are known as *Balanda*.² Yolngu people identify themselves primarily by their family, then by clan, language and their traditional country (Djandilnga and Barlow 1997). Clans are patrilineal, and represent the descendants of one man and his brothers (Reid 1982). Many clans are now living in Galiwin'ku, even though traditionally each would have lived separately. This is the reason given for much of the social tension perceived by Yolngu living in Galiwin'ku. Whilst each clan has its own language, Djambarrpuyngu has become the language commonly used as it was promoted by the church, mission and school authorities.

The population of Galiwin'ku is approximately 1800, according to a community-based census (Djamalaka, 2002, personal communication). According to the Australian Bureau of Statistics census of 2001, the median age for the people of East Arnhem Land was 22 years, the median household size was 6.5 and the median weekly individual income was between \$160 and \$199 (Anonymous, 2004c). These figures underestimate the household crowding within most Aboriginal communities. In a community-based census it wasn't uncommon for more than twenty people to be living in one house (Djamalaka, 2002, personal communication). The poverty created by low levels of income is exacerbated by the high costs of living related to food, power and transport.

Facilities in Galiwin'ku include a general store, run by the Arnhem Land Progress Association (ALPA), a not-for-profit cooperative. Fresh fruit and vegetables are subsidised so that costs are equivalent with urban centres, but the remainder of the

² *Balanda* is derived from the Macassan word for "Hollander".

foods and other products are considerably dearer than elsewhere. Two privately owned takeaways operate on the island, as well as the one run by ALPA. These sell large amounts of ‘fast food’ at prices up to double those of urban centres. There is a childcare facility and preschool. The school provides tuition up to year ten, but there are plans to extend services to cater for matriculation students. Less than fifty children attend high school, with a particularly steep drop-out rate for girls. Amongst Yolngu adolescents, competency in English is poor, and literacy in any language is low.

The community development and employment program (CDEP), amongst other commitments, has contracts for land clearing, house painting, fishing, and rubbish collection and ceremonial preparations. (Yolngu involved in CDEP programs are counted as ‘employed’ in the census, although they receive only a small supplement to the unemployment benefit.) The Traditional Credit Union has an office on the island. The art centre coordinates the distribution of materials to the artists and the sale of artworks both locally and internationally. The church has a Yolngu as well as Fijian minister. Work is proceeding at the Bible translation centre on a Djambarrpuyngu version of the New Testament.

Elcho Island is currently the site for numerous Darwin-based medical research projects. The *Yalu marngithinaraw* (nurturing centre) is a self-constituted organisation for the protection and nurturing of Yolngu culture that often takes on the role of cultural mediation between the community and researchers. A knowledge centre was founded in 2003 with the aim of preserving Yolngu knowledge through the archiving of cultural materials and the recording of new material of cultural relevance.

The health centre is run by the community council, and is staffed by one permanent doctor, five nurses and between one and ten Aboriginal health workers. Health care, including medications, is free of charge to Aboriginal residents. Medical specialists and dentists visit the community on an infrequent but regular basis. There is no facility for inpatient management on the island. All patients requiring radiologic investigation or admission are transferred to the nearby Nhulunbuy or the more distant Darwin hospitals.

The majority of Yolngu in Galiwin'ku are unemployed, although they are occupied with childcare, looking after the elderly, hunting, fishing, producing art, and ceremonial duties. Organised sport is very popular, with football and basketball competitions held annually. Other popular pastimes include playing cards, playing and listening to music. Substance abuse is not uncommon: kava, marijuana, petrol sniffing and alcohol abuse is apparent, even though alcohol is officially prohibited.

As in many Aboriginal communities, there is inadequate housing and maintenance services, although efforts are being made to rectify this. Houses range from simple demountables on concrete slabs, to architect-designed tropical pavilions made from modern materials. Homes are crowded by non-Aboriginal standards, with each room typically sleeping four or more people. There is little furniture; some houses have washing machines and refrigerators. Few houses have a private telephone. Food is often cooked outside around a campfire, but few choose to sleep outside, except during times of ceremony.

There is a sense of close family bonds and an active social life. People are rarely alone, and express concern when *Balanda* live alone, or walk around alone. Even when there are few people about, there are many dogs, which are, on the whole, looked after well and domesticated effectively. Connections are maintained with relations in the nearby communities and those living in Darwin. Galiwin'ku not only has familial ties with other communities, but there is some evidence of cultural ties in the moiety system that exists throughout Arnhem Land, the close relationship of the languages of the area, and the links with myth and ceremony (Wilson 1993).

There is no public transport. Within the island transport options include private vehicle or boat. Outstations off the mainland are reached by chartered lights. Fuel (AVGAS) is expensive by Australian standards, as are airfares. Travel beyond the community required for medical reasons is fully subsidised. Within the community the health clinic provides transport for patients on request.

12.2 The Yolngu medical paradigm

12.2.1 Illness and disease causation

I will now turn from the historical and social setting, to the intellectual and conceptual setting: Yolngu perceptions of health, disease, and the body, with particular reference to the heart. I will make some observations about Yolngu beliefs, but acknowledge that generalisations must be somewhat superficial and inaccurate. Mainstream Australian beliefs will be termed 'Western', whilst recognising that this term has its own history and biases.

There are a few accounts of Yolngu people's experiences of and explanations for disease (Reid 1983; Trudgen 2000) (Scarlett, White et al. 1982) as well as accounts from west Arnhem Land (Berndt 1982) and Central Australia (Nathan and Japanangka 1983). A combination of traditional and Western beliefs is often described. Reid, in her detailed investigation of health beliefs of the Yolngu of Yirrkala in 1974/75 found *galka* (*djängitj*, or sorcery) to be the main cause of serious illness. The underlying reasons for the use of sorcery was usually a transgression of social or sacred laws by the victim. Reid also identified a number of 'natural' as well as 'emergent' causes of disease according to Yolngu, to which I have added those observed by Trudgen (see table 16).

According to Reid the causes and the categories of disease were neither fixed nor exclusive. People held more than one explanation for a death, either because they were undecided, or because both were true, at different levels. *Galka* was commonly believed to be the cause of sudden, fatal illnesses, such as heart attacks. But despite a belief in the supernatural cause of disease, many Yolngu chose Western treatment, whilst herbal remedies and the healing powers of the *marngitj* (traditional healer) were also respected. Reid commented that although Yolngu did not use biomedical language to explain disease causation, perhaps due to insufficient grasp of the paradigm, they accepted the *prima facie* evidence of the efficacy of Western treatments (Reid 1983).

Table 15 - Yolngu perceptions of the cause of disease

| Social and spiritual | 'Natural' | 'Emergent' |
|--------------------------------------|-----------------------|---|
| Sorcery | Contagion | Alcohol |
| Transgression of sacred laws | Emotional state | Medical mismanagement |
| | Exposure | Illness, defined in Western medical terms |
| Failure to observe food restrictions | Food | |
| | Heredity | Motor vehicle accident |
| Breaches of social norms | Neglect | Sin |
| | Old age | Smoking |
| | Injury | Poor living conditions |
| | Sting or envenomation | Living in settlements |
| | Pregnancy | Conspiracy |
| | Self-neglect | Race |
| | Suicide | Disrespect for social laws |
| | No attributable cause | |

Sources: (Reid 1983; Trudgen 2000)

12.2.2 The Yolngu body

Just as Yolngu and Western concepts of the causes of disease differ, so do their understanding of the body and the function of its parts. From the outset, the Yolngu body is firmly situated within society and the land, unlike the more individualised and decontextualised Western body (Wilson 1993). A linguistic connection is made between landforms and body parts: a point is called a 'nose', a waterhole is an 'eye', bushland is the 'back', a river bend is an 'elbow' (Wilson 1993). Similarly, body parts

also represent relations and can be used in an effective sign language: breasts represent the mother (and her traditional lands), the forehead the father (and his traditional lands), the back the maternal grandmother, the hip the husband or wife, the lower leg represent younger brothers and sisters.

The heart is the organ that is most damaged by RF. In her analysis of Yolngu perceptions of the body, Wilson states that the heart, or the chest area in general, is the “centre of the life force or soul of the individual” (Wilson 1993). Whilst distinct terms for breath and pulse exist, the concepts appear to be frequently conflated and the heart is said to be the source of both (Amery 1986). Yolngu do not traditionally hold a concept of the circulation of blood - blood was said to be a connecting medium, rather like a river (Wilson 1993; Trudgen 2000) (Devitt and McMasters 1998). The common words for artery, vein, tendon, ligament, nerve are *gurrkurr*, meaning root, and *raki*, meaning string, and therefore do not indicate a hollow structure that could act as a conduit for blood (Amery 1986; Christie 2002).

Due to its importance the heart is the area most susceptible to attack by sorcery (Wilson 1993). Reid provided a frightening account of how *galka* might work:

A *galka* finds a person and makes him unconscious ... sometimes his hand goes up through the anus and rectum and he pierces the heart in several places with small needles so the blood flows everywhere. (Reid 1983)

There are no references to Yolngu concepts of RHD or its causation in the literature. The accounts above are by no means comprehensive descriptions of the Yolngu view

of causation of disease, or concepts of the body, and so answer few of our questions about Yolngu ideas about RHD. Still, they serve to illustrate the marked differences in the Western and Yolngu paradigms.

13 Qualitative research methods

13.1 The role of qualitative research in health

Advances are constantly being made in medical knowledge, however frequently there is failure in applying this knowledge in the real world. Thus the population at large is not experiencing all the potential benefits of basic medical research. Qualitative research methods are appropriate for studying clinical scenarios as events complicated by issues of human psychology, society and culture (Green 1998; Ritchie 1999; Meyer 2000; Malterud 2001).

Qualitative methods are useful for the study of human and social experience, communication, thoughts, expectations, meaning, attitudes, and processes, especially related to interaction, relations, development, interpretation, movement, and activity – all core components of clinical knowledge. (Malterud 2001)

Qualitative research methods have been used to study treatment uptake ('adherence'), acknowledging the value of understanding patients' perspectives. Studies typically highlight the impact of patients' beliefs on their willingness to take treatment as prescribed (Green 1998). However the usefulness of qualitative research methods can be extended beyond the investigation of patient beliefs to look at physician practices, service delivery and institutional issues. Qualitative research can complement quantitative research; quantitative research has strengths in describing and assessing the magnitude of a phenomenon but qualitative research can provide an interpretation or explanation of it (Ritchie 1999).

13.2 Qualitative research methods

13.2.1 Introduction to qualitative research

Qualitative research is a broad approach of inquiry encompassing many methodologies. The choice of the most appropriate method will depend on the research question, and will in turn influence the method of data collection, the selection of participants, the focus, outcomes and narrative form of the study (Creswell 1998). Data for qualitative analysis can take many forms. Commonly used techniques include questionnaires, in-depth interviews and focus group interviews. A single study may make use of multiple forms of data in order to reduce the biases inherent in any one method of data collection. Likewise, there are a number of described ways of performing data analysis. Some qualitatively obtained data, such as questionnaires, can even be analysed quantitatively.

13.2.2 Interviews

Interviews are a common method of data collection in qualitative research. Individual in-depth interviews form the basis of a case study. Group interviews may be ‘focused’, or informal. A focus group consists of between six and ten people, previously unknown to each other, but purposively chosen for their homogeneity. The interview is formal in the sense that the location and questions are preset, although the moderator attempts to generate an open discussion where each participant has the chance to air their opinions. Informal group interviews, by contrast, can be spontaneous, convene in a natural setting, involve varying numbers of self-selected participants and be relatively unstructured. Informal groups typically convene around

natural social groups, and participant numbers may vary during the course of the interview (Khan and Manderson 1992; Carey 1994; Schensul 1999).

Group interviews have a number of advantages over individual interviews. A large number of people can be interviewed in a relatively short time, thereby producing a lot of data. Furthermore, the interaction between the participants can generate data that would otherwise be inaccessible to the interviewer, such as participants' reactions to each other. Group interviews "elicit useful 'natural language discourse' that allows the researcher to learn idiomatic expressions, common terminology, and communication patterns in the community" (Schensul 1999). Group interviews reduce the potential for participants to misunderstand questions or produce responses to please the researcher (Khan and Manderson 1992). A practical advantage of informal interviews is that the groups are self-selected, so "such natural clusterings of people represent, in a loose fashion, the resources upon which any member of the group might draw, both in material terms and with respect to information and advice" (Khan and Manderson 1992).

Disadvantages of group interviews include the potential for censoring and conformity of the participants, domination of the group by an individual, and concerns regarding confidentiality (Carey 1994). Difficulties can arise in maintaining the formal aspects of the focus group interview in the field, as participants' priorities may not coincide with the researchers' (Khan and Manderson 1992). When the researcher is not fluent in the local language, the interview will depend on the skills of the group moderator, and the ability to translate the words and concepts elicited in the interview.

13.2.3 Grounded theory analysis

Grounded theory, first described in 1967 by Glaser and Strauss, emerged from the field of sociology (Glaser and Strauss 1967). It was expanded and updated by Strauss and Corbin (Strauss and Corbin 1998), and interpreted in a simple and applicable manner by Creswell (Creswell 1998). (See table 16 for a definition of terms used in this section.) The focus of this tradition is to ‘discover’ theory. The process begins with an area of interest, or a phenomenon. A hypothesis is not formed prior to the study, as the theory is created inductively. Data may be collected through interviews, or looking at documents relevant to the phenomenon of interest. Enough data should be collected so that no new information is discovered and ‘saturation’ is achieved. Data analysis is performed concurrently with data collection, so that further investigation of the phenomenon can be purposively directed, not only by ‘theoretical sampling’ but also by altering the interview themes, questions or techniques.

Analysis consists of a systematic approach beginning with the interview data and building a theory from this ‘ground’, through processes of conceptualisation, reduction, organisation and relation of the data. The first stage - ‘open coding’ - is the process of abstracting units of information found within the data to form concepts. These basic building blocks are then organised into categories, which may have properties and dimensions. ‘Axial coding’ relates categories to each other to describe processes and structures. Processes have conditions (either casual or intervening), actions/ interactions and consequences, which may in turn impact on conditions. ‘Selective coding’ involves identifying the central issue and refining the theory. Classically, a visual model, diagram, or chart-like matrix is devised to assist the analysis and the presentation of the theory.

Table 16 - Glossary of terms used in grounded theory

| Term used in grounded theory | Explanation/ definition |
|-------------------------------------|--|
| Phenomena | Central ideas in the data |
| Theoretical sampling | The selection of study participants so that a range of responses are elicited |
| Saturation | The point in data collection or analysis where no new information is produced |
| Open coding | Conceptualising the data to form concepts and categories, and to identify the properties and dimensions of these |
| Concept | Unit of abstracted information |
| Category | A group of concepts |
| Axial coding | Making relations between categories, thereby identifying structures and processes |
| Causal conditions | Conditions that positively influence a phenomenon |
| Intervening conditions | Conditions that negatively influence a phenomenon |
| Consequences | Outcomes of actions |
| Selective coding | Identifying a central category and refining the theory |

Sources: (Creswell 1998; Strauss and Corbin 1998)

13.2.4 Assessing the quality of qualitative research

Assessing the quality, rigour or validity of qualitative research has been a concern for many researchers, and a consensus has not been achieved (Dreher 1994; Gifford 1996; Creswell 1998; Gifford 1998; Mays and Pope 2000; Malterud 2001). Some authors reject outright the notion that qualitative research should be assessed by means akin to quantitative research (which concerns itself with internal validity, external validity, reliability and objectivity). Creswell argued that rather than striving for the

inappropriate goal of validity, qualitative researchers should aim to create an understanding of a phenomenon (Creswell 1998). Other authors argue that qualitative research should “conform to basic scientific principles and that explanations generated must be open to refutation” (Dreher 1994). I have adopted the stance of Mays and Pope: that qualitative research can be critically evaluated, but the criteria for quality differ from those used for quantitative research (Mays and Pope 2000). They suggest the following strategies for ensuring the quality of research:

1. “**Triangulation**”, a term taken from surveying, meaning looking at one point from a number of perspectives. This could include data source triangulation (using different kinds of data), researcher triangulation (having more than one researcher, particularly useful if the researchers hold differing perspectives), methods triangulation (using different methods to obtain the data) and theory triangulation (using alternative explanatory frameworks to analyse the data) (Gifford 1996).
2. **Respondent validation**: checking the researcher’s account with the research participants in order to reduce error.
3. **Transparency of methodology**: the methods of inquiry, data collection and analysis must be clearly explained so that the reader can assess the appropriateness of the methodology for the issue at hand. Unlike in quantitative research where methodologies can be ranked in terms of the validity of the results they produce, in qualitative research the variety of methodologies each have their own strengths, and should be selected appropriately (Creswell 1998).

4. **Reflexivity** entails that the researcher is aware of the paradigm within which he or she works, and acknowledges the effect that his or her own beliefs have on the collection and analysis of the data.
5. **Attention to negative cases** (known in quantitative terminology as ‘outliers’): instead of discarding data that doesn’t fit the emerging theory, these cases should be explored further in order to enrich understanding of the issues, and expand the theory.
6. **‘Fair dealing’** focuses on the sampling methods used, and calls the researcher to task to ensure that a range of responses is elicited.
7. **Relevance, usefulness and general interest** should guide the conception and design of the project.

Transferability is a particular problem in qualitative research. One strength of qualitative research is its ability to enhance the understanding of an issue in its lived context. The assumption that context is important limits the extent to which any results can be generalised. Yet, as with quantitative research, with adequate description of the context, sampling and methods, the reader should be able to make an assessment of the relevance of the results to his or her own setting.

13.3 Challenges for qualitative researchers working with Aboriginal people

13.3.1 Consent

Whilst required by many ethics boards, the value of information sheets and consent forms in a society with limited literacy is unclear. Smith, working in a Northeast

Arnhem Land Aboriginal community, found that obtaining written as opposed to verbal consent was problematic, leading participants to become suspicious of potential exploitation, as described below.

Many respondents who were neither English nor *Yolngu-matha* literate appeared both anxious and embarrassed when asked to sign the [consent] form. Others seemed to have had negative experiences in the past with signing ‘bits of paper’ and expressed concern about potentially having their name ‘put up on the wall at a conference’ or ‘being taken to court’ as a consequence of signing the form (Smith 2002).

13.3.2 Intent

The suspicion with which consent papers were regarded reflected concerns regarding the intent of the researcher. The researcher may be perceived as representing a government organization (such as ‘welfare’) with the power to intervene in people’s lives (Smith 2002). Whilst researchers are not in the habit of stealing children, their work can at times be seen as exploitative, with the primary aim being the adornment of the researcher’s academic career, but yielding little benefit for the people being scrutinised (Smith 2002); (Anderson 1996; Leeder 1998). It is important that the research should be seen as having benefit to the community. This can be through the engagement of community members in the research, or through the project outcomes. It is important to note that there are differences in what Aboriginal people and the researchers may regard as a benefit (Anderson 1996). Delayed or future benefits may hold little value for Aboriginal people, who have experienced the ‘empty promises’ of researchers in the past (Nathan and Japanangka 1983).

13.3.3 Communication

Communication is a major issue for researchers working in a cross-cultural context, especially when conducting group interviews. Beyond the not-so-simple requirement of having a good command of the local language, it is important to have an understanding of the local rules of communication. The questioning style of interview-based research is problematic when applied in an Aboriginal context. Questions may be seen as manipulative and inappropriate if the respondent feels that the interview is a knowledge test (Nathan and Japanangka 1983). Furthermore, for Yolngu questions are rude, as explained by a *Yolngu matha* translation team:

Knowledge is owned. It is not just for anyone. If you want knowledge, you must sit respectfully with those who own that knowledge. When they believe you are ready for it, they will give it to you. You must not ask for it.

(McLellan and team)

The following recommendations made by Donovan (Donovan and Spark 1997) and Morgan (Morgan, Slade et al. 1997) address these problems:

1. Avoid direct questioning, or acknowledge the discomfort caused by this means of interaction. Alternately, engage in an interchange of information.
2. Use an interviewer who is already known to the respondents.
3. Respect respondents' privacy, the right not to answer questions and the practice of not speaking for others.

4. Be conversant in the local language, use an interpreter, or have a good understanding of Aboriginal English.
5. Avoid questions that require a numerical response.
6. Recognise the differences between Aboriginal and non-Aboriginal concepts of time.
7. Be aware of communication mores such as delayed responses, avoiding prolonged eye contact, group acquiescence, and the use of sign language.

Whilst a community member is often a crucial part of the research team, invaluable in gaining the trust and facilitating the participation of the respondents, this can also cause some problems. Some Aboriginal researchers have experienced discomfort due to transgressing cultural communication mores during interviews (Devitt and McMasters 1998; Smith 2002; Nathan and Japanangka 1983). In other situations, respondents may not feel comfortable talking to people they know and interact with socially (Sibthorpe, Bailie et al. 2002).

Finally, the use of an interpreter, and the requirement for the translation of interviews produce a multitude of opportunities for misunderstandings and lost opportunities for enriching the interview, in addition to being time-consuming and expensive (Devitt and McMasters 1998; Smith 2002).

13.3.4 Cultural mores

The social identity of research participants has the potential to affect their interaction with researchers. Smith found many young Aboriginal people to be shy and difficult to engage in interviews. Similarly, men were reticent at times to be interviewed by

women. Further biases could arise from the use of kinship ties to select study participants (Smith 2002). On the other hand, kinship can be a strong motivator for participation in research, and some researchers acknowledge the usefulness of a pre-existing relationship with the community in facilitating their research (Hecker 1997). Reid described the impact of her own social identity within the community on what information she received:

First, what a person told me about an illness or death depended on the speaker's relationship to me, the event itself and the people involved. I was perceived as a member of a particular family and clan for some purposes, and as a non-Aborigine and outsider for others...

Second, what I was told depended on what I had a right to know and what the speaker had a right to say ... What one knows and can say depends on one's family and clan membership, birth order, marital status, child-bearing status, sex, authority and age...

(Third) once I came to be regarded as a familiar and reliable person, I gained the right to know whatever I saw, whatever I was invited to take part in, or what ever most women (my age) would be told. (Reid 1983)

Similarly, Nathan had to establish her credibility with the participants in her research, in ways that surprised her, unable to rest on the laurels of her academic achievements.

(Learning bush skills) allowed Dick (Nathan's co-worker) to introduce me in a way credible to himself and his people. Introductions were prefaced in the vernacular with 'This is *Naparula* (skin name), who is doing some work for

congress and who is all right because she speaks some Anmatjirra, eats kangaroo, can shoot a perentie, and camps in creek beds...’ (Nathan and Japanangka 1983)

13.3.5 Choosing a co-worker

Reid and Nathan described their preconceptions of an appropriate co-researcher and how this changed after time in the field. Both had assumed that young, professional women with a good knowledge of English would be ideal. Later they recognised the value of older, respected community members, with or without good English, but with fluency in local languages. The appropriate Aboriginal co-researcher ensured the credibility of the non-Aboriginal researcher and the project itself (Nathan and Japanangka 1983; Reid 1983).

14 Methods

14.1 The research origins

The impetus for this research came from physicians concerned about the increasing rate of recurrent RF in the Top End, and the lack of a shared understanding between patients and doctors as to the value of the prophylactic penicillin injections. The phenomenon for study was defined as “the care of people with RHD, with a focus on the secondary prevention of RF”. A secondary aim was to understand patients’ perceptions and lived experiences of RF, and the Yolngu explanation of its cause.

Funding was obtained from a grant from the Department of Health and Ageing Regional Health Services Program. The project was approved by the Joint Institutional Ethics Committee of the Royal Darwin Hospital and the Menzies School of Health Research.

14.2 Community selection and support

Galiwin’ku community (Elcho Island) was chosen for this study partly for reasons of convenience, but also because of the high incidence of RHD as well as the degree of community support for the project. Members of the Galiwin’ku community accepted the research problem as real and appropriate for investigation. Support was obtained from the community health services, the town council and the *Yalu marngithinyaraw* (Yalu nurturing centre), a locally constituted organisation that (along with other roles) mediates between the community and outside researchers. People with RHD and family members also showed support.

14.3 The researchers

As described in chapter 13, the social identity of a qualitative researcher bears some effects on the research project, and should be acknowledged. As principal researcher and author of this thesis, I describe myself as an Australian woman with a European heritage. I am a medical doctor and my partner was the resident general practitioner for the community. I held some responsibility for the clinical care of patients with RHD prior to the commencement of this project. In the process of clinical duties, and whilst conducting the research described in part one of this thesis, I had met most of the patients with RHD in the community. I had spent time performing patient and family education relating to RHD, in English and in *Yolngu matha*. An identity as a doctor interested in the ‘heart disease patients’ was clearly established prior to the commencement of this project.

I sought a local co-researcher. A number of potential candidates showed initial interest but withdrew for personal reasons. Joy Bulkanhawuy was recommended by Yalu and accepted the position of co-worker on this research project. Joy grew up in Galiwin’ku, but had also lived for a considerable length of time in Darwin. Joy speaks Djambarrpuyngu, the most widely spoken of the languages in Galiwin’ku. She is a member of the large and powerful Dhamarrandji family. Joy had previously held positions as a health worker as well as hospital interpreter and had previous research experience. By Yolngu kinship terms she is my *dhuway*, meaning cousin and sister-in-law.

14.4 Sampling

Patients with RHD or a past history of RF were approached either directly or through the health centre. Participants were selected to produce a range of ages, sexes, clan affiliations, clinical severity and level of compliance - as perceived by health care providers (theoretical sampling). Patients living in outstations (small settlements beyond the main community) as well as patients who had relocated to Darwin but were visiting the community were interviewed. Family members were encouraged to participate; in some cases, family members were approached independently. All the health staff working in the community were interviewed, including the visiting physician. Only two subjects (both patients) directly declined to participate. A number of community members initially agreed to participate but repeatedly failed to be available at interview times, probably indicating their underlying dissent. Whilst we were disappointed by their non-participation in the study, we were also reassured that they felt free to refuse to participate, which might imply that the consent of others was similarly freely given.

In all, fifty people were interviewed in twenty-three interviews (see table 18). Fifteen patients, eighteen relatives and seventeen health staff were interviewed. This constitutes approximately half of the patients on the island on the RHD register. Participants ranged in age from approximately twenty years to sixty years. Twelve clans were represented. Only six interviewees were male: four male health staff, one patient and one relative, probably indicating the cultural hesitation of discussing health matters with members of the opposite sex. This was an issue particularly for Joy who had to recognise Yolngu kinship laws which dictate what can properly be discussed between people, especially those of the opposite sex.

Active respondents in an interview ranged from one to five people, but there were often a number of silent participants. Groups were self-constituted; attempts by the researchers to convene focus groups were generally unsuccessful. The main contributors in interviews were the patients and their mothers. In fact, it was seen as mildly inappropriate to interview a young person in the absence of their guardians, even when that person was of adult age. Thus for practical reasons, interviews were conducted with informal, family-based groups, which did not fulfil the definition of focus groups, but have been used by other researchers working with Indigenous communities (Devitt and McMasters 1998; Smith 2002).

We purposely sampled the population of RHD patients and their families to elicit a variety of experiences, beliefs and behaviours (“theoretical sampling”). Thus whilst certain themes and points recurred, true *saturation* of data was not achieved, and may have required a much larger study group (see section 13.2.3).

Table 17 - Numbers of persons interviewed according to category and sex

| Participant category | Female | Male | Total |
|---|------------------|-----------------|------------------|
| Patients with RHD or a history of RF | 14 | 1 | 15 |
| Relatives | 17 | 1 | 18 |
| Health workers | 8 | 1 | 9 |
| Nurses | 6 | 1 | 7 |
| Doctors | 0 | 2 | 2 |
| <i>Total</i> | <i>45</i> | <i>6</i> | <i>51</i> |

14.5 Interviews

The project was explained in English and *Yolngu matha* and written consent was obtained. Participants were given the choice of being interviewed at home or at the home of the principal researcher. When the interview occurred at the researcher's house there was difficulty in engaging other family members. On the positive side however, this provided a space away from other distractions and allowed for a more confidential interview.

Yolngu matha and English were used interchangeably during the interviews, which were structured around a theme list. Interview plans were modified according to whether a community member or a health staff member was being interviewed. The two researchers in collaboration developed the themes and specific prompting questions, with attention to cultural appropriateness and individual contexts. The questions were revised throughout the research process to develop emerging themes and to curtail unfruitful enquiries. The themes in their latest formulation were:

- Getting sick with RF: tell me about the time when you (your relative) got sick.
- Who cares for people with RHD, and how?
- Who makes the important decisions about health?
- What is the best way of looking after people with RHD?
- Tell us about ...
 - The needles
 - Tablets and operations
 - The health centre
- What causes this disease?

- What is the best way to find out more about this disease?
- What ideas do you have about looking after patients with RHD?

We initially planned to ask demographic questions relating to educational status, the number of people living in the household and the person's clan affiliations, but these questions were seen as socially clumsy and instead of 'breaking the ice' led to an uncomfortable start and were thus quickly deleted. Two other areas of enquiry appeared fruitless, but the lack of response they elicited was in itself informative. The question "what do Yolngu people think about RHD?" or "what is the true story (*yuwalk dhäwu*) about RHD?" produced silence. My co-researcher eventually rephrased the question to ask whether there were any traditional therapies for heart disease, breathlessness or palpitations. After several negative replies, the subject was eventually dropped. Another second area that I wished to pursue, but that mostly led to silence or curt closure, was the subject of death and the risk of dying with RHD.

Interviews were recorded, translated then transcribed in English. When participants declined to be recorded (three cases) the interviews were transcribed directly as translated. The translation process allowed the identification of key terms, and a fruitful discussion of the possible meaning of the interview content from a Yolngu perspective. The two researchers discussed interpretations and implications of the interview data, thereby paving the way for the analysis.

In addition to the interview data, informal observations during my working life (as a clinician, researcher and community member) were noted in a journal. Minutes were kept of meeting with my advisory panel from the *Yalu Marngithinyaraw*. These data

were treated in exactly the same way as the interview data - coded, categorised and built into theory – and thus my own observations are integrated into the final conceptualisation.

14.6 Analysis – grounded theory with modifications

The interview data were analysed according to the principles of grounded theory (Strauss and Corbin 1998) (Creswell 1998). Grounded theory was chosen as a methodology as it is suited to an exploratory investigation. In principle it allows theory to emerge from the data and does not pre-empt either the structure or the results. Non-Indigenous researchers occasionally find themselves at odds with their Indigenous research subjects, having fundamentally different approaches to the phenomenon of interest.³ By using grounded theory I hope to reduce my own influence on the direction of enquiry – a difficult task.

Interviews were initially coded according to broad phenomena observed. These were:

- Getting sick
- Staying well/ looking after people with RHD
 - Role of self (looking after yourself)
 - Role of family
 - Role of the health services
 - The best way of looking after people with RHD

³ Smith discusses this problem in her study on childhood growth in a neighbouring community. In this case, the researcher was concerned primarily with the physical growth of children, whilst the Aboriginal participants in the study were focussed on the personal and social development of the children Smith, D. (2002). Community Development and indigenous Health (thesis). Department of Public Health; Faculty of Medicine. Adelaide, Flinders University: 357..

- Problems with care
 - Not looking after yourself
 - Problems with the family (not looking after their relative)
 - Problems with the health services
- The logistics and role of the injections
- Communication issues
- Knowledge and understanding of the disease
- Taking responsibility for health
- Religious beliefs

Interviews were dissected and rearranged into thematic documents based on these phenomena, keeping track of the origins of each interview excerpt. Within these thematic documents the data were analysed line-by-line for concepts, which then formed the basis for more general categories (open coding). Categories were organised into processes and structures in an attempt to explain the phenomena observed (axial coding). In this way theory was built from the ‘ground’ of the interview data (selective coding). The principal researcher performed the analysis, but the co-researcher was involved at each stage in order to assess the authenticity of the conclusions, as a means of triangulation. Phenomena, categories and concepts were revised throughout the analysis process in order to make the most cohesive theory from the data.

14.7 Audit of penicillin prophylaxis coverage

The medical records of patients with RHD in Galiwin’ku were audited for the number of penicillin injections received from January 2002 through to September 2003

inclusive, a total of 21 months.. Bicillin coverage was calculated as a percentage of prescribed injections administered per patient, and per month. This audit was not intended to represent a definitive description of the ‘actual’ situation to be compared with the ‘subjective’ viewpoint of the patients and staff. The clinical data is likely to have limitations in its accuracy and the lack of documentation for patients travelling outside the community. However, the audit contributes to the information obtained through the interviews and adds another perspective on the same issue (triangulation of methods). It also provides a point of comparison with international data on Bicillin coverage.

14.8 Community meeting

The research plan included a meeting to feedback the preliminary results of the interviews to the community, to achieve respondent validation. It was hoped that an open discussion would ensue and that a series of recommendations would be formulated outlining the preferred ways of looking after patients with RHD. This idea grew from the suggestion of Humphery et al. of convening a reform group, which should be self-constituted, although institutionally supported, and would be a means to “bring about change which is specific, local, small-scale and, above all achievable and effective” (Humphery and Weeramanthri 2001). A practical outcome was sought as a way of delivering some benefit to the people who had contributed as subjects.

15 Primary results (open coding)

The results reported in the following chapters are derived from the analysis of the interviews. People with a past history of RF or RHD will be called ‘patients’.

‘Relatives’ refers to any person recognising kinship connections with the patient.

‘Health worker’ refers to Aboriginal health workers. ‘Interviewer’ refers to either of the two researchers, Zinta Harrington (ZH) or Joy Bulkanhawuy (JB). ‘Yolngu’ will be taken as meaning Aboriginal person from the area, and ‘*Balanda*’ the local term for non-Aboriginal person.

In this chapter I present a description of Yolngu patients’ experiences of RF and RHD. The responses of patients, family members and health staff as to the ways of looking after people with RHD as well as the failures of care are described.

15.1 Stories of getting sick

Descriptions of how people became sick were mostly distant recollections of a childhood illness. Symptoms recalled by these young patients commonly included breathlessness, palpitations, unusual body movements, swollen joints or body aches, weakness, cough, weight loss, blackouts, fever and chest pain. Those with medical training would recognise the key features of RF in these descriptions:

Relative Everything was swollen; his face and body swelled up. When he got here I saw him doing a little bit of funny actions, like he was shaking.
(Interview 10)

Patient The sign: that time when I was bleeding through the mouth

Relative And she was swollen up...

Patient ...feet, ankles and knees. When I first got sick I was getting skinny. I was walking only a little way and I got breathless. (Interview 15)

Another account of the same patient's illness is quite different, and more difficult to recognise as RF:

Relative First I'll tell you the story when she was a little girl, 'round about, I don't know how old she was, she was dying with this disease. When she was only school age and she didn't know she had this heart disease, when she was lying there dying, we had a prayer for her then she woke up. After that she was walking around alright. Now she is a mother, and this sickness came back again... She was playing all the sports. She got stiff, and she lies still for a long time... We went hunting to get some fruits and she got the same again. Her body got all stiff and we thought she was looking at an ant. But she couldn't even move, although her eyes were open! I don't know how many minutes she was like that. Then she woke up. (Interview 15)

It is important to emphasise that this description is not 'wrong', even though it differs from the account given by the patient herself, as well as a medical definition of the disease. Understanding the speaker's perspective and language brings some light to the subject. Yolngu may define symptoms more in terms of loss of function than actual bodily sensations. So "she lies still for a long time" tells us that the patient was unwell, without revealing specific details. Whether pain, breathlessness, paralysis or loss of consciousness was the cause, we don't know from this account. Likewise the phrase "walking around alright" is used frequently to mean that a person is quite well. Furthermore, in translating the original *Yolngu matha*, the precise meanings of some of the key terms are lost, and substituted with best-match English words.

For some people the acute illness wasn't diagnosed in childhood and the condition of RHD was detected as an adult. Adults presented along a clinical spectrum from mild chronic symptoms to dramatic acute presentations, such as the episode of acute pulmonary oedema described in the following quote.

Relative I saw her when she was lying down and I saw blood coming from her mouth. Then I called out "what happened to you?" Her aunt said, "She's sick, she's short of breath." Then we went to the clinic. She hopped out [of the car]. Then when I took her inside we found out she had blood everywhere in her body. She was crying out, "I'll die, I'll die". (Interview 12)

The suddenness and unexpectedness of the illness was frequently remarked upon. The possibility of sorcery being involved in disease causation was not directly mentioned, although this last account echoes the description of the action of *galika* (see section 12.2.1).

15.2 Good health - a cooperative approach between the patient, the family and the clinic

15.2.1 Looking after yourself

As suggested above, being healthy was described in terms of what you can do rather than how you feel.

Patient I'm walking around normal, alright. I am working hard, lifting up heavy things, I carry damper [a 10kg tin of flour] from the shop to home, so I do more exercise helping my heart. Sometimes I work at home, in the garden, inside the house. (Interview 5)

Relative He goes hunting, he hunts and he doesn't get sick. He chopped down trees for mangrove worms!

Interviewer Everything is alright?

Relative Yes. He doesn't get sick or lame any more. He has three kids now. (Interview 14)

Many family members had opinions on how patients should look after themselves so they could “walk around normal”. People with heart disease should exercise, eat well, not smoke, care about their health and body, and maintain good relations with the clinic and their family. Whilst the healthy lifestyle message (exercise, good food, not smoking) had undertones of public health messages to reduce ischaemic heart disease, a preoccupation with food has a strong cultural basis. A *Balanda* with more than twenty years of experience in the community described a ‘Yolngu stomach-centred world’ (Journal 10/6/03). This is intuitively valid where nourishment was a daily concern until ready access was procured through the establishment of a store. Even today, whilst food is readily procurable, its expense limits the availability and variety. Thus tea and damper is the staple diet for many Yolngu. Furthermore the distribution of food within a household and over the pay period may be erratic.

Food was described as the cause as well as the remedy of disease (see section 16.2.1). Getting enough food was a prime concern, and eating well was synonymous with health. ‘Bush food’ was described as ideal, and has special properties. In Galiwin’ku the most commonly hunted foods are fish, turtle, shellfish, dugong, wallaby, goose, yams and bush fruit. Shellfish has a fundamental place in the Yolngu diet – it is a weaning food, a tonic for the sick, and provides nourishment for invalids, according many respondents.

Relative ... shellfish builds your body and also your blood into your heart. It’s healthy. Otherwise if you don’t eat fresh shellfish, no fresh blood gets in, because you can’t get blood into your body with nothing. (Interview 14)

Balanda foods were said to be unhealthy if oily, sweet, or bought at he take-away, but porridge, vegetables, drinks with no sugar were all felt to be healthy. A combination of fresh seafood and store-bought vegetables was said to be a healthy diet.

Smoking was universally said to be unhealthy, and the cause of a deterioration in health.⁴ A moderate level of activity was desirable, according to health workers and family, but hard physical work should be avoided. Exercise was believed to be good for patients with heart disease, and this could include organised sport or hunting.

Relatives and health workers stated that patients should show due concern for their health: they should ‘think really hard’ about their health and they should ‘feel’ their

⁴ In other conversations I have heard smoking described as a Yolngu tradition. It was introduced to the Yolngu by Macassan traders. Older Yolngu speak of smoking bush tobacco, but this tradition appears to have vanished.

body.⁵ Careful contemplation of the body was associated with caring for the body properly, as in this account:

Health worker One man refused [his injections] lots of times. Then we give up, and later he started thinking, and then he came back to see Marthakal.⁶

Interviewer What changed his mind do you think?

Health worker I don't know. He *felt* his heart, he *thought really hard*, then he went [to see the health workers]. (Interview 22, my italics)

This concern for health may come from past experiences of serious health events, or may be associated with older age and maturity, according to health staff. It may also be the result of the patient's growing status in the community and a sense of responsibility to others, especially the patient's children. Taking responsibility for health will be discussed in more detail in section 8.3.

A good working relationship with the clinic staff, and a good understanding of the disease were important according to health staff. Patients were encouraged by family not to be shy or afraid of clinic staff or treatments. Health staff denied any problems with the current patients. A positive picture was painted of staff and patient relationships in almost all cases. Staff denied any difficulties administering the monthly penicillin injections.

⁵ 'Feel' the body rather than 'listen' to it, as English speaker would commonly say.

⁶ Marthakal is the administrative body for the homelands, and has its own health service staffed by nurses and health workers.

15.2.2 The role of the family in looking after patients with rheumatic heart disease

The family is intricately involved in looking after any Yolngu patient with RHD. Its role is often described by the words *djāga* (or *djāka*) meaning to care for, or look after, and *gunḡa'yun* meaning to encourage, or assist. The mother has particular responsibilities, but is not necessarily the person with the authority to make the important decisions for a patient; decisions are typically made by the most senior of the close kin.

An aside is warranted to introduce the Yolngu kinship system to those unfamiliar with it. Children call their birth mother as well as all her sisters *ḡāḡdi*. Likewise, children call their father, and all his brothers *bāpa*, their maternal grandmother and her sisters are *māri*, and the cousins of each lineage are *wawa* (brother) and *yapa* (sister). There are particular social and ceremonial ties between a person and their maternal uncle (*ḡapipi*) and their maternal grandmother and her brothers (*māri*) (Christie 2001).

Understanding Yolngu kinship is a good way of recognising the differences of Yolngu and *Balanda* paradigms. For example, in the Yolngu kinship system the word *waku* is used to signify a woman's children, but also a man's sister's children, a woman's father-in-law but a man's brother-in-law's father, and the maternal great grandmother of both women and men! The sense in this taxonomy comes from the systems of moieties, clans, skin groups and appropriate marriages.⁷

⁷ In the Yolngu ontology, everything animate and inanimate, including people are classified as belonging to one of the two moieties: Dhuwa and Yirritja. Clans are the descendants of one man. There are eight skin groups, each with a male and a female version, leading to sixteen skin names in total. These are determined by the mother's line, and change with each generation in a set pattern. Skin groups are one of the factors that dictate an appropriate marriage. An example of an ideal partnering is between a woman and her cousin on her father's side.

Many people are involved in caring for a patient with RHD. Although the specific people vary from family to family, important figures include the parents (*ṅāṅdi*, *bāpa*), the maternal grandparents (*māri*, *ṅathi*), the husband or wife (*dhuway* or *galay*), sisters (for female patients only, *yapa*), paternal grandmother (*momu*), and paternal aunt (*mukul bāpa*). For male patients the female relatives were still the most prominently involved in their care (mother, paternal aunts) but it is likely that if more men were canvassed, then brothers (*wawa*) and maternal uncles (*ṅapipi*) would also be recognised. It is likely that these people would remain relevant in other conditions, with the important proviso that men are excluded from discussing female sexuality and reproductive matters.

Family members work together and apparently exchange roles quite freely. The main requirement they have of each other is good communication.

Relative His aunty (*mukul bāpa*) always looked after him. We (the mothers) didn't take him to the clinic, but we got the story from the aunty. (Interview 10)

'The mothers' may form a seamless team in caring for their 'child', but one woman, either the birth mother or her oldest sister, takes on the primary role. She will often be the one accompanying the young child to hospital, the main carer, and the coordinator of the family support. One respondent vehemently claimed that the birth mother had the greatest right and authority. This opinion was unusual and may have been influenced by his particular relationship with this patient's mother and the subsequent family antagonism.

Relative [The mother] she's struggling for [the patient's] good, for her life ... [I said to the patient] "Your mother is important. No one else is important to you, only your mother is important, because she gave birth to you, she took you to Adelaide [for heart surgery], and she is caring for you, and she has every story from the doctor and she will care for you. The other people can't look after you. Only the mother has the right." (Interview 13)

Another mother gives a very poignant description of her role in looking after her son with RHD. This young man was discussed in several interviews, due to the difficulties family and staff had in looking after him. Unfortunately we were not able to secure an interview with him, at least in part because of the reluctance of the local researcher (JB) in approaching this man, possibly due to concerns about violent repercussions. This mother focused on the concept of encouragement to describe the care she gave her son.

Relative But today I am just encouraging him more, especially his mind and his body. Supporting him: like how he can eat food, brush his teeth, have a shower, before it is time for his operation. So this time I am just encouraging him more closely. Even when he feels upset, I just keep talking. Sometimes he is just thinking about walking away and smashing me, leaving me. But I always said, "if you leave me [its no good], it's the only hope that you have, it's your mum, right here". So for me, as a mother, it was very, very hard, because we struggled all the way. And that struggle with him, was when he was rejecting his medications, and my job was to talk to him about the

medications and the operation. And finally he has made the right decision.

(Interview 18)

Gunga'yun, or encouragement, is a central concept in the care of a young person. It implies supporting a person to hold their own convictions, make life decisions, take on adult roles and speak with authority. It encompasses the notion of raising a child into adulthood, all the while respecting their autonomy. It goes along with other features of care, such as the physical care and assistance involved in looking after a child (*djaka*), the concern for the health of the child, and the guidance and advice that a parent gives a child. Despite its abstract meaning, *gunga'yun* is frequently used without elaboration. For example, when asked hypothetically how he would look after a child with RHD, a patient replied: "I would strongly encourage him, all the time" (Interview16).

Djaka was the term used to indicate the process 'to care for' or 'to help'. Some respondents were unwilling to elaborate on the concept. This may have indicated their reluctance to discuss what they perceived as obvious, or what may leave them open to criticism. More concrete responses described good care as involving providing good food, especially bush food, helping sick children physically, providing a clean environment, and facilitating good health care. Value was placed on having a close and trusting relationship with the patient, and acting as a confidant. Carers should also provide advice, ranging from the mundane issues of daily living to existential levels.

Relative I helped her more: telling her to stop smoking, not to work too hard, eat more food, stop smoking. ‘That’s how you are killing yourself, your heart. When you eat good food, then everything is healthy, your heart.’ (Interview 7)

Relative That’s what I explained to him, about how to die, and how to live. That’s why he finally came up with good feelings. (Interview 18)

Close supervision is considered to be a crucial part of good care, and concerned mothers will ask their children not to leave the home. A simple explanation for this restriction is that it allows the parent to watch out for the physical wellbeing of the child and monitor the child’s behaviours. Another purpose of this close supervision may be to protect from sorcery (*galika*) that might occur when walking away from the home at night.⁸

An important responsibility for the family members that was described was to participate in decisions relating to treatment choices. The head of the family usually took on this role, and their decisions were rarely disputed. A relative who is confident and conversant in English may also take on this role. However, the patient retains the final right to accept or refuse treatment. Patient refusal to heed advice was a source of stress for carers. In the event of an adverse outcome, families could still take the blame (see section 16.4).

Relative Like this girl, she was sick. We asked her, tried to help her. She didn’t listen. When she gets very sick we can ask you [the health staff] to

come. And after that we can find out what is happening, whether the health centre is wrong, or the family. (Interview 16)

The significant role that the family played in looking after patients was recognised by the local doctor, who commented:

Doctor For Yolngu, kinship is everything. Without seeing that person within that kinship context, it's always going to be a great challenge [to provide medical care]. (Interview 2)

By contrast, the visiting specialist and the clinic nurses made no mention of the role of family. The health workers, mediating between the two paradigms are likely to hold strong views on the role of family, but gave little indication of this in the interviews.

There is some indication of support for patients outside the immediate family. Some families affected by RHD offered advice and support to families similarly afflicted. Other sources of support for patients included teachers, community organisations, the church, and employers. One respondent identified child health as a *community* concern.

Relative The time when you came, I was really excited to see you. Because the community are looking forward to see people who would get together and help each other, looking after the kids. Because a lot of the kids are sick and we don't know how to look after the kids, give them medicine and look after

⁸ Concerns about sorcery are still widely held. In the context of my role as a doctor, people have clearly

them. Because health is important, because health has got everything right for the kids. (Interview 13)

This was quite uncommon, however. Most respondents restricted their interest to their own family, either out of a sense of priority and loyalty, or alternatively, in respect for others' privacy or autonomy.

15.2.3 The role of the clinic in looking after patients with rheumatic heart disease

Interviewer (JB to ZH) Here in Galiwin'ku, when you look around, the health centre, it is always right there; the health workers, they are everywhere. (Interview 19)

Health worker The people who are sick, they know where the health centre is. They know that health centre is a place where they can come and get their medicine to make them feel better. (Interview 22)

These quotes, describing the ease with which the clinic and health workers could be accessed in Galiwin'ku, illustrate an important point. The clinic is centrally located, and many respondents portrayed the sense of comfort they felt in going to the health centre for their medical care. There was significant potential for bias in this regard, as the patients interviewed had all received considerable medical input. In addition, as the interview team had associations with the clinic, respondents may have withheld negative or critical responses. However, as section 15.3.3 will attest, people did report

stated their unwillingness to walk half a block to my house at night time for fear of *galka*.

problems they had with the health centre, so the effects of biased sampling and the social identities of the interviewers did not silence all critical voices.

The residents of Galiwin'ku may well value the fact that their health centre is community-run, but they didn't make any mention of it. Rather than the health centre belonging to the people, the residents of Galiwin'ku 'belonged' to the health centre, according to one respondent, in the sense that the health centre was the steward for that community's health. The implication was of a responsibility for, rather than authority over people.

It was apparent that the well-trained, local health workers were considered to be a great asset to the clinic. There were nine health workers employed at Ngalkanbuy Health centre, and two at Marthakal homelands health service. Health workers lived in all of the four major areas (camps) of Galiwin'ku. A number of families were represented, although one clan predominated at Ngalkanbuy. Clan or family affiliations were not said to interfere with the professionalism of the health workers.

Health workers, unlike *Balanda* staff, knew the families and patients over many years, and therefore had a long-term perspective on the patients' conditions. Most patients were familiar socially with at least one health worker, and this facilitated clinical interactions. Health workers were never 'off-duty' when it came to looking after their family members. Sharing the patient's culture and values, health workers enacted the principles of encouragement (*gunḡa'yun*) and care (*djāka*) for the patient:

Relative Now they [the health workers] are caring alright for her [the patient]. Because all the time [when] she sees the new moon she goes down to the health centre and they help her. “This is the girl who’s got this [RHD]. She came at the right time. She is due this injection,” [they say]. The health worker told her not to miss her injection, “Come all the time.” (Interview 11)

Maintaining kinship protocols that restrict interactions between some relatives did not seem to pose a major problem in most cases:

Health worker We can help him quickly: if we are not affected by the skin relation, or our brother, we can treat them. (Interview 23)

This statement is further qualified, in that cultural laws only restrict some forms of interaction. A health worker can give out tablets to her brother, for example, but avoids contact with his blood, and therefore cannot give an injection to him.

Ultimately, not only are the health workers integrated within the community socially and culturally, but they share the physical conditions of the patients too. On one occasion, where a health worker and I were examining a patient with RHD, the health worker demonstrated her own murmur and discussed its management, ostensibly as a way of educating the patient (Journal entry 7/4/03).

A doctor and five nurses also work at Ngalkanbuy health centre. A visiting specialist attends from Darwin every three months. The doctors and nurses occupy a spectrum of social positions, from friend or kin (when ‘adopted’ into a local family), to

nameless service provider, to visiting expert. This was partly determined by the personal approach taken by each of the *Balanda* staff, and partly by the historical and social context of colonisation. *Balanda* staff expressed a wish to be seen as approachable, even to be friendly with patients, at the same time limiting the level of intimacy of the interaction.⁹

Patients and relatives expected the following services from the health centre: making the right diagnosis, communicating with the patient and family, performing check-ups, treating patients appropriately, engaging in disease prevention, and educating staff as well as the community about the disease. When necessary, health centre staff referred patients for investigations, specialist reviews, for hospital admission and for operations. Maintaining an accurate medical record was another important role of health centre staff.

Home visits by health workers, nurses and doctors were valued in that not only did the patient receive medical care in a familiar, comfortable environment, but also the extended family had the opportunity to be involved in the patient's care. Home visits seemed to indicate that the health service really cared for the community, in a nurturing, comforting way. Health workers on home visits resembled caring family members more than officious professionals. One respondent connected a diminishment in home visits to worsening health outcomes in the community.

⁹ There is an active debate amongst *Balanda* residents in Aboriginal communities as to the relative merits of being 'adopted' into an Aboriginal family. Advocates say that adoption allows Aboriginal people to situate a stranger within their own paradigm, giving the adoptee an identity that has meaning to Aboriginal people. Opponents say that the system is open to manipulation, that *Balanda* typically become plagued by obligations to the extended adopted family, and that having a Yolngu identity, even if this is recognised as honorary, is an impediment to fair and equal interactions with patients.

Patient They have a lot of roles, the health workers: they've got a lot of responsibility, not just to work inside the clinic, but to come and visit the home. Like ten or fifteen years ago, the health workers used to go through the camps, so that we can feel the relationships and communication. There's two-way communication. They can come here and do regular check-ups at the camps, or we can go there, to the health centre... or they can come around to the home, so that they [the patients] can feel comfortable. Like the doctor always used to come around. After a few years it stopped [the home visits], a lot of people became sick, because of that. (Interview 16)

Patients variably appraised the treatments offered by health staff. Some patients felt that they benefited from tablets, others did not. Injections were mostly accepted as 'stronger', and able to travel around the body. (See section 16.1 for a discussion of the perceptions of *Bicillin* needles.) Operations were viewed as therapeutic, and life saving, not only by those who had experienced them, but also by those anticipating an operation in the future. This in no way obviated the fear associated with the operation, the interstate travel, and the possibility of dying away from home. It was repeatedly stated that operations needed to be carried out both early in the disease, and early in life, before it was 'too late'. This was sharply illustrated in an account by a mother who attended her daughter's cardiac surgery:

Relative She was very scared. She was shivering after the operation. She cried out, "I will die, what is wrong with me?" I was sitting down there and I told her she will be alright with this medicine. "Don't worry", I said, "because you

are a young person. If you were 21 or 30 you might die”, I said to her.

(Interview 11)

Patients and their families put a lot of emphasis on the services provided by Gove and Darwin hospitals. This is where patients get their ‘full check-up’ as opposed to the sometimes quick and rushed review at the local health centre. Royal Darwin Hospital was seen as the ultimate authority in terms of health decisions. The visiting specialist from Darwin was seen as the “heart-disease doctor”, an expert with the power to ensure good care for the patient, supervising and directing the work of the health staff at the local clinic.

The visiting specialist saw his role as predominantly clinical care. In the past he had participated in education sessions for RHD patients and their families, alongside the health workers. Patients were enticed to these ‘social evenings’ with a barbecue dinner. These events were recalled by patients, but without any overt or spontaneous enthusiasm. Yet one health worker was particularly positive in her recollections of these events, and the opportunity it created for educating the patients and was keen to repeat the event.

15.3 Poor care of patients with rheumatic heart disease

15.3.1 Patients not looking after their health

Most patients stated that they were fully compliant with treatment. Yet non-compliance was not a matter for shame for other patients who reported their own non-compliance in a refreshingly matter-of-fact fashion. This indicated that for these

patients, if not for staff, non-compliance was not a negative gesture, but one of many legitimate responses to medical advice. This sentiment reflects Humphery's unemotive explanation of non-compliance as "the material consequences of particular models and practices of health service provision, undertaken within particular institutional, political, social and cultural contexts" (Humphery and Weeramanthri 2001).

Explanations for non-compliance were offered infrequently. Some patients evaluated their prescribed treatments in personal clinical trials (sample size of one) and compared their observations to the information provided by the prescribing doctor. Reid suggests that whilst Yolngu patients may never adopt the foreign bio-medical paradigm used by their doctors, the ability of doctors to make predictions within that paradigm is respected (Reid 1983). However, if that prediction fails, then the doctor and perhaps the whole biomedical paradigm loses credibility. In the case below, the patient may well have discounted the doctor's recommendations, remaining unconvinced as to his ability to make accurate predictions.

Relative Sometimes she missed her tablets. Because I know, the doctor told me, if you miss anything [tablets] that's no good. If you have a cut, it's no good. That sort of things. When you are swollen up, it's no good. It was happening [she was missing tablets], but it was just normal ... Sometimes she had a cut, but she walked normally. Sometimes she missed tablets, but she walked normally, and I saw her.¹⁰ (Interview 13)

¹⁰ In this case the medication was an oral anticoagulant - warfarin. This relative is describing how the doctor warned about the risks of missing medication and also about excess bleeding and bruising with too much medication. The relative says, "she walked normally" to indicate that the patient was well, despite the doctor's warnings regarding missing medication.

Not believing that the illness is a chronic, persistent illness was the reason why some patients ceased their medication. Patients who felt better and looked better were able to convince themselves and their families that they had made a full recovery, and there was no need for further treatment. This pragmatic, ‘evidence-based’ approach to taking medicines was evident when patients regulated their medication according to their symptoms. In this case, the evidence is the patient’s experience, not what is published in peer-reviewed journals.

Patient With *LA Bicillin* I always get better for two months. If I get an injection one month, in May, then I am still well for June and July, until I get another needle (in) three or four months. (Interview 5)

However, many patients have discovered that their illness was indeed chronic or recurrent, and that the consequences of not complying with treatment could be quite serious. Health staff expressed their dismay at the suffering that ensued when patients deferred effective treatment, thereby compromising the eventual outcome.

Fear was suggested as a reason for non-compliance with injections, for failing to undergo medical review, and for not travelling to the city or interstate for treatment. Some patients became frightened, not by the injections, but as a result of the information provided to them about their disease, and “disappeared for a while”, avoiding all contact with the health centre (Interview 23). Conversely, when a patient embraced her disease as part of “God’s design”, health staff regarded her *lack* of fear as a problem in terms of compliance with treatment.

Poverty was implicated in some accounts of a patient's inability to care for him or herself. No car to go hunting, an intermittent supply of food, no phone, no money for a bus fare were all mentioned as impacting either on the patient's wellbeing, or on their ability to access medical services. (Few households in the community own a car or have a telephone connection, making them well below the Australian average in this regard.) One discussion led to reminiscences about a real or imagined past, when people lived 'outside', there was no pollution, and children didn't get skin sores. This was contrasted with the present situation of overcrowded houses and sick children (Interview 16).

Yolngu parents shared the concerns of their non-Aboriginal counterparts when they complained about their children smoking, eating unhealthy food, drinking alcohol or kava, playing cards and hanging out with the wrong crowd.¹¹ Whilst many preached a healthy lifestyle, few practiced it themselves. In these interviews focussing on patients with RHD, many relatives made the assumption that healthy lifestyle practices were more relevant to patients than unaffected people.

Behavioural and psychological traits had their impact on patients' compliance with treatment, particularly for adolescents. Conversely, this disease, with its onset between the ages of five and fifteen, had the potential to impact on the developing psyche of a young person, according to the local doctor. Some patients were described as immature, with the 'adolescent' characteristics of rebelliousness, irresponsibility, shyness, and excessive dependence on family members. Young people were more

often implicated in playful avoidance of treatment leading to the exasperation of some staff. Older adults were more likely to refuse treatment outright, or more subtly decline by saying that they will return later for treatment, in the “typical Yolngu non-confrontational way” (Interview 2).

Yolngu take on an adult role in society at an older age than most non-Aboriginal Australians. It was not part of this research to investigate the transition from adolescence into adulthood, but the shyness of young people was a common topic in the interviews, due to the difficulty in getting young patients to speak for themselves. Young Yolngu, up to the age of nearly thirty, may seem extraordinarily shy in a clinical context. When mothers exhorted their children to speak up, the children (in this case women in their twenties) sat quietly, and signalled for their mothers to speak for them. As a *Balanda* doctor and researcher I had thought this shyness was due primarily to the power difference inherent in the situation, as I had observed the carefree and exuberant behaviour of children in familiar contexts. However, a discussion on shyness led one mature woman to recollect her own apparently sudden transition from a person who did not even speak up within her family, to one with the courage to hold and convey her own views, finally becoming a community leader with professional achievements including public speaking before non-Aboriginal audiences. This shyness was a concern to family members. Relatives expressed their desire for patients to communicate, not only with their doctors, but also with their families and other young people with the same condition. Patients needed extra confidence to access medical services other than their local clinic. One patient claimed that shyness was the cause of not getting adequate medical care.

¹¹ Although prohibited in Galiwin'ku, alcohol is still available, as is kava. Playing cards involves long

Patient And if they get shy, they won't get any help from the doctor.

(Interview 12)

15.3.2 Families not looking after patients with rheumatic heart disease

As introduced in section 15.2.2, the family has an important and broad role in looking after the health of young people. Complaints about family members not fulfilling their obligations came from patients or from health workers. It was very uncommon for anyone to comment on the business of another family. Gossip may flourish, but interviewees deliberately obscured the names, the identifying details and specifics of the accusation. Openly criticising others (at least in front of *Balanda*) signified significant social friction. Thus in an anonymous fashion, some families were described as 'lazy', too 'busy' playing cards, or ambivalent about their children's wellbeing. Some mothers were accused of being poor parents: not using bush medicines to treat their children when they were sick. A relative expressed concern over escorts who put their own needs ahead of the patient's when accompanying them to hospital. In other instances, patients openly blamed their families for not supporting them. One mother complained that she received no help (either from family or from the clinic) in caring for her sick son. Other patients were scathing of their family's lack of physical help when they were unwell.

The families' perception of the disease and their understanding of the treatment offered, affected the extent to which they supported medical interventions. Their own relationship with the health workers and the level of trust they had for their advice

sessions of gambling with relatively large sums of money. Little attention is paid to the activities of

were also important. Poor interactions with patients' families led to health workers becoming disappointed and frustrated. This in turn caused health workers to accuse parents of not caring for their children.

Health worker They [the families] don't help us. It's their feelings towards us, the workers: when we go there then people ask, "Why, what's this [injection] for?" It's their precious child, but it's he or she who has to take that injection. They just asked us what is this for. They say, "Come back tomorrow", but they are not telling the truth. They don't even care about the body of the child, that she is sick. (Interview 22)

Rather than taking it as a criticism of the health care provided, this health worker blamed the parents for not knowing the purpose of the injections. By implication, if they were good parents, they should have known what the injection is for, and should have encouraged the child to comply.

15.3.3 Problems with the care provided by the clinic

Unlike the paucity of complaints directed at relatives, there was plenty of material depicting problems with the care provided by the clinic. This does not necessarily imply that the community was dissatisfied with the service provided, rather that it was more socially acceptable to discuss these problems. Whilst criticisms may have been politely softened due to the interviewers' association with the clinic, the vehemence of some responses indicated that this was not absolute. I will outline the general

daily living such as feeding the kids, or refilling dosette boxes with medications (personal observation).

complaints about the clinic and health services in general in this section. Issues pertaining to the RF prevention program will be covered in section 16.1.1.

Patients made numerous suggestions that the care delivered by the clinic was inadequate at times. A common complaint was that check-ups were infrequent and perfunctory. When sick patients present after hours, they are required to see the health worker first, who either treats them or refers them on for medical review urgently, or during clinic hours. Many patients claimed that care was compromised by a delay in being properly assessed and receiving definitive treatment. One young woman who felt that she had been brushed off by the clinic returned to her outstation without being medically reviewed. She next presented with life-threatening acute pulmonary oedema.

Relative The health centre, that time, they didn't help me. I always took her for a check-up. At night time I always took her, and the health workers gave her *Panadol* and told her to come back in the morning for a check-up. And I told them she has got this shortness of breath, and they told me that the *Panadol* will help, and tomorrow she will get a check-up. (Interview 12)

Patients described the staff in these instances as being 'busy', 'lazy' or 'not caring'. Much of this blame fell on the health workers, and some patients stated that they therefore preferred to see nurses or doctors directly. Thus 'caring' could be deduced as meaning a 'full check-up' as well as bringing the nurse or doctor in for a second review. In other situations, health staff were accused of incompetence, such as when there was a delay in diagnosis, or the medical records were inaccurate. Care may be

seen as substandard if the patient's management appeared to differ from that of other patients with the same condition.

The community's expectations of the health centre extended well beyond simple clinical care, to include family support, community education, and physical support such as transport and perhaps even providing food. Some of these expectations may have arisen from the history of missionary colonisation, with its food kitchens and 'round-ups' for vaccinations and other medical treatments. People who have grown up under the missionary system may still be expecting a somewhat paternalistic level of assistance. In the contemporary setting however, such a benevolent service does exist in the form of the aged care program. Community members commented that old people received food, transportation for recreational purposes and a clothes-washing service. By implication, this was set as the standard of good care that could be extended to other deserving groups, such as 'heart disease patients'. This desire for a holistic health service that would look after the well being of patients appeared to be the basis for some patients' assertion that the clinic doesn't *look after* them, although it provided their medical care.

Access to the local clinic was generally regarded as good. Transportation was often available, and the regular monthly injections were administered either at the clinic, or at the patient's home. However waiting times at the clinic could be long, and some patients opted to see a health worker instead of waiting to see the doctor, or in the worst scenario, left without receiving treatment. For one patient, the opening hours of the clinic coincided with her own working hours, preventing her attendance for medical reviews and prophylactic injections. For patients living in outstations access

to health services was generally more difficult: health services were provided on an intermittent basis and, according to some patients, were not reliable.

Just as good communication was an indicator of the quality of the service provided, poor communication by clinic staff caused serious dissatisfaction amongst relatives. The relaying of 'the story' to the right people has significance in Yolngu culture where knowledge is perhaps a stronger currency than money itself, as suggested by McLellan (McLellan and team). Relatives and health workers spoke of the sense of restriction or concealment of knowledge.

Relative When they [the health staff] hear this story [the patient's medical results], they keep it for themselves at the health centre, what's wrong with him. They don't share it with us, and not even to him. (Interview 10)

Health worker Before, no one even told these patients why she is getting all these injections and tablets, maybe. We don't know. Maybe they didn't tell the straight stories to them, until they started getting worse and worse. (Interview 23)

The doctor and nurses indeed admitted that time and resource constraints prevented comprehensive patient education, especially of the less seriously ill patients.

The Yolngu social structure, with its laws governing the interactions between kin, families and clans had a negative impact at times on health workers' ability to care for patients. Most health workers are female, and women are discouraged from having

close physical contact or discussing matters of a personal nature with their brothers (which includes a number of male cousins). Likewise male health workers avoided treating their sisters, their mothers-in-law, and their ‘poison cousins’. This put stress on the health service, as patients waited longer to see a staff member with the appropriate social connection. Yet the doctor’s assertion that the health centre was an “unfriendly place for young men”, based on these observations (Interview 2), was not confirmed by other respondents.

The extension of the family is the clan. Patients from clans that are not represented at the health centre might not feel as comfortable accessing health services. This was denied by the health workers, but supported by two observations. Firstly, one woman declined to be interviewed by us, stating that she “didn’t know us”. JB felt that although she knew the woman’s identity, they were not related and thus any social interaction between them would be superficial, if not unnatural. This was construed as the reason why the woman declined to participate in the research. If we extrapolate from this observation, then *knowing* or *being related* to a health worker might be a crucial element in a constructive relationship between patients and health staff. Secondly, one relative recounted a story from the past when a health worker complained about treating a patient who wasn’t from Galiwin’ku. The mother retorted, of course he was from Galiwin’ku, as he was born there; he was only (temporarily) living elsewhere. Is the claim of impartial professionalism made by the health workers real? Is it a coincidence that the strongest grievances against the health centre came from people who had no connections there? Reid, working in Yirrkala in the 1970s, noted that health workers were accused of favouritism based on clan allegiance (Reid 1983).

It is not surprising that there is a considerable toll on the clinic staff responding to the demands of the job. Whilst there is a cohort of dedicated health workers at Galiwin'ku, one third appear to be absent at any one time, often for prolonged periods. Similarly, there is a frequent changeover of *Balanda* staff, with each new employee needing to become acquainted with a seemingly overwhelming amount of clinical and cultural information (many haven't had any experience in Top End communities prior to their employment at Ngalkanbuy). *Balanda* staff repeatedly commented how they were unable to provide an ideal service: the health service lacked structure - "a little bit 'hit and miss'" (Interview 21), there were insufficient resources to provide patient education (Interview 22) and a primary prevention program was out of reach (Interview 2).

Amidst their other responsibilities, the efforts of staff to follow up 'non-compliers' and patients 'lost to follow up' were seen as yielding a low return. Staff described the frustration of tracking down patients for their monthly *Bicillin* prophylaxis.

Health worker But how long do we have to stand there, from morning until afternoon, trying to chase them for nothing? (Interview 22)

Balanda staff also spoke of their personal distress in looking after young patients with serious disease. Health workers did not explicitly describe distress relating to illness and death in young patients, but were clearly unsettled by the conflicts with family that ensued. It was common for health workers to decline to look after patients with a terminal illness in Galiwin'ku. Being the last person to attend a dying patient left that

person open to accusations of malpractice, although, to my knowledge, this has never been followed through to litigation (see section 16.4).¹²

15.4 Suggestions for patient care

Every participant was asked to make suggestions for the ‘best way to look after patients with RHD’. The answers to this question form the basis of this section. These ideas were reported back to the community and health staff at the feedback meeting, where comments were invited (see chapter 17). The recommendations do not necessarily reflect the views, or the analysis, of the researchers. They serve to indicate the (at times quite divergent) priorities for patients, families and health staff.

Two health workers should be nominated to coordinate the care of patients with RHD. Ideally, the health worker should be the same sex as the patients they are responsible for. A consistent, familiar person would secure the trust and confidence of young patients. This reflects the emphasis on familiarity and trustworthiness of the health service provider.

Patients should be able to choose where to receive their injections. Some patients preferred to go to the clinic for their monthly prophylactic injections; others preferred to receive their injections at home. Men needed special consideration, because of the possibility that they felt uncomfortable attending a clinic dominated by female staff. All staff needed to be aware of the principles and practical aspects of administering

¹² The suspicion with which health staff are perceived depends on the family’s perception of the illness. In a recent case in Galiwin’ku, numerous family conferences were held to discuss the serious and terminal nature of a young man’s liver disease. Despite this, when he died, suspicions were raised that health staff had hastened his death.

prophylactic penicillin to patients with RHD. Displaying a lunar calendar at the clinic might assist staff in planning clinical duties.

Patients and their families value check-ups. A ‘full check-up’ reassures the family that the patient is being cared for properly, and is part of proper treatment. A check up should involve a careful clinical review, and communication with the family. Full check-ups might involve being referred to Gove or Darwin for an echocardiogram, or admission to hospital. Patients and families also expressed a wish for screening check-ups to exclude serious disease.

Patient It would be good if we can take the children in for check-up, the older people too, and tell them [the health staff] to do a full check-up, instead of just giving *Panadol*. When they give only *Panadol*, the inside of our bodies is still aching, and sick. (Interview 12)

Many patients requested ‘**reminder**’ cards for appointments, check-ups and even their *Bicillin* injections. As there is no domestic postal service in the community this would involve health staff delivering the cards. The perceived inefficiency of this practice had led to the abandonment of reminder cards a few years previously. Reminder cards not only served to notify patients of an impending appointment, but also to legitimate an appointment if no warning was given. A nurse described her practice of handing out appointment cards to patients at the same time as she picked them up for clinical reviews. In her opinion, patients were much more likely to attend if an appointment card was offered at the time of the pick-up (Interview 21).

Good communication between health staff and patients and their families is important. Relatives said that patients needed to understand the disease, and the results of their investigations. People wanted to hear the ‘*dhudhi dhāwu*’, the true or ‘root’ story relating to the disease. The need to inform the whole family was stressed, especially when there was an important treatment decision to be made.

Communication lines between family members must also be maintained. Good communication helped to prevent accusations of wrongdoing from flourishing after an adverse event, according to health workers and relatives. A mother recommended extensive family consultation prior to making important treatment decisions:

Relative ...I think all the family have to come together to sort out these problems, the sickness for these kids. So we have to talk it over, like uncles, mothers, grandmothers... So if anything is going to happen to them, we know. We know they have suffered from this disease. So we can't turn against doctors and nurses, we know. (Interview 6)

If patients and their families didn't receive a satisfactory explanation for an adverse event, not only might they blame the health staff, but also they might invoke sorcery as the cause and refuse what was seen as irrelevant medical treatment.¹³ Suspicion could be prevented by education, best given ‘around the camp’ according to another mother:

Relative All the time we should give education around the camp. I think this is the best, so that people can know. In future time the people with heart disease

might die because we thought about it the wrong way: “it might be sorcery (*djāngitj*)”. Because we Yolngu people don’t know, we don’t know about this sickness. (Interview 15)

Patients and health workers valued **home visits**, as an opportunity for providing health education, but also to ‘comfort the family’. When health workers used to perform more home visits, there were fewer deaths in the community, according to one patient. New *Balanda* staff were uncomfortable at times approaching patients in the community, as they were unfamiliar with social codes. In the past, some community members had expressed their dislike of *Balanda* staff visiting their homes, as they were seen as critical of people’s homes.

Health worker One thing we stopped visiting around the camps, because they just felt that we were spying on them, which house is good, which house is bad. A lot of people were saying that to us when *Balanda* visitors came around to check their skin for scabies. We were trying to help them, but they were thinking the wrong way. (Interview 23)

A ‘**social night**’, or educational event, for RHD patients was much discussed. Even though few patients had much to say about the past social nights, many expressed enthusiasm for an opportunity for patients to get together, share their experiences, and to hear personal accounts of operations and procedures. Health workers and relatives saw these social nights as an opportunity to teach young patients about healthy living, such as good food, exercise, and quitting smoking. Almost as an afterthought, it was

¹³ Reid recounts a story where a young man refused medical treatment for his alcohol induced hepatitis, as he believed that sorcery was the cause of his illness. He subsequently died. (Reid 1983)

said to provide an occasion to deliver education about RHD. Ideally a doctor, as the person possessing the authority of the medical knowledge, should deliver this education with the assistance of an interpreter and using visual aids such as an anatomical model, pictures or videos.

Special measures should be initiated for patients who travel. Key ideas to facilitate treatment outside the community included patients taking a record of their medical history, a reminder card, or a prescription for *Bicillin* with them whenever they travelled. Health staff preferred that patients notified them of their travel plans, and presented to the clinic for review when they returned. If informed of the patient's contact details, health staff were generally happy to contact the relevant organization to organise ongoing *Bicillin* prophylaxis.

New health staff requested **more education** on RF and RHD. This included finding the least painful injection technique for *Bicillin*.

The ideas outlined so far are already in place, or have taken place in the past. This predilection for supporting the status quo may be the outcome of a state of relative powerlessness associated with colonisation and socio-economic disadvantage. It may also reflect that the question and even the whole research 'problem' had been defined by an outsider, and respondents strove to satisfy this outsider by making suggestions that reaffirmed that person's beliefs and paradigm. This is particularly apparent in the health workers' support for an educational night, despite ambivalence by patients for these 'Western' methods of transmitting information. By contrast, the following ideas

I would classify as novel and thus may represent independent thought, not requiring the tacit approval of the researchers.

Patients could receive a payment for getting their *Bicillin* injections on time. This suggestion came independently from a patient, a relative and a specialist. It engendered interest, but also concern as to its feasibility. Alternatively, **staff could be rewarded for delivering the *Bicillin* injections on time.** This would appeal to a Yolngu sense of competitiveness and fun, according to a nurse. It would be one way of “balancing (Yolngu) culture with the way *Balanda* do stuff” (Interview 21). The possibility that these strategies would result in a loss of the patients’ control over, and responsibility for their health was raised.

Another nurse suggestion was that **an announcement over the loudspeaker** be used to remind people that their *Bicillin* injection was due. Public announcements are made on a daily basis via a loudspeaker that can be heard throughout Galiwin’ku. A subtle, cryptic remark, which would be recognised only by the relevant people, could be made. Although announcements had been used in outstations in the past, the health workers felt that it would be ‘too embarrassing’ for Galiwin’ku patients today.

One relative saw the need for **a special building for young people** to get together. This would provide a comfortable place for young people, away from the clinic and the home. It would primarily be a social centre, but space and opportunity could be provided for health staff to meet with young patients for clinical and educational purposes. Advantages would include privacy in a comfortable environment, and this would promote good communication.

Relative We need to have something, a good building, everything good. So the health workers go to the right place, not to the home. I am feeling right now, because some of us are missing this part. We can't go around to the homes of the children, sometimes the kids are too shy and they might not give you their stories. (Interview 13)

This comment is somewhat at odds with the other accounts of young people being most comfortable within their family sphere, but probably reflects this respondent's experience working with youth who abuse illicit substances. When there is conflict within the family, a space away from the home may indeed promote better communication. This respondent went on to say that the building should be purpose built structure, with its purpose identifiable by the community, and also by government organisations. He expressed a need for official recognition and legitimacy in the eyes of 'government people'.

Others suggested **supplying bush food** to RHD patients, either directly, or by taking them hunting, or by making bush foods more available, for example, at the take-away.

One patient, with a professional background, suggested **more research**:

Patient It's just my idea, we could do research work: how we can care for our body. We should work with the take-away, like we can find out which is good food and which is bad food at the take-away. (Interview 16)

This further illustrated the widespread interest in healthy food. Perhaps it also reflected the fact that food and nutrition have been the subject of a disproportionate amount of research in Aboriginal communities (Anderson 1996).

A doctor suggested **an audit of patient records** to identify the demographic characteristics associated with better treatment compliance. Research of this nature has provided few solutions in the past (see chapter 11).

Finally, health staff called for an **active response to non-compliance**. Staff should not ‘give up’ on people who refused or missed treatment. Non-compliance need not be seen as an outright rejection of medical advice. It may mean that patients require more information, or more precisely, a better *understanding* of the likely benefits of treatment. Non-compliance may represent a ‘cry for help’ and may necessitate *more* attention, rather than a withdrawal, by health staff (Devitt and McMasters 1998). Even when patients were absent from the community, there was an expectation that the staff from Galiwin’ku would maintain an active interest in their care. Failure to do so was seen as not caring for the patient.

15.5 Conclusion

A number of concepts believed to be central to good care of patients with RHD and good health were described. These included the processes of care (*djāka*) and encouragement (*gunḡa’yun*) of patients, a healthy lifestyle, a good lifestyle, good relations with family and health services. Health services were expected to be professional, efficient and familiar.

Poor care was said to result through patients not heeding the advice of family or health staff, negligence of the patient, family or health staff, miscommunication or poverty. Fear, immaturity and 'laziness' were described as causes of patient non-compliance. Whether these factors are indeed significant in the uptake of prophylaxis for RF will be discussed in the following chapter.

16 Processes and connections (axial coding)

16.1 Why do some people miss their injections?

16.1.1 Introduction

In this section, the process of “getting the needles” – the monthly prophylactic injections of benzathine penicillin or *Bicillin* prescribed to patients with a history of rheumatic fever- will be discussed in detail. The analysis will take on the form of ‘axial coding’, with its principles of relating concepts to each other to form theory. According to the grounded theory methodology the context, the actions and the consequences of the process are identified. The emergent theory can be expressed with the assistance of diagrams or matrices. ‘Matrix’ is used in the sense of a dynamic table, which assists the analyst in generating theory (Strauss and Corbin 1998). The result is intentionally abstract, yet ‘grounded’ in the data.

In the process of “getting the needles” there are two sub-processes that I will analyse according to context, motivation, action and consequence (see table 18). The first involves health staff, working within a medical context, and will be called “service provision”. The second involves the patients who receive the needles and whose physical and intellectual contexts are different and varied. This sub-process I will call “treatment uptake”. There are distinct differences in the motivations and the consequences for both parties. Factors affecting each stage of the process will be discussed. Both processes occur within a larger historical and socio-economic context, which I will touch on briefly. By looking at both components of the process, the

narrow view of compliance as a problem originating in the patient, or the clinical interaction alone, is avoided.

16.1.2 An analysis of the process of health service provision

To begin with the broadest context, it is important to acknowledge the process of the colonisation of Arnhem Land, and the foundation of the settlement of Galiwin'ku by missionaries. The present day reality of a health service run largely by outsiders, albeit sympathetic and well-meaning suggests parallels with the history of colonisation: cultural difference, inequalities and communication issues are inevitable features of the process of health service provision and uptake in Arnhem Land.

There are two health services on Elcho: Ngalkanbuy Health, and the Marthakal homelands service. The nurses' experience of working in Yolngu communities ranged from three months to many decades; the health workers all had over five years' work experience. Knowledge of the *Bicillin* program varied according to duration of residency in the Top End: the majority of the *Balanda* clinic staff did not know the timing, the method of recording, or the purpose of the injections, whilst the health workers all seemed to have a working knowledge of these issues. One consequence of not knowing the practicalities or the reason for the *Bicillin* injections, was that *Balanda* staff did not give injections opportunistically, leaving it to the regular monthly run.

At Ngalkanbuy Health, injections are administered to patients by staff of the same sex (see table 18). Effectively this meant that the local doctor, assisted by a male nurse or health worker, ran the program for the men, and a female health worker gave the

injections to the women. At the commencement of this study one health worker had taken responsibility for the female patients over a number of years, but she resigned during the study and was not replaced.

The Yolngu staff's familiarity with their patients and the ease with which they approached people in their homes assisted in the endeavour of administering the needles. Some of the *Balanda* staff were unsure as to the social protocols governing treating people at home.

Nurse It is quite amazing. You go out and find someone playing cards, or somewhere else, and say, "do you want to come to the clinic for your *LA Bicillin*?" And they say "do it here", and so you go "OK, in the car." And then it's over and done with. (Interview 21)

It is not possible to construct from these interviews a comprehensive account of the motivation for staff to carry out this at times tedious task, but a few observations can be made. Motivation to administer the needles, it seems, came either by instruction or by having a personal dedication and sense of responsibility to the patients. Factors that impeded staff motivation included being overwhelmed by urgent clinical priorities, and the effort involved to ensure injections in difficult situations, such as for patients who were refusing injections and patients absent from the community.

Perceiving a benefit from the treatment was an important motivating factor for giving the injection, not just for receiving it. The perceptions of the program appeared to differ between the Yolngu and *Balanda* staff, with *Balanda* staff conveying a more

negative impression. *Balanda* staff spoke about the process of giving injections as ‘intrusive’, patronising, exploitative of the power differential, painful, ‘persistent’, unpredictable and chaotic. These negative statements were presumably overridden by the drive to fulfil job requirements. By contrast, Yolngu staff spoke more about the *benefits* of the *Bicillin* treatment, in terms of saving lives and preventing further disease. *Bicillin* was seen as ‘strong’, ‘long acting’, and able to penetrate the whole body and travel through to the heart.

Health worker Before, there was no medicine. It is lucky that we’ve got medicine for the heart now, because we would have died. (Interview 22)

Staff accepted the need to respect patients’ wishes for receiving the injections either at home, or at the clinic. The health workers were particularly accepting of the practice of administering the injections at patients’ homes. When a patient was absent from the community, staff did not feel that they held the responsibility for ensuring that the patient received their injections. During a short-lived absence the relevant health service was usually contacted, but there was no attempt to ensure that the injection was actually received.

Staff spoke of the “art of giving the needle”, concerned primarily with reducing the pain experienced by the patient. The doctor was the only person who expressed uncertainty as to how to administer the injection; others spoke confidently about their technique. Health workers were indignant over patients’ complaints and did not necessarily indulge patients’ preferences for a particular staff member to give the injection.

Health worker Sometimes they get hurt, and they tell the health workers that we have given them the wrong medicine, its gone into the bone. They don't know! (Interview 23)

Outcomes for the *Balanda* staff ranged from the satisfaction of getting the job done quickly, to the sense of frustration when the patients couldn't be found, or were not agreeable to immediate treatment. By comparison, health workers reported positively on the experience of giving out the injections. They claimed a much better success rate, giving fifteen injections in one morning, for example. These experiences impacted on the motivation of the staff for carrying out the program. Thus 'consequences' impacted on 'context', and the cyclical nature of processes was confirmed.

Table 18 - Matrix for the process of ‘getting the needles’

| Process | Context | Motivation | Action | Consequence |
|--------------------------|---|--|---|--|
| Service provision | <ul style="list-style-type: none"> • Duration of employment in health services • Knowledge of the Bicillin program • Cultural familiarity • Past experiences of giving the needles | <ul style="list-style-type: none"> • Professional obligation • Duty of care • Perceived benefit to the patient of the treatment | <ul style="list-style-type: none"> • Staff give Bicillin needles • ‘The art of giving the needle’ | <ul style="list-style-type: none"> • Job satisfaction • Frustration • Ambivalence |
| Treatment uptake | <ul style="list-style-type: none"> • Diagnosis of RF • Knowledge and understanding of the disease • Familiarity with the service provider • Past experiences of getting the needles | <ul style="list-style-type: none"> • Perceived benefit of the treatment • Belief that the disease is serious | <ul style="list-style-type: none"> • Patients receive Bicillin needles | <ul style="list-style-type: none"> • Symptom control • Disease control • Disease prevention |

16.1.3 Analysis of the process of treatment uptake

For patients the starting point of the process of treatment uptake is the diagnosis with RF (see table 18). This usually occurs between the ages of five and fifteen, but the condition may also be discovered during an antenatal check-up, or in the later stages of chronic valvular heart disease. As RF is known to run in families, patients may know of a relative with the condition, and be aware of the medical experiences of that person.

Treatment uptake was facilitated by familiarity with the service providers, and the routine and timing of the injections. The importance of this should not be underestimated: patient allegiance to ‘their’ clinic, their feeling of *belonging* to a service mediated their interactions with other services. This was brought sharply in focus when patients were absent from their community, when there was an explicit expectation by patients and their families that the home clinic should ensure the ongoing care of the patient. A relative was often nominated to contact Ngalkanbuy Health centre, which was then expected to arrange ongoing services by another provider. Patients expected or hoped for a similar arrangement to that which occurred in Galiwin’ku such as receiving ‘reminders’ and being picked for injections and check-ups. Failures in any stage of this process were not actively followed up by any party. The following quote is just one example of this story. This patient knew when her injections were due, and actively sought out medical treatment when in Galiwin’ku, but failed to make the connections required to receive injections in Darwin.

Interviewer When is the time for your injection?

Patient Every month, every new month.

Interviewer Did you miss your injections?

Patient No. But I missed all my injections when I was over in Darwin.

Interviewer Why? Too busy? Your home was too far from the clinic?

Patient My sister used to ring the Galiwin'ku health centre to transfer the injection to Danila Dilba.¹⁴ No one from there contacted us and let us know if the injection is ready or not.

Interviewer How long was that for?

Patient A few years. When I came back to Galiwin'ku I saw the doctor who started the injections straight away.

Interviewer Did you ever contact Danila Dilba yourself?

Patient No.

Interviewer (JB to ZH) She relied on Ngalkanbuy to sort it out. (Interview 19)

These problems were not limited to patients travelling to Darwin, and are not necessarily an indictment on any particular medical service. The same events occurred in the microcosm: patients from outstations failed to receive their *Bicillin* injections on time when they were in Galiwin'ku. It appeared that context and relationships with service providers were critical for patients. Disruption of established connections and pathways led to derailment of the whole process of treatment uptake. Patients attempted to recreate familiar patterns by using relatives and their home clinic as mediators for health service delivery. This implied the need for the clinic to adopt the

¹⁴ Danila Dilba is the Aboriginal medical service in Darwin.

care and concern similar to that of a responsible relative, care based on *relationships* not geographical demarcations.

These observations suggest that Yolngu people situate themselves firmly within their families. According to Rowse, the family, rather than the individual, is the primary social unit for Aboriginal people, and this is a far larger organisation than the nuclear family that is standard for non-Indigenous people (Rowse 1996). In this study, Yolngu families were linked through association, rather than by geography, to health service providers. When at home, the boundaries between families and health staff were blurred: patients may be (literally) related to health staff, and health staff were ‘everywhere’ integrated within the community. Other health service providers were seen to be located in a metaphorical ‘outer circle’, which families preferred to access via the home clinic (see figure 15). Being a dedicated Aboriginal health service was not sufficient to make another service familiar or accessible to these patients.

Discussions with health staff from other communities supported this finding that Aboriginal people in the Top End were strongly loyal to their local clinic, or at least, hesitant to use alternative health services.¹⁵

¹⁵ A nurse at another bush community told of community members becoming unwell in Darwin, then driving some 500 kilometers back to their ‘home’ clinic for medical assessment, only to be evacuated by airplane to Darwin hospital.

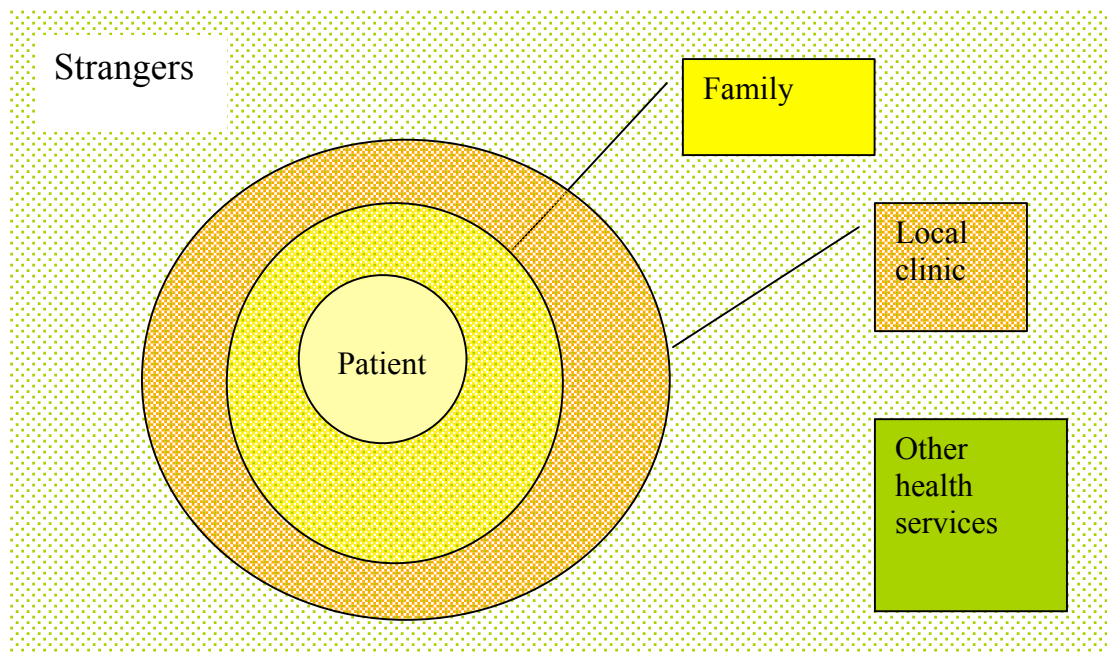


Figure 15 - Inner and outer circles of care

Mincham, working in the Kimberley, found that whilst a close relationship between the patient and the health service provider was associated with treatment compliance, patients from small communities were also concerned about confidentiality, the transience of the non-indigenous staff and perceived incompetence of staff (Mincham, Toussaint et al. 2003). This difference in health service provision between towns and communities in the Kimberley contrasts with our findings, and may relate to the smaller size of the communities in the Kimberley. For Kimberley patients, towns provided a greater choice of services with easier access to specialists, whilst community dwellers had to contend with intermittent medical services.

Factors that affected a patient's likelihood of accessing medical services when away from home included the purpose of the travel, whether for work, social or ceremonial reasons. The destination was also relevant: some Yolngu abused alcohol when in

Darwin, and subsequently were ambivalent about ensuring ongoing medical treatment.

Within the local context patients differed in their preferences for receiving the *Bicillin* needles. There was evidence that men had particular concerns for privacy. For some men this meant receiving their injections at home, where the greater community wouldn't be privy to the event, for others it meant receiving their injections at the clinic, presumably to avoid the gaze of the family. The preference for same-sex staff to administer the injections did not apply as strictly with *Balanda* staff who were less implicated by Yolngu customs.

The strongest recommendation for a treatment was a patient's own positive past experience. Generally *Bicillin* was regarded favourably. It was said by patients to relieve symptoms of chorea and breathlessness. It was also felt to 'help the body', in particular the heart, and to be 'good for your health'. Patients said they were able to sense when their body 'needed' *Bicillin*.

Relative Because inside his body he can tell if he misses injections or tablets. His body can tell there is something happening to him, and then he asks me to get him some tablets or an injection. "Can you ask the health workers for an injection?" he says. "Because I think my body needs an injection." When he takes this, he feels happy. (Interview 18)

Lacking this, patients were motivated by trust of the health carer. This was generated either through personal contact, by reputation or association, and could be

strengthened by the experiences of other family members. A biomedical understanding of RF was not essential for a patient to accept injections. (This will be discussed more fully in section 16.2. Instead, knowing that the disease was both chronic and serious was said to be an important incentive for patients to get injections.

Relative What I understand is that she didn't get the full story from the doctor and she didn't know about this disease, like the full story. That's why she was missing all these injections. She thought that it was a superficial (*garramat*) sickness. But sometimes I was concerned. I told her it wasn't a *garramat* sickness, "its inside your heart, it will stop your heart all at once". (Interview 15)

Patients had two basic approaches to getting their injections: either passively waiting for health staff to deliver the injection, or actively seeking treatment. From their responses it was clear that patients who did not attend the health centre on their own initiative could not be considered non-compliant. Rather, they willingly accepted treatment that was delivered to their home.

The consequences for patients were an inevitable combination of the side effects of the injection (pain, fever, swelling) and the perceived benefits, as described above. The pain associated with the needles did not seem to be a significant disincentive for patients. Only one of the patients spoken to refused injections and, although her reasons were obscure, they did not relate to the pain of the needles. Another respondent, a relative, stated emphatically that he disapproved of needles on principle,

but upon hearing the justification for the regular injections he recognised their necessity.

The two sub-processes of service delivery and treatment uptake were not independent of each other, but interacted at each stage to differing degrees (see figure 16). For example, the context for the processes was quite distinct for *Balanda* staff and the patients, but rather more shared for the health workers and the patients. Furthermore, the consequences of each process formed part of the context for future processes, in the recurring monthly cycle.

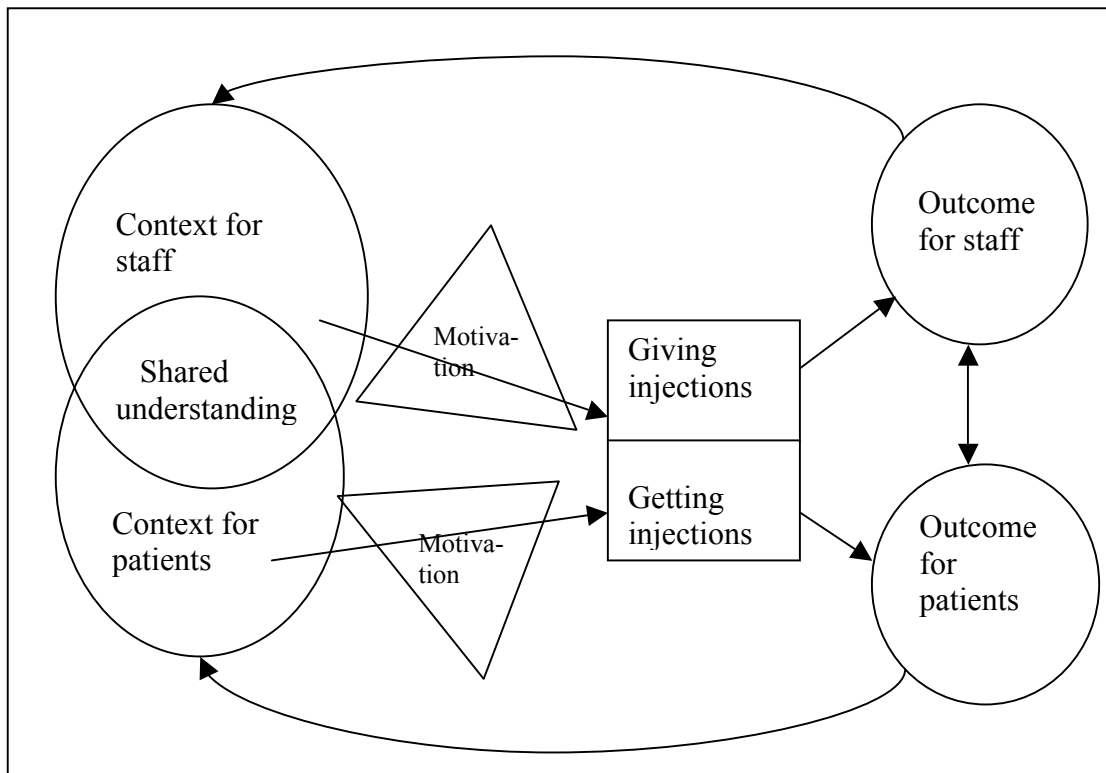


Figure 16 - Diagrammatic representation of the process of getting the injections

16.1.4 Causal and intervening conditions for the process of ‘getting the needles’

Finally, to fulfil one of the stated aims of this enquiry, I will propose a set of facilitators and impediments to the process of RF patients getting regular *Bicillin* injections (see table 19). In doing so I fall back into the role of a clinician attempting to solve the clinical ‘problem’ of non-compliance, but acknowledging the greater social context softens my fall a little. In addition, my conclusions are drawn from patients’ stories, albeit abstracted through a doctor’s mind. The lists of facilitators and impediments are neither exhaustive nor definitive. A discussion of how ‘patient passivity’ can feature both as a facilitator and as an impediment to getting needles is found in section 16.4.

Table 19 - Facilitators and impediments to "getting the needles"

| | Causal conditions for 'getting the needles' | Intervening conditions for 'getting the needles' |
|-----------------|--|---|
| Staff factors | Appropriately trained staff | Negative perception of the Bicillin program |
| | Socially and culturally competent staff | Conflicting priorities for staff |
| | An active recall system | No effective strategy to deal with absent patients |
| | Staff willingness to treat the patient at home | Staff fatigue and frustration |
| | | |
| Patient factors | An appropriate location for receiving injections | Conscientious refusal of treatment |
| | Belief that the disease is chronic and serious | Inconvenience to the patient |
| | Confidence in the health service | Being away from home |
| | Family support for the treatment | Lack of family support for the treatment |
| | Patient passivity | Patient passivity |
| | Belief in the efficacy of the treatment | Lack of confidence in the treatment |

16.1.5 Audit of *Bicillin* coverage

Patient records and the clinic's RHD register were audited to assess the coverage of RF/RHD patients with monthly *Bicillin* prophylaxis. The period studied was from January 2002 to September 2003. All patients appearing on either the clinic's list or on the Darwin-based RHD register were included. The percent of *Bicillin* injections administered each month was calculated by dividing the number of injections

administered, by the number of patients eligible for the injection. Patients who were marked absent from the community were not included in the denominator. The results are depicted in figure 17. Using this data the average rate of *delivery* of *Bicillin* injections in Galiwin'ku was 76.5%, which compares favourably with international figures and those found in the Kimberley (Nordet 1992; Mincham, Mak et al. 2002).

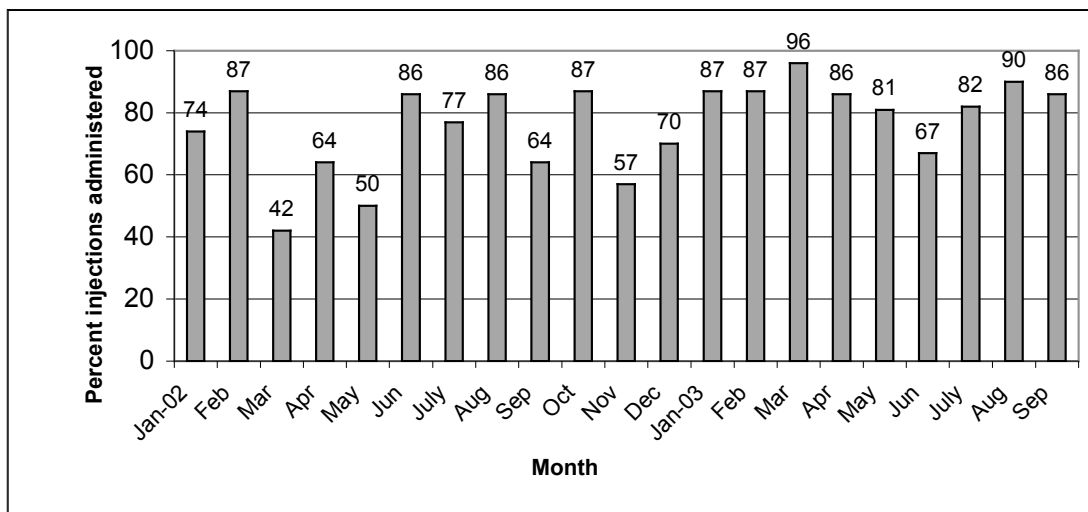


Figure 17 - Percent of *Bicillin* injections administered to patients in Galiwin'ku by month

The proportion of penicillin injections received by each patient was calculated by dividing the number of injections received (according to the records held at Ngalkanbuy), by the number of months the patient was eligible for an injection. An assumption was made that no injections were received when the patient was marked as 'absent', or 'in Darwin'. Thus this graph depicts the worst possible scenario in terms of *Bicillin* coverage . The results were arranged from highest coverage to lowest, and are depicted in figure 18. The average proportion of *Bicillin uptake* is 72.4%.

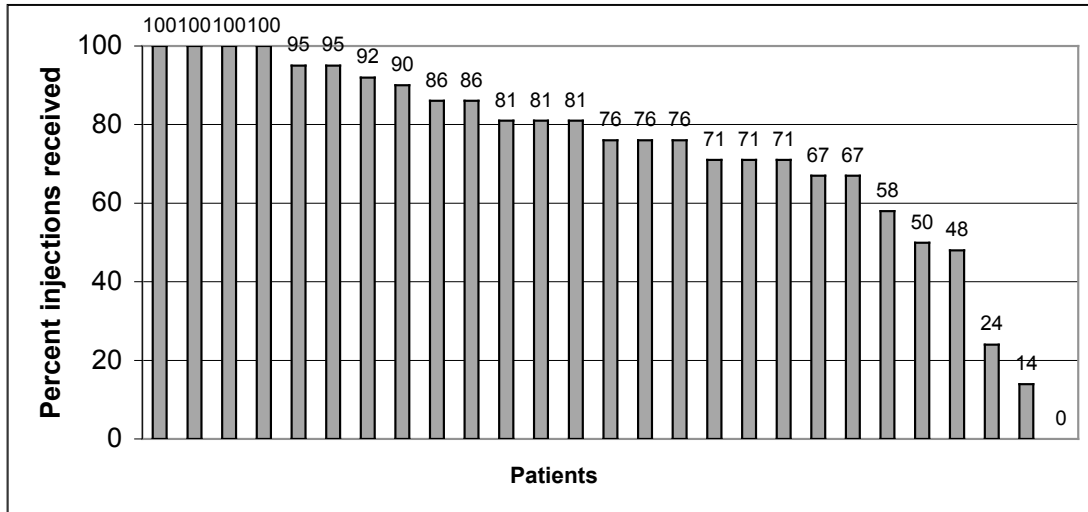


Figure 18 - Percent of *Bicillin* injections received by each patient

16.1.6 Discussion of the results of the audit, and correlation with the interview data

This small sample compares reasonably with the WHO reviews of *Bicillin* coverage in third world countries in the 1980s, described in section 10.3, where the average rate of coverage was 63.2% (Nordet 1992). Expressed in another way, 59% of patients in Galiwin'ku received full prophylaxis (more than 75% of injections), 26% achieved irregular prophylaxis (50 to 75% of injections), and 15% achieved occasional or no prophylaxis (less than 50% of injections), while the WHO rates in the 1970s were 60%, 20% and 20% respectively (Strasser 1985). The consistency of these results over time and across the globe might suggest that a certain degree of failure to deliver prophylaxis is unavoidable, and represents the inherent impossibility to achieve perfect outcomes in a real world context, reminding us once again that non-compliance is the “material consequences of particular models and practices of health service provision”(Humphery and Weeramanthri 2001).

Looking at the local data further, it is apparent from figure 17 that whilst the clinic in Galiwin'ku delivers a good proportion of the prescribed injections each month, there appears to be considerable month-to-month variability. Clinic factors that would affect the ability of the staff to carry out the program such as low staffing levels, competing priorities in the health centre and local events such as cyclones, ceremonies, funerals and festivals may account for this. It seems plausible that having a dedicated person responsible for the *Bicillin* program would improve service delivery, which concurs with the suggestion made by an interviewee.

The health staff's self-congratulatory stance regarding *Bicillin* coverage should be revised to account for the patients who were absent from the community, and most likely not receiving any *Bicillin*. If the responsibility for ensuring that injections were administered to all patients, whether in or out of the community was attributed to the clinic (as stated by the patients), then the clinic's rates of coverage with *Bicillin* would appear lower.

Another observation concerns the group of patients who received less than 50% of their needles. The patient who achieved no prophylaxis at all represents an important case in point. This patient appeared on the RHD register, but not on the clinic's working '*Bicillin* list'. The medical notes indicated that the diagnosis of RHD had not been conclusive, but that the patient had been lost to follow-up, even though she appeared to have remained in the community. Thus an administrative oversight (anecdotally combined with patient resistance to treatment) meant that a local patient had received *no* RF prophylaxis.

From the medical records, and with their consent, it is possible to comment on the other patients who received less than 50% of their needles. Two had spent considerable time in Darwin, and spoke of their inability or unwillingness to access medical services in Darwin.¹⁶ The third patient was the only person who overtly objected to having the injections, even though she was amenable to persuasion at times. Her reasons for objecting to the injections were unable to be fully explored as she exhibited significant cognitive impairment in her interview.

Referring to the group who received irregular prophylaxis, no great generalisations can be made. Some travelled regularly, others were ‘too busy’, another regulated the frequency of injections according to her symptoms, and one was described by health staff as mischievous and occasionally belligerent. These patients neither conformed to any particular classification, nor differed significantly from those who received full prophylaxis.

These findings suggest two possible approaches to improving the *Bicillin* coverage for RF/ RHD patients in Galiwin’ku. The first approach would be to target the group with the lowest rate of *Bicillin* uptake, and would focus on administrative improvements to ensure that patients are not lost to follow-up and that the working *Bicillin* list used by the clinic is regularly compared with the RHD register. In addition, specific efforts should be made to facilitate patients’ access to urban medical services.

¹⁶ I acknowledge that the quantitative data is less objective than it might appear, as I relied on the accounts of these patients in making the assumption that patients received no injections when the record indicated that they were ‘in Darwin’.

The second approach would be to introduce measures to improve the uptake of *Bicillin* for all patients, in order to raise the level of coverage in the group with irregular prophylaxis (26% of the patients). This might include some of the suggestions made in section 15.4 such as the offering of incentives to patients, reminder cards, providing more education, and improving communication between patients and their health carers. A holistic approach by health services that acknowledged the Yolngu concepts of care (*djāka*) and encouragement (*gunḡa'yun*) is likely to be well received, and may result in increased satisfaction and participation by community members.

16.2 Knowledge and understanding of the disease and medical treatments

16.2.1 Patients' accounts of disease causation and pathology

A secondary aim of this study was to elicit a Yolngu narrative for RHD, its cause, patients' experiences and its outcomes. The reason for this was so that staff and patients could better achieve a *shared* understanding of the disease. The idea of a shared understanding acknowledges the differences between the patients' health beliefs and the medical paradigm, and confers an equal respect for both. The Aboriginal health workers, with their education in both systems, personify the intersection of the two paradigms.

Expectations of finding a 'pure' Yolngu narrative were quickly dashed. Few respondents had anything at all to say about a Yolngu perspective on RF and RHD. Most respondents either flatly denied having such knowledge, or implied that 'before'

(that is, before colonisation) either the disease was non-existent, or that Yolngu were not aware of the disease. For some time I persisted asking questions in different ways before recognising reasons for the lack of results.¹⁷ Firstly, as a doctor practicing in the community I would have been seen as an authority and respondents may not have felt able to express their own beliefs about the disease and its cause. Secondly, I had been looking for an authentic Yolngu narrative, something I believed to exist if only the skill of the interviewers could bring it to the surface. However, many of the discourses that were elicited bore evidence of the biomedical model that had been conveyed over the years by health staff as well as via other sources such as the mass media. Thus the Yolngu RF story appeared to be a mosaic of knowing and not knowing, of the biomedical and the intuitive.

Many gave no explanation for their illness, whilst others held what appeared to be discordant views, without any apparent tension. Some patients and relatives had absorbed the biomedical explanation for the disease, whilst others had not. Many acknowledged that they had heard about the streptococcus germ that caused heart damage, even when they didn't volunteer this initially as the cause of the disease. The clearest account relayed by a patient linking germs and heart disease was not entirely biomedically accurate, yet resembled closely an account given by an experienced nurse.

¹⁷ Questions included:

- Is there a Yolngu story for RHD?
- Are there any bush medicines for heart trouble?
- What do you do for people who have palpitations or who are breathless?
- There are many stories about what causes RHD, can you tell me all the stories you know?

Patient A little worm (or germ, *mewirri*) goes into your blood, and then it goes into your heart. When it gets into your heart it makes holes in the heart.
(Interview 17)

Other explanations of disease causation offered by patients and relatives ran the spectrum from medical explanations to intuitive observations. They included intrauterine events, such as the mother eating bad food, an injury to the womb, or the baby swallowing its own faeces (probably referring to meconium aspiration).¹⁸ Some respondents recognised that the disease appeared to be inherited.¹⁹ Patients also described contracting the illness after eating bad food or falling out of a tree, because of cold weather, walking in salt water, carrying a jerry can of water, or from a lack of exercise. Smoking was believed to cause deterioration in the condition. The references to food, smoking and exercise may indicate conflation of medical evidence relating to ischaemic heart disease. These findings support the observation of the local doctor who felt that Yolngu often focussed on situational events, coincidences or “clustering occurrences” as the cause of disease (Interview 2). Another cause of disease he suggested was “kinship”, referring to the transgression of traditional kinship laws. Indeed, Reid lists the transgression of sacred laws as one of the Yolngu explanations of disease causation (Reid 1983).

Patients’ explanations of medical concepts were unclear at times. Despite working closely with the Yolngu co-researcher on these elements I am still unsure whether patients were uncertain about their ideas, or whether they were simply expressed

¹⁸ Reid found that neglect of the Yolngu laws governing the diet of pregnant women was thought to be the cause of some congenital malformations. (Reid, 1983: 53)

¹⁹ See part 1 of this thesis for a summary of the evidence for an inherited susceptibility to ARF.

ambiguously. The use of English words such as ‘artery’ suggested that patients did not have a clear understanding of the concept. A few examples illustrate this concern:

Patient I have a problem with my right or left, that kind of thing like a net, your blood goes in and out. It’s not closing properly, that’s why it’s leaking back. (Interview 5)

Patient My heart wasn’t working properly.

Relative The heart valve?

Patient Yes. Artery valve. My heart is not pumping well.

Relative The heart valve?

Interviewer (JB) The heart pumps the blood through the body.

Patient It just goes one side of my heart, this blood. I think it clotted. It is sort of like an umbrella. (Interview 15)

Whilst the treating specialist believed that Yolngu have an excellent knowledge of anatomy and “how blood flows” it has been described elsewhere that Yolngu do not have a concept of the pumping of the heart leading to the circulation of blood. Instead there appears to be a central idea of *ɲir* that represents both pulse and breath (Trudgen 2000). I found evidence for this in two instances. A remedy for breathlessness was demonstrated to me, which consisted of rubbing the arms to push the ‘breath’ back into the body. The following quote also suggests the conflation of pulse and breath:

Patient I think the heart vein (*raki*) maybe it was blocked. That’s why the blood was not pumping well. When I breathed in, it just stays in, not coming

out. But I don't know what caused it, whether it's from my heart beating?

(Interview 12)

The lack of clarity in medical explanations for RF can be partly explained by the nature of the explanations the patients received from health staff. Some health staff themselves had poor knowledge of the disease, and others, did not put a high priority on explaining disease causation. An example of an explanation provided by a health worker with ten years experience illustrates the lack of detail given to patients, and the focus on the implications rather than the cause of the disease.

Health worker We will tell them that this part of the body is very sick, so you might get an operation. Because in your heart, there is something blocking your heart. (Interview 23)

The *Balanda* staff reflected on what was the desirable level of knowledge to convey, and suggested that, in many cases, patients didn't need or want to know all the facts. A nurse commented that "we probably know too much" (interview 21), echoing a common belief that too much knowledge, particularly of the possible negative outcomes of a disease might lead to anxiety. But is it true that 'ignorance is bliss'? It has been said that the control of knowledge for Yolngu is strategically important.

Yolngu knowledge was structured according to the senses of their concept of *marnggi*. This word can be translated 'know' or 'knowledgeable' but it had something of the sense of 'can' or 'be able' as well. Thus ... knowledge was not separable conceptually from power. (Keen 1994)

Some patients and relatives conveyed the sense of knowledge as a valuable commodity that was denied to them. It was said by one family that clinic staff withheld important information. Others said that only *Balanda* possessed knowledge of the disease, but that they should share that knowledge with members of the community. However, *being Yolngu* was said to be the cause of ignorance about this disease, by a Yolngu woman (a relative) working in health education.

Relative At the moment Yolngu don't know these stories. They don't know the full story. They want to know about the disease... Because we are Yolngu people we don't learn much about this disease. Only *Balanda* people can let us know. (Interview 15)

Of particular import to this study was patients' and family members' perception of the *Bicillin* injections. The injections were generally seen as therapeutic rather than preventive (see section 16.1.2.). There may be a community memory of some of the earliest treatments administered by missionary nurses working in Arnhem Land like the powerful and painful injections that cured diseases such as yaws (Kettle 1991). Patients described a feeling of well-being following an injection, and an internal sense of derangement when an injection was delayed. Many respondents regarded missing injections or tablets as the cause of deteriorating health. A few respondents described the injections as killing the germs that cause the heart disease. Yet health workers were concerned that *Bicillin* had a long association with the treatment of impetigo, and therefore was not seen as an appropriate treatment for RF.

No respondent described any traditional treatments for RHD, although a number of people spoke of remedies for cuts and sores, sore throats and other minor ailments. General measures such as a healthy diet with bush food, exercise and healthy living were all thought to be beneficial.

Thus it appears that a Yolngu RF story contained elements of personal experience, intuitive deductions and medical explanations, although the latter may be rendered unrecognisable via the process of imperfect cross-cultural communication. The *Balanda* disease story was hardly more cohesive, which was not unexpected because of the inexperience of the staff. Furthermore, *Balanda* staff perceived Yolngu as having a congruent anatomical and physiological system to their own, when this was not the case. Yolngu, on the other hand, suspected *Balanda* of hoarding their medical knowledge, and this was true to some extent.

16.2.2 The relationship between knowledge of the disease and ‘getting the needles’

People co-operated well with treatment, but a large proportion of the community did not yet believe the facts relating to the mode of infestation by this parasite (hookworm), despite much and varied teaching. This greatly retarded the control of the disease.

Northern Territory Administration, Welfare Branch, Annual Report, 1967-1968 (Berndt 1982)

What observations can be made regarding the relationship between a biomedical knowledge of the disease and its treatment and getting the *Bicillin* needles? A relative

contended that it was lack of knowledge about the disease that led her daughter to be ambivalent about seeking out treatment. Staff concurred that knowing about the disease was a crucial factor for engaging patients in their treatment, and also for patients to take responsibility for their health (see section 16.3).

However, there was little evidence to suggest that knowledge of the disease was a prerequisite for getting the needles, or that the lack of knowledge prevented patients from accepting treatment. Most patients received most of their needles; few patients had a solid biomedical understanding of the disease. Patients who received few needles seemed to know as much about the disease as those who received theirs regularly.

Interviewer Did they tell you that you have heart disease?

Patient No. I didn't know.

Interviewer Until when?

Patient Until 1991, then I knew. I asked them, "why did I get all these injections, what for?" Then the doctor told me, "its for your heart that you are getting this injection." (Interview 17)

Patient I don't think about it. I just accept the injection, that's why. (Interview 16)

Although knowledge of the disease was not directly related to compliance, there were some connections. As suggested in 16.1.2, the belief that the disease was chronic and serious was a causal condition for accepting treatment, along with trust in the health

service. This trust was, in part, founded on a history of good communication between patients and staff. Patients valued the effort and time staff took to communicate medical stories to them, yet did not necessarily concern themselves with retaining the information conveyed. Perhaps by giving a medical explanation of disease, the health staff earned the authority to decide the treatment, as knowledge, in both cultures, is associated with power.

Unlike the finding in the Kimberley that patient's desired more information about their disease, it was uncommon for Yolngu patients to spontaneously request information, and some directly declined (Mincham, Toussaint et al. 2003). Whilst regional variation may account for this difference, no definite comparative conclusions can be drawn between these two sets of qualitative data.

16.3 Communication

16.3.1 Communication about illness between community members

Communication between community members about illness appeared to be restricted. Cultural protocols dictated the content of communication about disease, as well as to whom the message could be communicated. Few respondents admitted to knowing other people's medical histories, including major operations. Participants were reticent to talk about unrelated community members, and provided polite but negative responses to questions in that area. It is possible that communication between community members was freer than it appeared, but that respondents were not willing to reveal their knowledge in the context of a recorded interview with a *Balanda*

researcher. Respondents rarely indicated a curiosity about other patients; only one respondent openly admitted wanting to hear about other peoples' experiences.

Patient Like me, I didn't get my operation, but the other people got theirs. So I would like to know why those people got their operation, and where.

(Interview 3)

Sanctioned pathways for communication existed between the doctor, the patient and specific members of their family – primarily the 'parents' (which included the sisters and brothers of the birth parents), the paternal aunts and the maternal grandparents. There are specific restrictions on the communication between brothers and sisters, including information conveyed by a third party. Reproductive issues should never be discussed with different sex siblings, but there was also a sense of restraint in discussing other health matters. For example, my co-researcher initially declined to interview her 'brother'.²⁰ Later, on reflection, she decided that it should be acceptable to discuss matters relating to RHD, and the interview proceeded without any obvious tension.

The secrecy surrounding disease existed even between some close relatives and was either the cause or the result of conflict. Two 'mothers' expressed dismay that their adult son excluded them from his circle of confidence, thereby preventing them from participating meaningfully in his health care.

Interviewer How did he feel himself, or describe himself, your son?

Relative 1 He didn't. Only when his doctor comes in does he talk about himself to the doctor. He doesn't even tell us how he feels, he only talks to his doctor. We ask him whether he can share his story with us so that we can talk to Zinta and Joy when they come around, but no.

Relative 2 He says “no, I will only tell this story to my doctor and Zinta and Joy”.²¹ (Interview 10)

Another patient did not keep her mother informed as a retribution for the perceived lack of care. These observations suggested that some young people withheld their personal thoughts as a means of exerting power over their relatives. Information, including personal details such as symptoms and feelings, seemed to have a value and significance in this Yolngu community that exceeds the norm within *Balanda* society.

Death was generally avoided as a topic of discussion. The younger the interviewee, the more curt their responses to discussions about death. An older patient confirmed sanguinely that she was not afraid to discuss death, but that it was “no good” to discuss it with other patients. I was aware of at least two people who had died within the past few years directly as a result of RHD. Two respondents only admitted to knowing about a death secondary to RHD, but declined to talk about it. My co-researcher elaborated that the cause of death was usually not discussed within the community, unless there was grounds for the suspicion of misadventure.

²⁰ Even though the respondent was not a close blood relation, the fact that he was a brother (*wawa*) in kinship terms was the critical issue.

²¹ We took this statement as encouragement to seek out this patient for an interview, however he subsequently declined to participate.

In contrast to the reticence to engage in idle gossip, respondents indicated that talking about illness was acceptable if the reason for doing so was to ‘encourage’ or ‘help’ (*gunḡa’yun*) others through the telling of personal stories. One parent of a young girl who had undergone cardiac surgery had been approached on two occasions by families seeking advice on the matter of heart operations. Patients and families appreciated hearing about personal experiences when making important decisions.

Parents and health workers stated that the telling of stories would be beneficial to the speaker and listener alike. They were in favour of a ‘social night’, where patients could get together, share their stories, and learn about healthy living and RHD from health staff. These events should be held on a recurring basis, to reinforce the message.

Relative If we hold these social nights, it should help. Like the kids get shy, they should feel better through this. In this way they can help each other: so they don’t get frightened, so they get confident. (Interview 6)

However, whilst theoretically in favour of a support-group meeting, even the most outspoken patients wouldn’t commit to telling their own stories at such an event. Most patients indicated that they would only feel comfortable speaking to relatives.

Watching the way young patients reacted to the interview process convinced me that they were unlikely to speak up in a public forum. One mature patient made clear the link between helping others and talking about your own illness, with his sad comment about the effect of the culture of privacy amongst Yolngu:

Patient Yolngu people don't help each other, maybe. When people get sick, we don't talk about it. (Interview 16)

16.3.2 Communication between *Balanda* health staff and Yolngu patients

Marngitj, the Yolngu word for doctor, means person with knowledge and builds on the stem word “*marngithirr*”: to know. The transmission of knowledge from health care practitioner to the patient and their family was a highly valued aspect of the clinical consultation, beyond the actual content of the communication. Doctor-patient or nurse-patient communication had its own cross-cultural flavour, and was affected by the context, the technique, and the content of the communication, as well as the identity of the speaker. There appeared to me to be evidence of good communication occurring between the *Balanda* health staff and some Yolngu, who could recount convincingly what doctors had said to them, years earlier. At other times, I was reminded of Trudgen's warning, that a superficially cohesive narrative, especially when using technical language, may hide the most profound lack of understanding (Trudgen 2000). A well spoken patient referred to “something” pumping in her body, apparently unaware of the role of the heart in her symptoms.

Patient When I go up to see the doctor and he asks me how I am I say “alright”, but there is something pumping. It is pumping slowly, but if it gets fast the doctor can slow it down. (Interview 5)

Speaking with experience in cross-cultural communication research, one relative stated vehemently that communication between *Balanda* health staff and Yolngu patients was a problem.

Relative But in this area, with patients and doctor, the communication isn't working. They don't understand each other. Sometimes the patients get angry or upset. The doctors use a lot of medical words. (Interview 15)

Another relative stated that a patient (her son) and health staff inherently misunderstood each other, recognising perhaps the difference in world views of most *Balanda* and Yolngu. Health staff are equally pessimistic about the effectiveness of their communication with patients, and some stated that improving communication, for example through the use of interpreters, was made difficult by poor resource availability.

Staff, patients and relatives made recommendations for improving the process of communication, with attention to the context and content of communication as well as the techniques used. The location of the interaction was relevant: the patient's home was the preferred locale for communication for many. However one respondent stated that the home was inappropriate for many young people who were unlikely to feel comfortable speaking up before their relatives. If a social night was to be held, food should be provided to improve attendance and attention. Another aspect to the context of the communication was the individual context for each patient, in terms of his or her disease severity. Both doctors commented that it was harder to communicate with (that is convince) an asymptomatic patient than one who presented with symptoms. Thus the patient's condition affected the content of the communication.

Staff differed somewhat in their approach to what content to include when communicating with patients about RHD. The visiting specialist concentrated on conveying a biomedical explanation of the disease in order to justify the treatment prescribed. Nurses also presented the physiological basis to treatment, but were concerned that the content of the communication should be adjusted for ‘culture’ as well as the person’s personal context.

Nurse ...because I am not one hundred percent sure of the culture, how much people understand. And the stories that we tell them, whether that is really what they want to hear? Or how much understanding they have of the story, and how pertinent and important it is to them at that time of life anyway?
(Interview 21)

A simplified but practical explanation provided by a health worker emphasised the anatomical location and the possible outcomes of the disease, but did not include disease causation. The local doctor de-emphasised the biomedical explanation in favour of the social implications for the patient, believing that this was a higher priority for Yolngu patients. Yet patients were interested in the cause of the disease. It was not uncommon for patients to repeatedly request an explanation of the cause of RF, suggesting that the explanation we provided in both languages (an autoimmune response to streptococcal infection) was not perceived as full or cohesive.

It was considered appropriate to discuss the possible serious consequences of the disease, but it was suggested that this be couched in positive, encouraging terms, such as: “you have a serious disease, but we can help you”. Patients and relatives certainly

appreciated hearing good news from health staff. The possibility of death should not be discussed with patients, we were told, but may be mentioned to family members. Scaring patients into complying with treatment was considered improper and damaging, and had the possible consequence of frightening the patient away from further contact with health services.

In terms of the technical aspects of communication, patients and staff expressed a preference for interpreters to be present during clinical interviews. Competency in *Yolngu matha*, and awareness of non-verbal language and cues were recognised as relevant to good communication. Characteristics of the messenger were also thought to be important: the doctor relied on his “level of acceptance as a healer” (Interview 2), while a health worker suggested that ‘closeness’ with the people facilitated communication. A nurse suggested that there were ‘right’ and ‘wrong’ people to deliver certain messages, and this was in part determined by the cultural mores that surrounded communication.

Visual materials such as anatomical models, diagrams, videos and electronic resources were highly recommended as communication aids.

Health worker Sometimes we just tell them the story, just talking, and they say “What? What?” And if we talk and show them pictures at the same time it could be OK. (Interview 22)

Strikingly, very few respondents spoke of the importance for *Balanda* health staff to understand their patients. Only one respondent hinted at the bi-directional process of communication:

Relative For it is hard for you to understand what he is thinking, and it is hard for him to understand what you are thinking. (Interview 18)

The one sided focus on the process of communication between patients and health staff has multiple origins. Firstly, it comes from the power difference between the parties, and the perceived importance of the information conveyed by the doctors and nurses. Secondly it may also be fed by a major cultural difference in communication: the care with which Yolngu people convey information, based on their sense of privacy, and the value of personal information, as well as their discomfort in being interrogated. Many a patient has instructed me to look in their medical record as a way of avoiding the need to engage with a question.

16.4 Responsibility and blame

16.4.1 Who is responsible for what?

Table 20 summarises the responsibilities of the relevant parties from each perspective and highlights some of the differences and potential areas of conflict.

Table 20 - Perceptions of the responsibilities of the patient, the family and the clinic

| Perception of responsibility | Of the patient | Of the family | Of the clinic |
|------------------------------|----------------|---------------|---------------|
| | | | |

| | | | |
|-----------------------|---|--|---|
| By the patient | Variable | Care for the patient physically (<i>djāka</i>) | Provide transport Deliver medication Perform check-ups |
| By the family | Choose a healthy lifestyle Listen to advice from family | Care for the patient physically (<i>djāka</i>) Encourage the patient (<i>gungā'yun</i>) Make decisions about health | Provide transport Deliver medication Perform check-ups Communicate with the family |
| By the clinic | Present themselves for medication Comply with treatment Volunteer symptoms Make decisions about health | Care for the patient physically (<i>djāka</i>) Support the clinic Encourage patient to comply with treatment Help the patient make decisions about health | Legal duty to provide health care Responsible use of resources |

The interviewed patients varied in their sense of personal responsibility from a sense of distinct independence, to a reliance on family and health staff to provide every component of their health care. Most patients had an expectation of physical assistance from their families, and expressed their disappointment and even anger when they perceived this was lacking. The majority of patients saw the role of the clinic as delivering health care, literally, to their door. Even when patients knew when their injection was due, and had a short distance to negotiate to get to the clinic, they expected a ‘reminder’ and transport to be provided. This expectation is congruent with WHO recommendations for the care of patients with RHD and was met, more or less, whenever possible (Anonymous, 1995b). However, when the patient was outside the community, in Darwin for example, the health service was unable to meet this

requirement, and the balance of responsibility fell back to the patient. Patients were mostly unused to keeping track of dates, organising their own transport and appointments, and so they tried unsuccessfully to deflect the responsibility for organising the injections back to the local clinic. Thus, when patients were home, there was no great conflict over whose responsibility it was to ensure health care, but when patients left the community, responsibility was relayed between the parties, with family members seeming to maintain the greatest concern.

Families expected patients to live a healthy lifestyle, more so than the average community member. They also put pressure on patients to accept their advice and direction. One family member implied that she could overrule her adult son's refusal to consent to an operation, although it was unclear how she would have done so (interview 18). Relatives and health staff expected patients to comply with prescribed treatments. Some patients did this by accepting all treatment when offered; others actively and independently sought out their injections and tablets.

Interviewer Does anyone go with you (to get your needles)?

Patient I go alone.

Interviewer Do you go by yourself or does the health centre pick you up?

Patient I go by myself.

Interviewer How do you know when is the right time to go to the health centre for your needle?

Patient I go every new moon. I go by myself. I never missed any needles.

(Interview 9)

Likewise patients were expected to listen to the advice of their families. Conversely, becoming mature, independent, and making decisions for themselves was also recommended. In a harmonious system the patient makes his or her decision taking into account the advice from the correct people. If this protocol is followed, then the decision will be respected.

Families, as a rule, accepted the responsibility of making the important decisions about a patient's health, such as consenting to operations. While the decision makers differed from family to family, there was little conflict within the family as to who was responsible - typically the maternal grandparents or maternal uncles, and often a small committee. However, there was potential for conflict within a family over the allocation of the authority to care for a child.²² Family conflict was implicated as a causal condition for accusations of blame (discussed further in section 16.4.2).

Relative ...there is a lot of family looking after her. She has a lot of family, a lot of mothers, but it's lies: they don't help her [birth] mother ... there are a lot of bosses. If something happens to her, her [birth] mother might get into trouble from family, or from the police, or from the health centre. (Interview 13)

The family expected the clinic to provide comprehensive health services including home visits. They particularly stressed their expectation that clinic staff communicated regularly with the family.

Although it appeared that the health staff had cheerfully taken on the responsibility of providing an active recall system for *Bicillin*, many commented that patients should present to the health centre themselves. At Ngalkanbuy, patients were actively recalled for mental health reviews, antenatal care, and, on occasion, for immunisations. Patients with chronic disease, failure to thrive, or on long term medication were not systematically recalled. Thus it could be said that the patients with RHD were being prioritised, receiving a significant share of the clinic's resources.

Balanda health staff stated that a biomedical understanding of the disease was a causal condition for patients accepting responsibility for their health, but that 'Yolngu culture' was an intervening condition. Nurses spoke of the need to 'cajole' both the patients and the health workers to carry out their expected roles. Similar actions that might be seen as paternalistic have been a feature of the history of colonialism in Australia. This is not to suggest this approach should be abandoned just because its paternalism replicates the colonial encounter, as the benefits for the patients may outweigh the implications of the approach. In any case, this 'cajoling' of patients and staff could be seen as similar to the Yolngu concept of encouragement or *gunga'yun*.

Health staff struggled with these issues of accepting and deferring responsibility. The visiting specialist proposed "the promotion of individual responsibility, but with adequate fallback", hoping to foster patient autonomy, but believing that patients who fail to present for Bicillin needles, for example, should be actively retrieved (Interview 1). This position appears to give patients the choice of complying with

²² A woman complained to me that her sister had taken her son away to another community. The sister

treatment voluntarily or being strongly encouraged to comply. Patients were only permitted autonomy when their actions conform to medical recommendations. A nurse indicated how the allocation of responsibility was an unresolved issue at the clinic:

Nurse It's a challenge to take responsibility for your own health, and it's a challenge for the staff of a health service to follow that through. And where do you start, and where do you stop, and where do you – you often juggle this in your mind – where do you think that people have to *own* their illness, you know. Sometimes I feel we disempower people, you know, by pushing them to do things that they would rather not do, you know, like the *Bicillins*. I wonder about that. (Interview 21)

Health staff expressed respect for patients who 'took control' of their lives, but maintained reservations about the ability of patients to do so independently in difficult circumstances, such as when facing surgery. Health staff felt that they should 'work together' with the patient, 'help them', and not 'give up' on them. This implied that health staff might not be entirely ready to relinquish control over patients despite the lip service they paid to this ideal.

16.4.2 Illness and blame

A consequence for taking responsibility is receiving blame when a negative outcome is reached. A search for 'who is to blame' takes place after many of the deaths in Galiwin'ku. Accusations of inadequate care are common, and may be levelled at the

had accused the birth mother of inadequate parenting, hereby giving her custodial rights over the boy.

family or the health staff, but rarely at the unfortunate individual themselves.

Analogous to the Western custom of not 'speaking ill of the dead' Yolngu were unlikely to blame patients for their poor health until other (more sinister) reasons were excluded.

Health worker 1 A long time ago, a person passed away. When we heard that this person had passed away, then the health workers got the blame. The parents blamed us, that she hadn't been looked after properly. This was one of our family and she had the same problem: RHD...

Interviewer And how did that resolve? Was there a fight?

Health worker 1 No fight, just an argument. And then we found out that it was her: that she was in the wrong.

Health worker 2 The mother's side were OK. It was the father's side who was making trouble. Now her mother and father know what happened to her, what was wrong with her. (Interview 23)

Occasionally, wrongdoing by another community member via sorcery is suspected.

Events that were perceived as particularly suspicious were sudden or unexpected deaths, but also deaths in young people even when a serious condition was present.

Unexpected deaths also caused suspicion of outright murder or euthanasia.

Community members suggested that a coherent, convincing and detailed medical explanation for a patient's illness went a long way towards reducing such accusations.

16.4.3 A discussion of the intersection of “taking responsibility for health” and “staying well”

Patient I teach myself how to look after myself, look after my body. I don't rely on other people to look after me. (Interview 5)

Nurse I remember one person saying “If you want to give me an injection you have to find me, because I am only doing it for you”. She didn't think she needed injections. She thought she was doing us a favour by letting us give her an injection. (Interview 22)

In the first example, the expression of autonomy describes the ideal patient for health staff. Or does it? This patient also selectively missed treatments, smoked cigarettes, and rarely presented for check-ups at the health centre. The patient described in the second quote, who received all her treatment at home, didn't do anything without family facilitating the event, expected reminder cards for check-ups and required transport to the health centre, may be the bane of the health workers' lives, yet received optimal medical care. The necessary condition for patients to receive good health care was not that *they* took on the responsibility, but that someone did.

A shared understanding of who is responsible for caring for a patient is necessary in the effective collaboration between patients, their families and health professionals. When opinions diverge as to who is responsible, then conflict and poor outcomes for the patient occur. Will RHD patients take up the challenge of coordinating their own care in the near future, as per the wishes of the health centre staff? It seems unlikely. Whether an incentive such as money or a gift would increase the number of patients

who present themselves to the health centre for injections is unknown. School students in Galiwin'ku receive a gift each year for perfect school attendance, but the effect of the incentive appears not to be sustained over a prolonged period.

16.5 Religious beliefs – Christianity and Yolngu spirituality

An initial impression gained from the interviews was that the Yolngu people of Galiwin'ku had absorbed Christianity, as introduced to them by Methodist missionaries, into their worldview. Depending on the value judgement of the observer, the adoption of Christian beliefs could be seen as a contamination of traditional culture or a progressive evolution. For participants in this research, faith in God was a solace in hard times. Many patients and relatives referred to prayer as recourse in times of illness. Prayer was seen as having the potential to effect a cure for patients, improve the performance of the attending medical staff, and prevent further illness. Thus prayer was often the initial response to becoming unwell, before seeking medical assistance.

Because of these strong impressions I wondered whether faith in God was an impediment to seeking out health care. Two respondents directly denied this possibility, but in one case a patient did not “comply” with medical advice, instead accepting a fatalistic interpretation of Christian teachings. She probably died a premature death as a result of this attitude.

Nurse She just didn't take it seriously. She was the most joyous person; she had the most beautiful nature. She used to say: “it's alright about those tablets,

because I believe in God, and he will help me through all this, and don't worry about it so much!" (Interview 22)

On further reflection of the role of Christian beliefs in Galiwin'ku and the guarded references to sorcery, I wondered whether faith was used as a weapon against sorcery. This could be suggested by the fact that as there were reportedly no traditional healers (*marrgitj*) remaining in Galiwin'ku, causing patients to turn to the Christian faith for the spiritual healing they needed to combat illness of spiritual origins. This might explain why the initial response to illness for some patients and relatives was prayer. Reid also found that prayer was being used to counteract the effects of sorcery in this quote from a patient in Yirrkala:

They took me to hospital and operated and found I had appendicitis. It was due to *manggimanggi* (sorcery). I wasn't afraid though, because I was praying. (Reid 1983 p127)

Thus references to prayer in these interviews may contain allusions to 'traditional' Yolngu spirituality.

17 A practical outcome? Feedback to the community and the constitution of a reform group

Relative Like you people, you are good people helping us with your work. It's really true. Especially for those people with heart disease. (Interview 13)

After the completion of the interviews and a preliminary analysis of the translated transcripts, a meeting was convened to discuss the results of the research and proved an opportunity for respondent validation. Research respondents, as well as other patients, their families and health staff were invited. About forty-five people attended, many of whom had not participated in the interviews. A few older men who had not participated attended the meeting. Many of the female patients were present, including some regarded as compliant, but also some who were perceived as less compliant. A patient who had declined to participate in the research nevertheless attended the feedback meeting.

The findings were presented in English and *Yolngu matha*. We limited the feedback to “impediments to ‘good care’ for RHD patients” and “participants’ recommendations for patient care”, hoping to verify the research findings and stimulate a useful discussion. Very little discussion arose. This may have reflected the poor acoustics of the meeting venue (the health centre veranda), but also lent weight to the finding that Yolngu do not readily discuss personal issues in a public forum. The lack of critical comments cannot be taken as tacit support for the findings of the research.

One exception was the input from the mental health worker, who said that the issues raised by this research reflected some of the problems she experienced in her clinical practice. She stressed the need for a 'culturally appropriate health service'. When asked to elaborate she spoke of the need for having the 'right relationship' with the patient to care for them. She was referring, it seemed, to the need to accommodate kinship laws in clinical practice.

Because of the lack of discussion stimulated during the first meeting, a second meeting was convened in an attempt to transfer the research results more usefully to a selected target group. Vocal patient advocates (in this case, patients' mothers) and Galiwin'ku health staff were invited. Representatives of urban health services involved in health care delivery to Aboriginal patients were also invited.

Unfortunately, the Darwin visitors did not arrive due to 'engine trouble' in the chartered aeroplane, and declined a repeat invitation.

This meeting occurred in a quiet room, away from the health centre, and was much more interactive than the previous meeting. The discussion was steered to address the problem of delivering secondary prophylaxis to patients who were away from home, which I felt would yield results if addressed. The group concurred that there were two main factors relevant to patients who were travelling: the characteristics of the urban health services, and the reticence of patients to contact these services. A major concern was the apparent lack of an active recall system for RHD patients in Darwin, and limited transport services. The group also discussed the option of a hand-held patient record that could serve as a treatment record, as well as providing the phone numbers of the home clinic and relevant urban services.

Following on from these suggestions I (ZH) met with representatives from a number of urban health services. This included two Aboriginal medical services, the government-run community health centre, the RHD register program, cardiology services and a research project also focussed on the secondary prevention of RF. A number of members of this group were Aboriginal, but no remote community members were present. I presented the concerns of the community-based group regarding the availability and appropriateness of the urban services, and the recommendation for a hand-held patient record. This group of service providers enthusiastically made plans to enact change, thus simulating the “reform group” described by Humphery (Humphery and Weeramanthri 2001).

Urban health service representatives stated that they were unable to provide an active recall system for RHD patients with the current level of financing; financial constraints even affected the power of one organization to buy the required *Bicillin*. The group saw a role for lobbying for more funding in order to improve health service delivery. Furthermore, it was considered impractical to institute a recall system in the urban setting where patients were mobile and rarely had phone access. Instead, it was suggested that all remote clinics were informed of the range and location of the urban health service providers, providing patients with a greater choice.

The group also discussed how to increase the ease with which patients accessed unfamiliar health services. The representatives from the urban health services denied that there were significant cultural barriers for remote patients. They claimed to be culturally sensitive, having Aboriginal staff closely involved in patient care. They also

downplayed the issues of access to the health services and waiting times for patients. Although the limited transport options for patients were a concern for all providers, it was considered that a lack of funding prevented this from being addressed. The group did consider that a hand-held patient record, similar to a medical alert card, might help the patient negotiate the first contact of a patient with an unfamiliar health service. The details of the card, its format, and what information it might contain were discussed. The group hoped to secure funding for a trial of such a card. The group planned to reconvene in three months time to review the progress of these ideas.

18 Comments on the research process

18.1 Researcher bias and “going native”

Strauss and Corbin, the experts in grounded theory, warn against ‘going native’, whereby researchers “come to accept the assumptions or beliefs of their respondents”, as this would be a potential source of bias (Strauss and Corbin 1998). I believe that this position is erroneous, failing to recognise that the researcher has culture and assumptions of his or her own. It implies an outdated colonialism, raising the (undoubtedly ‘Western’ trained) researcher to a level of scientific impartiality, an outsider, and not open to influence.

Rather than trying to eliminate the internal biases of the researchers, I preferred to acknowledge them. A major source of bias was also one of the strengths of the research team: the fact that both researchers were part of the community being studied. In different ways we were affected by our location within a kinship network, which inherently played a part in each interaction with Yolngu.²³ We had our own personal experiences, cultural knowledge, and health beliefs: my own originating in a non-Indigenous, urban upbringing. I was educated as a doctor, and came to Galiwin’ku with my partner who was employed by the council as the general practitioner. Joy grew up and was educated in Galiwin’ku in ‘mission time’. She lived in Darwin for a decade then returned to Galiwin’ku. She has worked as a health worker, and also as a researcher in urban youth affairs. She has raised thirteen children, and has close relatives affected by RHD. We each looked at the data from

²³ I had been ‘adopted’ into a Yolngu family before I arrived in Galiwin’ku through my partner’s connections within the community.

our different viewpoints, thereby increasing the veracity of our theorisation ('triangulation'). Whether each researcher's position has equal validity in a cross-cultural situation is uncertain, in fact I wonder about the validity of the role of the 'non-native' researcher in Yolngu communities.

Another aspect to our cultural and social positioning within the community was the likely effect it had on the respondents. Community members saw us as researchers, as representing the National Heart Foundation (which we didn't), as the doctor looking after the RHD patients (which I was), the wife of the local doctor (true again), a health worker (true in the past), as a close or distant relation, as mothers, or as strangers. Participants would have been influenced by these perceptions. It clearly affected who we recruited in the research, as well as the ease with which each respondent participated. It probably affected what was relayed in the interviews as well.

Despite the contribution to bias, I strongly believe that 'going native', in the sense that the researchers immersed themselves in the world of the respondents, was a strength of the research. I suggest that no cross-cultural research should be undertaken without a 'native' guide, and that the elimination of bias, either pre-existing, or acquired, is philosophically flawed, and unattainable.

18.2 Comments on working with a Yolngu co-researcher

The origin of the project, the funding, the timetable and methodology were all devised by the principal researcher. I subsequently sought to engage a local co-researcher. I experienced false starts in making this selection, similar to the experiences of Reid

(Reid 1983) and Nathan (Nathan and Japanangka 1983).²⁴ Whilst I had hoped that JB would subsequently take on ‘ownership’ of the project and that there would be a collegiate relationship between us, the reality fell short of this. This arose inevitably, perhaps, from the different origins of our participation in the research, the differences in our background and motivation. In the end, JB and I had a respectful, consultative and, at times, collaborative working relationship.

JB contributed substantially to the theme list and question design for the interviews. She was invaluable in explaining the project to potential respondents and gaining their consent. JB’s relationship with the participants facilitated many an interview. I benefited from her social integration, her insider situation, and frequently consulted her for advice on local social practices. Conversely, JB displayed some reticence in organising and participating in interviews with people she didn’t know well, where I was less inhibited. She appeared fearful of one potential participant and persistently avoided recruiting him for the project.

We worked collaboratively on the translations. I gained most of my understanding of the interview data during the discussions that ensued around the translation process. JB observed rather than participated in the processes of coding and analysis. Time constraints, her apparent disinterest and my own inexperience prevented me from training her to be an independent analyst. JB acted as interpreter rather than presenter

²⁴ One woman who agreed to work on the project left the community for three months. Another worked with me for two weeks before leaving for a holiday from which she did not return. Becoming anxious about the research timeline I approached the Yalu organisation in Galiwin’ku, which recommended JB. With little deliberation we proceeded to work together on the project. Either by luck or good judgment, JB was indeed a perfectly suited co-researcher who, despite her considerable obligations to family, was able to see the project through.

at feedback presentations, indicating to me that whilst she was happy to be involved in the research, she didn't feel 'ownership' of it.

The community members praised her for the work she had done. JB expressed her satisfaction with the work in a report to the funding body:

This research has really helpful findings for the community, especially for the people who have heart disease. It was a good experience to do this research.

The only problem was that I couldn't do any interviews with my *gurrung* (avoidance relations). I found the work interesting and I learnt something from it. If there is another research project coming in from the Heart Foundation, or other research work, I would be happy to work on it. Everybody was telling me this was good research. A lot of people came and told me it was good research. (Final report to the Northern Territory Department of Health and Aging, 30th October, 2003)

18.3 Deficiencies and difficulties in sampling

There were far fewer men than women with a diagnosis of RHD in Galiwin'ku (consistent with the epidemiologic observation that in the Top End the ratio of males to females with ARF is 1:2²⁵). The difficulty we found in recruiting these few men to the study reflected the hesitation Yolngu had of discussing health matters with members of the opposite sex. This was particularly an issue for JB, who observed Yolngu protocols determining which subjects could be discussed openly between kin. Discomfort was obvious: young men, who had apparently agreed to participate, were

repeatedly unable to be located, or seen disappearing as we arrived at the appointed time. We discussed the possibility of engaging a male co-researcher to overcome the reticence of male respondents. This was more difficult than supposed, as the person selected would need to be of appropriate kinship with JB and myself, have good skills in English, and a basic knowledge of health issues. We did not pursue this approach. Despite the lack of direct evidence from men, a large amount of data were collected about the few male patients, through the participation of their relatives and carers. Multiple viewpoints were gained on various individuals, thereby strengthening the validity of these secondary sources.

Young women were equally difficult to engage. Whilst more cooperative on the surface, giving consent and attending interviews, the profound social shyness they exhibited prevented free discussions, particularly when the recording device was present. Young people constantly deferred questions to their relatives, yet it was considered mildly inappropriate to interview them alone.

Thus, where possible, we encouraged the participation of informal family groups. Allowing the self-selection of people with the authority to speak on the issue of caring for people with RHD had a theoretical justification: it is this group of people who were most relevant in the patient's care, and for whom the research results may yield some benefits. However, self-selection of participants was a potential source of bias. Community members unwilling to engage with researchers, and those with a lack of concern about RHD have valid and potentially crucial viewpoints relating to the subject.

²⁵ Carapetis 2005, personal communication

18.4 Comments on communication issues pertaining to research

Good qualitative research requires a good command of language, and an ability to interpret and analyse language-based data. For most non-Yolngu researchers working with Yolngu not knowing *Yolngu matha* is a major impediment. JB and I had some command of both English and *Yolngu matha* (although JB was clearly more strongly bilingual than I), as did many of our respondents. Interviews were conducted primarily in *Yolngu matha*, which I encouraged, as I was often able to follow the discussion enough to participate. On occasion English was used so that I could be more fully involved. Undoubtedly the accuracy of the narrative deteriorated whenever the speaker used their weaker language.

Interviews were carried out in various locations, mostly either at my house or the home of the respondents. Each had their advantages: my house had novelty value for some respondents, and also provided privacy. Being interviewed at their own home created a more natural, comfortable environment for most respondents, and allowed family members to ‘drop-in’. The recording device (in this case a digital recorder) was both a help and a hindrance. Despite its tiny size, it was rarely unobtrusive to the respondents. It could not be used with some young women who became mute before it. In addition, much communication between Yolngu is voiceless; sign language and gestures are commonly used, as are half-whispers and silent mouthings. The recording device was switched off a number of times when the discussion strayed into ‘women’s business’ or issues of a private nature.

Working with a Yolngu co-researcher helped prevent cultural ‘faux pas’ such as aggressive questioning, or straying into culturally forbidden topics. Even so, some people had difficulty with our questions, especially if we sought opinions about other people, or we attempted to discuss a hypothetical situation.

Interviewer (ZH) If you had kids with this disease, how would you look after them?

Patient Look after them well.

Interviewer (JB) It’s hard to answer these sorts of questions. (Interview 19)

JB would interject at other times when she perceived that the question had already been answered, or that the answer was obvious. It was not uncommon for her to phrase a question in a leading way, an observation also made by Nathan, working with Japanangka in central Australia (Nathan and Japanangka 1983).

Interviewer (JB) We don’t have any bush medicines for heart disease, do we? We have other medicine, but no medicine for the heart. (Interview 16)

We were careful to emphasise the confidential handling of the interview material, and strove not to identify the origins of the concepts we discussed with research participants. An exception was when we felt that it would be acceptable to encourage potential participants by naming previous respondents. Concern was expressed over the confidentiality of ideas expressed in the interviews and their potential inclusion in a book or the publication of names at a conference. Yet the same concerned individual wanted to be sure that his ideas would not be unacknowledged (interview 13). It was

unclear how to tread the line between confidentiality and acknowledging the origins of the ideas in this thesis.

18.5 Translation: the joys and the difficulties

Translation and transcription was carried out as soon as possible after each interview. The recording was verbally translated into English by JB, which I transcribed directly. The process was facilitated by the fact that JB was present at all of the interviews conducted in *Yolngu matha*. The translation-transcription process was an ideal opportunity to discuss the interpretation of the data, as well as pivotal words and concepts. It was at this level that Joy contributed most to the analysis; I relied absolutely on her translation. I altered the English translation as little as possible to make it intelligible to a readership unacquainted with Aboriginal English.

I felt that the concepts that were the most difficult to translate were potentially the most interesting, representing significant differences in worldview. One example was the Yolngu vocabulary referring to kin, many terms not having equivalents in English. More specific to this research, were the differences between *Yolngu matha* and English in concepts pertaining to the body, symptoms, disease and pathology.

Whilst it is commonly assumed that Yolngu have an excellent working knowledge of anatomy, it was my experience that this did not necessarily translate into a full vocabulary of anatomical terms. In my two years of clinical practice in Galiwin'ku, I was unable to find a recognised term for the spleen, and Yolngu patients often named lungs, liver and kidneys incorrectly. Furthermore, there was a lack of congruence of Yolngu conceptualisation and the medical model of the function of the heart and

lungs. *'njir'* refers commonly to pulse as well as breath, and whilst the term *'nojy dupdup'thun'* is onomatopoeic and refers to a noticeable heart beat (palpitations), it can also mean breathlessness. Breathlessness and palpitations occur concurrently during heavy exercise but may occur independently in disease states. Doctors are concerned with accurately identifying medically defined symptoms that Yolngu may have no inherent grasp of.

Another area of incongruence relates to the pathological cause of RHD: bacterial infection. Some work had been done by the Aboriginal Resources Development Services (ARDS) organisation to develop a vocabulary in *Yolngu matha* to enable discussion of microbiology (Anonymous 1995). This work was disseminated some years ago, but there are mixed opinions as to how successful this has been. The term selected to represent 'germs' was *'mewirri'* or worms. This induced some debate amongst linguists, some of whom felt that the loose translation was misleading.²⁶ Nevertheless this is the only term I heard being used by patients and health staff in Galiwin'ku. The term is pictorially reinforced by the worm-like creatures depicted leaping out of the throats of patients in the RHD information booklets (Angeles, Bengier et al. 1996; Angeles, Bengier et al. 1996).

A few other words attracted my attention. The common translation of the word *dhäwu* as 'story' fails to convey any of the gravity that Yolngu associate with spoken information. *Dhäwu* has no elements of fabrication or imagination, although *dhäwu* that is incorrect or contains lies is *'nyäl dhäwu'*. When Yolngu ask for the *dhudhi dhäwu* - the true story or root of the story - they are asking for the underlying essence

²⁶ M. Christie, personal communication.

of a phenomenon. These requests can leave a medical doctor lost for words when they feel that they have already given a full explanation of the aetiology of a disease. The answer probably lies beyond the individual in a larger world view.

Two further words indicating chronicity of disease appeared to strike a chord of recognition amongst Yolngu patients: ‘*dhudhi weyin rerri*’ meaning disease that lasts a long time, or chronic disease, and ‘*binbapthun*’, the propensity for something hidden to reappear. I found that the use of these terms helped to justify the long-term need for secondary prophylaxis for RF.

19 Conclusion and implications

The central concept that emerged from the interviews with patients with RHD and their families was that good care consisted of more than just getting regular *Bicillin* needles. Yolngu wished for a level of care from the health centre that went beyond what is provided by a standard medical service. RHD patients wanted their health service to provide holistically for their well-being, just as the aged care program cared for old people. Essential components of such holistic care for Yolngu included physical care – expressed by the term *djäka* - as well as encouragement and guidance – *gunga'yun*. Patients wanted food, transportation, ‘reminders’, ‘full’ check-ups, medication and *Bicillin* needles. As well as this they wanted their carers to be reliable and professional, but most of all familiar, *like family*. Good feelings were generated through home visits and also through the friendly and encouraging behaviour of staff.

Patients *belonged* to their clinic, just as they belonged to their families and clans. A strong sense of attachment to their home clinic was described, although this didn't prohibit quarrels and grievances between the parties. The bond between patients and their health centres persisted when the patients left their community. Thus the health centre could be seen as the extension of the family, and in some cases it *was* family. Sometimes the very closeness of the relationships between the patients and the health staff interfered with the delivery of good care. On the other hand, patients without close relationships with health staff were also at risk of receiving a lesser standard of care. It is possible that this strong feeling of connection was unique to this particular clinic, and might reflect the isolation of island life, the community's missionary origins or even simply the personalities involved, but I don't think so. In discussions

with other health care providers across the Top End, many recognised the dependency of patients for their own home clinic, in both urban and rural settings.

The concern that originally drove this research - the perception that the high rates of recurrent RF was due to patient non-compliance with *Bicillin* needles - was revised. As a rule, patients did not reject or refuse treatment. Failure to get needles was not, as commonly believed by health care providers, related to patients' perceptions of the painfulness of needles. Nor was a convincing connection made between knowledge of the disease and the getting the needles. Instead, most patients followed the instructions from health staff although they varied in their approach.

Many patients conferred the responsibility for providing their health care onto the health staff. Accustomed to a paternalistic health service and feeling entitled to special care because of the seriousness of their disease, many RHD patients waited (patiently) for their injections, check-ups and home visits. Any delay or omission was regarded as failure on the part of the health service. Other patients actively sought out health care, and took responsibility for ensuring their own medication and clinical reviews. These two 'groups' of patients were not clearly distinct in terms of their demographics, the severity of their clinical condition, or their knowledge of the disease. Thus patient characteristics leading to non-compliance could not be readily identified. Indeed the assumption that patients are obliged to accept medical advice rests on an outdated notion of physician supremacy, and generates a habit of blaming patients when advice or treatment is not taken up.

To overcome the limitations of this one-sided view we looked at the process of service delivery. Whilst generally performing this task effectively and graciously we found that, at times, the health service failed to deliver *Bicillin* needles for reasons such as lack of resources, ambivalence, inexperience, and oversight. The most glaring omission was the failure to look after patients who had left the community, due not only to the lack of responsibility that clinic staff felt for patients out of their direct reach, but also from the practical inability to accommodate patients' lifestyles and needs. Patients expected the home clinic to organise their health care when in Darwin, but health staff, unaware that urban services did not provide an active recall system, rather easily relinquished any sense of responsibility they felt for the patients. On the other hand, urban service providers were unable to simulate the familiar, supportive, encouraging service that patients were used to. Urban health services require patients to be responsible for booking their own transport and for initiating appointments. Thus patients from communities became disconnected with all health services, and failed to receive proven, effective treatment.

School-based RF control programs are recommended by the WHO and have been shown to be effective in some communities (Anonymous 1980; Iyengar, Grover et al. 1991). Aboriginal children attending boarding school in Darwin receive their prophylactic injections via a school visit by a nurse. However, in the remote community of this study, the rate of school attendance is so low that a school-based program is not warranted, and by far the majority of patients were adults.

Following on from this, two broad approaches could be taken to improve the uptake of prophylaxis for recurrent RF. One approach would be to refocus the health service

to provide better holistic care: a better administered, more communicative, more responsive and resourceful service. This should not need to be justified with proof of a better biomedical outcome (although health funding is often linked to this); Yolngu should set the priorities for their own health service. For Yolngu patients, better care is not measured in terms of the number of needles administered, but in a sense of well-being, and the feeling of being nurtured. This vision may well lead to downstream improvement in biomedical outcomes, if patients respond by working more closely with their health centre.

A second approach would be to concentrate efforts on the group of patients with the poorest level of health care. In the case of patients with RHD, we identified patients who travelled as at most risk of missing out on adequate care. The root cause for this situation was the failure for any party to take responsibility for their care.

Geographical translocation redefined relationships and responsibilities for staff, but not necessarily for patients. We convened a 'reform group' to address this particular problem and hope to see some changes implemented in the near future. Numerous obstacles to improving the urban health services for patients with RHD were identified, including lack of funding to expand the program, the impact of Yolngu clan and language group on their interactions with other Aboriginal people and each other, the difficulty in accessing relevant medical records on remote-living patients, to name a few.

Administrative oversight was another cause of failure to deliver health care. A centralised RHD register alone was not sufficient to ensure that patients weren't 'lost'. It requires commitment and perseverance from staff to maintain administrative quality

when clinical needs are so compelling. This is a common problem in all areas of patient care, not just in small, chaotic Aboriginal health services, and requires constant vigilance

There is no reason to believe that these conclusions pertain only to patients with RHD. Chronic disease, pregnancy and mental illness all require ongoing cooperation between patients and health staff, and often involve issues of compliance with medical treatments. Based on this research, clinicians could usefully concentrate on their own practice, the way they relate to their patients, the way they communicate with them, and the focus they put on holistic care- recognising not just the physical care but also emotional care needs of their patients - rather than on specific therapies. It is imperative for clinicians and all patients to openly negotiate the responsibilities of each party, and discuss contingencies.

I did not discover a cohesive and universal Yolngu explanation for RF or RHD. A purely indigenous or 'pre-contact' narrative was not elicited. Contemporary Yolngu have combined notions of bodies, health and sickness from Western culture and their own paradigm to form a mosaic picture that appeared rather unstable. Patients and their relatives did not place much stress on obtaining a western understanding of the disease. When a biomedical explanation for the disease was proffered, patients and relatives often found it unsatisfying, or unconvincing, and continued to ask for the *root cause (dhudhi dhāwu)* of the disease. This is not to say that communication was irrelevant or unnecessary. On the contrary, communication from health care providers was highly valued (and indeed ethically required), and provided it was carried out in a culturally appropriate way, was the means of earning authority as a healer. Even so,

relatives valued the information relayed by patients as much as that from health care providers. Private knowledge, irrespective of its source, was valuable.

I cannot generalise about people beyond Arnhem Land, or even beyond Galiwin'ku. Although there may be a temptation to homogenise the people of East Arnhem Land based on the similarity of languages and traditions, Yolngu tend to emphasise their differences. Nevertheless the concepts of *djaka*, *gunḡa'yun* and belonging to a health service have an intuitive veracity that might prove to be generally relevant.

Appendix

Yolngu matha orthography and pronunciation

I have adopted the orthography developed by Beulah Lowe that is commonly used for *Yolngu matha* languages. ‘Yolngu’ and ‘Ngalkanbuy’ are treated as proper nouns and remain in the Latin alphabet in this thesis, but might be more correctly written as *Yolṅu* and *Ṃalkanbuy*. ‘*Yolngu matha*’ means Yolngu language.

Vowels

| Short | Long |
|-------|------|
| a | ä |
| i | e |
| u | o |

Consonants

| Stops | Retroflexed | Interdental | Lamino-alveolar |
|----------------|-------------|-------------|-----------------|
| b | | | |
| d | ḍ | Dh | dj |
| g | | | |
| k | | | |
| p | | | |
| t | ṭ | Th | tj |
| ‘ | | | |
| Nasals | | | |
| m | | | |
| n | ṇ | nh | ny |
| ṅ | | | |
| Liquids | | | |
| L | ḷ | | |
| rr (trill) | r | | |

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