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Prevalence of rickets-like bone deformities in rural Gambian children

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ABSTRACT

The aim of this study was to estimate the burden of childhood rickets-like bone deformity in a rural region of West Africa where rickets has been reported in association with a low calcium intake. A population-based survey of children aged 0.5–17.9 years living in the province of West Kiang, The Gambia was conducted in 2007. 6221 children, 92% of those recorded in a recent census, were screened for physical signs of rickets by a trained survey team with clinical referral of suspected cases. Several objective measures were tested as potential screening tools. The prevalence of bone deformity in children <18.0 years was 3.3%. The prevalence was greater in males (M = 4.3%, F = 2.3%, $p < 0.001$) and in children <5.0 years (5.7%, M = 8.3%, F = 2.9%). Knock-knee was more common (58%) than bow-leg (31%) or windswept deformity (9%). Of the 196 examined clinically, 36 were confirmed to have a deformity outside normal variation (47% knock-knee, 53% bow-leg), resulting in more conservative prevalence estimates of bone deformity: 0.6% for children <18.0 years (M = 0.9%, F = 0.2%), 1.5% for children <5.0 years (M = 2.3%, F = 0.6%). Three of these children (9% of those with clinically-confirmed deformity, 0.05% of those screened) had active rickets on X-ray at the time of medical examination. This emphasises the difficulties in comparing prevalence estimates of rickets-like bone deformities from population surveys and clinic-based studies. Interpopliteal distance showed promise as an objective screening measure for bow-leg deformity. In conclusion, this population survey in a rural region of West Africa with a low calcium diet has demonstrated a significant burden of rickets-like bone deformity, whether based on physical signs under survey conditions or after clinical examination, especially in boys <5.0 years.

Introduction

Rickets is a childhood disorder of bone mineralisation at the growth plate, usually caused by inadequate concentrations of extra-cellular calcium or phosphate. The delay in or failure of endochondral ossification leads to deformation of the growth plate, the development of bone deformities and a reduction in linear growth [1,2]. Children with bone deformities may be severely disabled, have increased morbidity and decreased quality of life. The burden is currently greatest and the public health impact most substantial in developing countries, where crippling deformities reduce physical capacity and drain economic prospects [3]. Rickets is most commonly caused by vitamin D deficiency, although rickets in Sub-Saharan Africa, India and Bangladesh has been reported in children with a biochemical profile that does not suggest vitamin D deficiency but who may have calcium deficiency [2]. Prevalence estimates, however, are limited by the use of subjective assessments of deformity in large-scale surveys; there is a need to develop standard prediction tools for population screening [3–5].

We published a case series of rickets from The Gambia, West Africa, in which the aetiology was unknown but was associated with a very low calcium intake and elevated plasma fibroblast growth factor 23 (FGF23) [6]. The plasma 25-hydroxyvitamin D concentrations of these patients did not suggest vitamin D deficiency as a causal factor [6]. Rickets had not been formally described in The Gambia previously, a population typical of many Sub-Saharan African communities. The study reported here was designed to establish the prevalence and characteristics of rickets-like bone deformities in rural Gambian children aged 0.5–17.9 years in the province of West Kiang, which has been the focus of nutrition and health studies by the UK Medical Research Council for many years [6,7]. The survey was conducted using screening methods commonly employed in population surveys. In addition, five simple anthropometric measures (wrist width, wrist circumference, interpopliteal distance, intercondylar distance and intermalleolar distance) suitable for use by trained, non-medical staff were included to test their potential as

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screening tools. Suspected cases were referred for detailed clinical examination and diagnosis.

Subjects and methods

West Kiang is a rural region of The Gambia, West Africa, latitude 13°N. The people are of Mandinka, Jola and Fula ethnicity and follow the Islamic faith, although purdah is not commonly practised. Undernutrition is prevalent and foetal, infant and child growth retardation is common. Most families are subsistence farmers, with men and women working the fields during the single rainy season (July–November). The diet is deficient in several micronutrients and calcium intakes are very low, averaging around 200 mg/d in children and 300–400 mg/d in women. Tropical sunshine is abundant throughout the year, and customary dress, although relatively conservative for women, does not restrict sunshine exposure of the face, arms and hands, and, in younger children, the lower legs and upper body [6,7]. Vitamin D status in this population is good, with plasma concentrations of 25-hydroxyvitamin D in children and adults well above those associated with vitamin D deficiency rickets [6–8].

The Rickets Prevalence Survey was conducted in the 30 villages and hamlets of West Kiang between February and October 2007. Sampling was framed within the 3-monthly West Kiang Demographic Surveillance System. All 6767 children who were alive, resident in West Kiang and aged $\geq$0.5 years and $<$18.0 years on census day in February 2007 were eligible. The survey was approved by The Gambian Government/MRC Laboratories Joint Ethics Committee. Written informed consent was obtained from a parent or guardian of each child; all assessments were conducted with the implicit assent of the child.

The survey was explained to the villagers via a meeting with the village elders prior to the arrival of the survey team in each village. Each eligible child was traced, the family contacted and the survey explained by a member of the survey team.

Each participant was screened for bone deformities consistent with rickets by trained fieldstaff: knock-knee (genu valgum), bow-leg (genu varum) and windswept deformity of the lower limbs, ribcage deformities, bossing of the skull, and enlargement of the wrists, ankles and costochondral junctions. A brief questionnaire was completed to record if the child had difficulty in walking and running, bone pain at rest and during activity, and a greater tendency to fall than their peers. Prior to the survey, the survey team of six experienced community fieldworkers of the Medical Research Council were trained by a paediatric nurse (HLJ) to recognise rickets-like bone deformities and were provided with a photographic aide-memoire. They were also instructed in standardised anthropometry and the importance of regular equipment calibration.

A portable stadiometer (Leicester Height Measure, SECA, Hamburg, Germany) was used to measure the height of children $\geq$2.0 years. The child, with no shoes or head-dress, was positioned upright with a horizontal Frankfort plane. Feet were positioned together, parallel and flat on the floor. In children $<$2.0 years, length was measured using a length board (Kiddimeter 100, Raven Equipment, Essex, England). The child, lightly clothed, was laid supine on the board with a vertical Frankfort plane. The spine and legs were kept straight with toes pointing upwards.

Wrist width was measured at the ulnar styloid using a Vernier caliper (model 675037, Silverline, Yeovil, England) with the forearm positioned lightly away from the body, fingers outstretched and palm facing downwards. Wrist circumference was measured using a small paper insertion tape (TALC, St Albans, England) by locating the ulnar styloid and encircling the radial margin at the widest part of the wrist.

A short, solid ruler held parallel to the floor was used to measure the distance between the legs at various points. The child was asked to remove or lift up clothing that would obscure the anatomical landmarks, and to stand upright, facing forward, with ankles or knees together, and feet parallel and flat on the floor. If this posed difficulties, the feet were positioned as close together as possible while keeping them parallel and flat on the floor. Interpopliteal distance (IPD) was defined as the distance between the two tendons of the semi-membranous muscle on each leg at the level of the popliteal fossa when in the standing position. Intercondylar distance (ICD) was defined as the distance between the two medial tibial condyles with ankles together. Intermalleolar distance (IMD) was defined as the distance between the two medial malleoli with knees touching. The anatomical landmarks were located by palpation and marked prior to the measurement. All leg measurements were obtained with the child standing.

Suspected cases of rickets were referred to the MRC Keneba Clinic for medical examination by one of two clinicians and, when clinically appropriate, for radiography of the wrists and knees and blood sampling for the measurement of total alkaline phosphatase. All radiographs were scored using the Thacher Radiographic Scoring Method [9] and reviewed by a consultant paediatrician (JP) experienced in the diagnosis and scoring of rickets. Total alkaline phosphatase activity was measured in plasma anticoagulated with lithium heparin by Kone Analyser 20i, Finland using the Kone Lab Alkaline Phosphatase (IFCC) kit. For the purposes of the survey, active rickets was defined as bone deformity with a Thacher Score $>$1.5, an alkaline phosphatase $>$960 U/l, or both.

Relative risks, calculated using MedCalc Software v12.7.8 are presented as RR [95%CI]. To account for the wide age range being surveyed, height (or length if $<$2.0 years) was expressed as height-for-age SD-score (SDS) relative to the British Growth Reference [10]. Linear Model software was used for age and sex adjustment of height-SDS, IPD and wrist measures. Logistic regression and Receiver Operating Characteristic (ROC) curve analysis were used to assess the probability of a screening measure correctly identifying a clinically-confirmed lower limb deformity (DataDesk version 6.3, Data Description Inc; IBM SPSS version 22).

Results

The results are given in Fig. 1. Of the 6767 children aged 0.5–17.9 years living in West Kiang and eligible for the survey, 6221 (92%), 3260 boys (52%) and 2961 girls (48%), were enrolled and screened for signs of rickets-like bone deformities. The median [IQR] age was 8.2 [4.3–12.1] years and was not significantly different between boys and girls; 1777 (28%) were $<$5.0 years.

The survey team referred 206 children (3.3% of those screened) for clinical examination (Table 1). Of these, 58% were suspected of knock-knee, 31% bow-leg, 9% windswept deformity. Growth plate enlargement of the wrists, ankles or costochondral junctions was suspected in 2 other children (1% of those referred). No child was suspected of cranial bossing or rib-cage deformity. There were twice as many boys referred than girls referred (139 of 3260 (4.3%) vs 67 of 2961 (2.3%), RR = 1.9 [1.5–2.6] $p < 0.001$). A greater proportion of children aged $<$5.0 years were referred than older children: 101 of 1777 (5.7%) and 105 of 4444 (2.4%) respectively, RR = 2.5 [1.9–3.2], $p < 0.001$ (Table 2).

Of the 206 children invited to the clinic for medical review, 196 attended. Of these, 36 children (0.6% of those screened, 18% of those reviewed) had a deformity considered by the clinician to be outside the normal range and consistent with rickets; 47% knock-knee, 53% bow-leg (Table 1). None was considered to have growth plate enlargement. The majority of cases of clinically-confirmed deformity were boys (83%), and boys appeared to be more likely to have bow-leg than girls, although the difference was not statistically significant (18 of 30 (60%) compared to 1 of 6 (17%), RR = 3.6 [0.6–22.1], $p = 0.2$).

The median [IQR] age of those with clinically-confirmed deformity was 3.6 [2.1–7.4]y: 72% were $<$5.0 years old (Table 2). The prevalence of rickets-like bone deformity in the population of children $<$5.0 years was 1.5% compared with 0.2% in older children (RR = 9.1 [4.4–18.8], $p < 0.001$), and was greatest in boys $<$5 years (2.3%, Table 2). In the younger age group, knock-knee was more common than bow-leg (17 of the 26 cases (65%)) but bow-leg was more common in older children (8 of 10 cases (80%)).