Congenital talipes equinovarus in Papua New Guinea: a difficult yet potentially manageable situation

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Abstract Reports in the literature have suggested a high incidence of congenital deformities, including congenital talipes equinovarus (CTEV), in many Pacific Islands. This study performed a retrospective analysis of cases of CTEV in an isolated region of Papua New Guinea over a 2-year period. Data was collected on the incidence of CTEV, together with an analysis of initial treatment and outcome. The incidence of CTEV was 2.7 per 1,000 live births per year. A peak incidence of CTEV births in September suggested that maternal anaemia secondary to malaria was a significant risk factor. Good functional outcome was confirmed in only 20% of cases following initial treatment. The authors suggest the Ponseti method as a realistic option for treating CTEV in this region and that it could be instigated with minimal resources and training.

Résumé De nombreux rapports montrent la fréquence importante de la déformation en pied varus équin congénital dans beaucoup d'îles du Pacifique. L'étude actuelle a pour but de réaliser une analyse rétrospective du pied varus équin congénital dans la région Papouasie Nouvelle Guinée sur une période de deux ans. Toutes les données ont été rassemblées sur le plan épidémiologique ainsi que sur le plan du traitement et de son évolution. L'incidence du pied varus équin congénital est de 2.7/1,000 naissances par an. Il existe un pic plus important au cours des naissances du

mois de septembre faisant envisager la possibilité du rôle néfaste de l'anémie après malaria. Les résultats ont été rapportés dans seulement 20% des cas. Les auteurs suggèrent que la méthode de Ponséti sont une des options thérapeutiques du pied varus équin congénital soit appliquée et enseignée de façon large.

Introduction

Musculo-skeletal deformities can have major consequences for the affected individual, frequently resulting in significant disability. This is particularly pronounced in developing countries, such as Papua New Guinea (PNG), with a high rural, self-sufficient population, coupled with poor health and welfare services. The incidence of congenital musculo-skeletal deformities, in particular, congenital talipes equinovarus (CTEV), has been shown to be higher in individuals from nearby Pacific Islands, Maoris and Aboriginal groups [4, 8, 11, 22]. However, it appears that very little is known about the frequency of such disorders in PNG itself. There is some evidence to suggest that CTEV is a problem in PNG, but this does not appear to have been explored in any depth [11].

Conditions for children in PNG continue to be difficult; infant mortality is 77 per 1,000 and a health budget of only US\$10 per head of population limits the help that can be provided to conditions demanding intensive medical and surgical input [12]. This becomes particularly apparent when treating a condition such as CTEV. The excellent outcome seen in children treated in the western world can largely be attributed to early recognition of the disorder and effective treatment regimes, such as that seen in the Ponseti method, commonly practised in the UK. This method of serial manipulations and castings with or without Achilles

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tenotomy and subsequent orthotic management requires well trained medical personnel, parent education, orthotic supplies and surgical facilities, all of which are taken for granted in the western world [9]. The Ponseti method is designed to re-align the foot to alleviate pain and deformation, whilst allowing plantigrade weight bearing with adequate joint motion, providing excellent functional outcome and reducing the need for extensive surgery [17, 18].

The aim of this study was to explore the burden of CTEV in the East Sepik region of PNG. Together with the collection of epidemiological data, the study set out to gain an insight into the treatment capabilities, prognosis for affected individuals and cultural attitudes towards deformity.

Materials and methods

We conducted a retrospective analysis of children admitted and treated for congenital deformity (any deformity present since birth) and whose date of birth fell between March 2001 and March 2003. This study was carried out during August 2004 at Wewak General Hospital, PNG. Wewak General Hospital is the main referral centre for the East Sepik Province and Manus Island, serving a population of approximately 350,000.

Patient identification

Meticulous written records of obstetric care, births, including birth defects, hospital admissions and surgical procedures are kept routinely in Wewak General Hospital. Hospital logbooks were examined by hand and the available data for each patient with CTEV was recorded. Only CTEV in live births were included. To insure the inclusion of all individuals and the monitoring of follow up hospital unit numbers, the patient names and dates of birth were recorded and verified against surgical records, paediatric ward logbooks and physiotherapy logs. The patients in the study were either born within the hospital or presented later as hospital admissions.

Data collection

Only infants born at term ± 14 days were included and this was verified by obstetric logbooks. Data were obtained regarding sex, date of birth and diagnosed abnormality (recorded by the paediatric registrar either on initial baby check or on presentation to the hospital). Hospital numbers were used to identify patients and prevent duplication. The treatment for each patient, including type of surgical procedure performed, was recorded. Not all children in the study were born in the hospital—many were born in the community and presented to the hospital at a later date.

To calculate the incidence of deformities, census data was used to provide the total number of live births within the region served by Wewak General Hospital.

Supplemental data

In order to place the difficulties faced by affected individuals and the health services into context, it was important to gain an understanding of health beliefs and customs in the region. Discussions and structured interviews with a spectrum of health professionals (outreach workers, immunisation doctors and local health workers) and carers were vital to this process. The opinions of those interviewed were categorised and each individual could list the three most important factors that they believed were responsible for: children not presenting to health services, refusing treatment or not attending follow up with the hospital or outreach service. Excursions into rural and isolated regions of the East Sepik Province provided the opportunity to see children with disabilities in the community and provide insight into the difficulties and prejudices experienced.

Results

Based on the PNG population census for the year 2000, there are 11,215 births a year within the population served by Wewak General Hospital [25]. Unfortunately, the exact numbers for the study period cannot be accurately known; however, public health organisations, including UNICEF, consider this to be the only method to obtain birth data [24]. Over the two-year study period, the incidence of CTEV presenting to hospital in the East Sepik Province of PNG is 2.7 per 1,000 live births per year. There was a predominance of males ($n=45$) with CTEV compared to females ($n=15$), giving a male:female ratio of 3:1. Males had a predominance of bilateral (62%), right (36%), followed by left (2%) clubfoot. Whereas females have a predominance of bilateral (53%), left (27%), followed by right (20%) clubfoot.

Age of presentation and treatment

The age of presentation varied greatly, with many patients presenting after 12 months of life (Fig. 1). Male children appeared to present earlier, with female children mainly presenting after 12 months. This had a major influence on the child's initial treatment regime, with surgery becoming more likely the older the child was at presentation (Fig. 2). No children who presented in the first 3 months of life required surgical correction of their clubfoot deformity, whereas all patients presenting after the second year of life

Fig. 1 Age at presentation of children with congenital talipes equinovarus (CTEV) ($n=60$)

had surgical intervention. There was, thus, an increase in the number of children requiring initial surgical treatment with increasing age. In total, there were 3,790 operations carried out at Wewak Hospital during the 24-month study period; CTEV operations accounted for 1.58% ($n=60$) of the total theatre time.

Table 1 shows the outcome of initial treatment in those diagnosed with CTEV deformity, but no long-term follow up is available due to the limited study period and the long period of treatment required. Unfortunately, a large proportion of children treated for CTEV were lost to follow up. As a result, it is difficult to comment on the efficacy of the varying treatments for clubfoot performed at Wewak General Hospital.

Problems identifying, treating and following up the children were assessed by a structured questionnaire. Thirty-eight individuals were questioned about their knowl-

Fig. 2 Initial treatment for CTEV against age at presentation ($n=60$)

Table 1 Beliefs and opinions regarding the presentation, treatment and follow up of children with CTEV

Treatment Outcome	Percentage
Acceptable functional outcome*	20%
Still undergoing treatment	16%
Did not attend review	38%
Absconded before first treatment**	19%
Health centre review†	8%

*As decided at doctor assessment 1 year following treatment conclusion. Defined as able to function independently without aids, where pain or discomfort does not limit activities

**Often, children absconded following diagnosis, but before treatment could be started; no explanations for their decisions were recorded

†These children had the option to attend a health centre for follow up by a physiotherapist (no doctors or surgeons were based at these centres). This study did not have access to records kept at these centres.

edge and attitudes towards CTEV. These included ten immunisation workers who had spent between 1 to 8 years working with local communities, 20 PNG parents whose questionnaire was translated by a local outreach doctor and eight outreach/missionary workers working stationed in rural tribal villages (Table 2). The most commonly reported problems preventing follow up and presentation to hospital were:

1. Time: isolated regions and poor infrastructure
2. Money: much of the rural population is self-sufficient and would struggle to pay for the cost of petrol needed to travel or the costs incurred for a hospital stay and treatment
3. Ingrained belief system: parents frequently felt ashamed to present children with deformities, as there is much stigma associated with birth defects

Seasonal variation

There was a predominance of CTEV and also bilateral deformity in children born between June and November (Table 3). CTEV births were most common in September, giving a conception time somewhere around December.

Discussion

This study showed a high incidence of CTEV 2.7 per 1,000 live births per year. This figure is considerably higher than that seen in the UK, where the incidence is quoted as between 0.89–1.24 per 1,000 live births per year [3, 26]. This is consistent with the only previous study of birth defects in PNG, which also indicated that there were a

Table 2 Beliefs and opinions regarding the presentation, treatment and follow up of children with CTEV. Each individual could provide 3 answers (total for each group equals 300%)

Group studied	Explanation given for difficulties with presentation, treatment and follow up of children with CTEV					
	Attitudes	Infrastructure	Availability	Climate	Cost	Cultural
Immunisation workers (<i>n</i> =10)	0% (0)	100% (10)	80% (8)	0% (0)	80% (8)	40% (4)
Parents of children* (<i>n</i> =20)	10% (2)	50% (10)	50% (10)	10% (2)	100% (20)	80% (16)
Outreach workers (<i>n</i> =8)	50% (4)	100% (8)	50% (4)	75% (6)	25% (2)	0% (0)

*Not necessarily parents of a child with a deformity

significant number of children born with musculo-skeletal defects, including CTEV [11]. The possibility remains that the figure of 2.7 per 1,000 live births shown in this study may underestimate the true incidence in the region, with not all congenital deformities in PNG being detected at birth. During visits to rural communities, children were seen with CTEV who had never sought treatment or presented to medical services. A great deal of anecdotal evidence suggested that this is not a rare occurrence and that there were many reports of parents keeping children with deformities hidden due to cultural beliefs. Many parents believed that a deformed child reflected a curse upon their family and would continue to cause bad luck.

The high prevalence of CTEV from this study, in line with other results from this geographical region [4, 6–8], may present further evidence to suggest that the aetiology of CTEV involves genetic factors, although this does not exclude possible environmental causes. Studies have suggested that there might be seasonal factors at work in the aetiology of CTEV [3, 19, 26]. Some authors have noticed an increase in babies with the condition born in the winter quarter (December to February) [3, 19]. With a winter peak, the 16th week of gestation would fall between July and October. The 16th week is the time where environmental influences have been suggested to have their maximum impact, this being the point that the talus attains its correct alignment [19]. PNG has an equatorial climate and is warm and wet for most of the year, suggesting that other factors must be at work to account for the increase in infants born with CTEV towards the end of the year (peak month September), as shown in this study. This suggests that conception would be around December and, thus, the 16th week of gestation in March. This coincides with the peak of the malaria season and, hence, maternal anaemia

[13]. As previously noted, maternal anaemia has been shown to be related to CTEV [6, 7, 21]. Low birth weight is another important factor related to CTEV incidence and frequently results from malaria, accounting for up to 40% of low birth weight babies in disease-prone areas [5]. However, many causes of low birth weight are attributable to a multitude of influences, including socio-economic factors, physical stresses and poor diet [2, 15, 16]. Pregnant women are rarely spared the physical stresses of daily subsistence farming, whilst the average diet is poor, consisting largely of starchy foods, such as sago and sweet potato.

In the developed world, treatment with the Ponseti method has proved to be successful, but requires a constant supply of good quality resources, input, motivation and commitment from the parents and healthcare team [10]. Frustration amongst health carers working with these children was significant. Often, they had the skills to apply the casts and mould the feet, but lacked the funds and resources, such as splints and shoes, required to complete treatment.

The fundamental aim of treatment in CTEV is to form a foot which can fit into a normal shoe and carry the weight of the body safely without discomfort. In PNG, closed shoes are expensive and infrequently worn, altering treatment aims, whilst the additional stresses of daily bare foot walking heighten the need for a good functional outcome. Many patients present late or not at all due to different cultural values, poverty, geographic isolation with no public transport, unavailability of health services and a lack of understanding of what can be achieved. Undoubtedly, these barriers adversely affect compliance, attendance and management, especially where regular follow up and treatments are required, such as using the Ponseti method

Table 3 CTEV and month of birth

Season	Total number of CTEV births	Bilateral CTEV	Right CTEV	Left CTEV
December to February	11	6	3	2
March to May	12	5	5	2
June to August	17	11	6	0
September to November	20	13	5	2

[1]. This is reflected by the fact that, of the cohort of patients studied, 38% did not attend review and 19% absconded before any form of treatment could be commenced. Non-compliance has been shown to be the main factor associated with the risk of recurrence and poor treatment outcome; the educational level of parents is also a significant risk factor [10]. The often late presentation of children with CTEV also influences the treatment options, with surgical techniques being increasingly likely the later they present. Interestingly, males presented to the medical services earlier than females, where no females were seen before 6 months of age. Although the numbers were too small to draw accurate conclusions, one explanation may be that families place greater importance on the health and fitness of male children due to the self-sufficient nature of family life in PNG.

This study found a high incidence of CTEV in this region, whilst treatment options and outcomes remain unsatisfactory. The lack of adequate treatment and recognition of CTEV as shown by this study is particularly concerning, as neglected clubfoot deformity causes major pain and disability. This will hinder a child's ability to live an independent life and support their family in what is largely a self-sufficient community. Recently, it has been shown that the Ponseti method of treatment achieves good results with physiotherapist-delivered systems [20] and orthopaedic clinical officer systems [23]. Furthermore, the Ponseti method seems to present a safe, effective and low-cost treatment for neglected idiopathic clubfoot presenting even after walking age [14].

The three major factors preventing follow up and delaying initial presentation were the time and distance taken to travel to a medical centre, lack of sufficient funds to pay for travel and the costs of medical care, and the local health beliefs. Although these problems are substantial, it identifies areas in which health services can work in order to improve the outcome for these children. One option would be to provide more extensive outreach services that could be provided by physiotherapists based in rural locations or even by training many of the community workers already based in isolated villages. The number of skilled physiotherapists in PNG who are able to perform this technique is increasing. Outreach projects including physiotherapists would be able to deliver a good outcome to many of these children with only a small number of visits to the hospital (tenotomy if required). This process could incorporate an education role in a hope to modernise health beliefs. This idea was met with great enthusiasm by teams currently operating in this region and presents a very realistic option that will greatly improve the quality of life for children with CTEV in this region. However, this does not solve the problem with the supply and acquisition of the vital materials necessary. It would be our aim to establish a

fund and direct charity organisations to the cause of the children suffering from this potentially treatable condition in PNG.

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