CRANIOTABES IN THE AFRICAN CHILD

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It is generally accepted that up to the age of 3 months craniotabes can be found in otherwise normal children ;[1-5] and some authorities consider that craniotabes may be a normal finding over the age of 3 months.[1, 6] Bille[6] is of the opinion that every third child presents a non-rachitic craniotabes during the first year of life. After 3 months of age (or even before[7]) the presence of craniotabes may be associated with rickets, either as its earliest sign,[1, 3-5, 7-11] or as its only remaining sign.[1] It has also been found in association with prematurity,[7] congenital syphilis,[12] hydrocephalus and osteogenesis imperfecta[4, 5, 12] and with hypervitaminosis A.[12] Craniotabes has not previously been associated with kwashiorkor.

This paper records an unexpectedly high incidence of craniotabes observed by the author in African children seen in general practice. The incidence of rickets and kwashiorkor, and the correlation between craniotabes and these diseases, is discussed.

METHODS

This clinical survey was conducted in an entirely non-White (mainly African) practice in an urban area on the Witwatersrand, during the summer months December 1960 to February 1961. Only African children aged between 3 months and 2 years were the subjects of the survey.

There were no cases of congenital syphilis, osteogenesis imperfecta or hypervitaminosis A; and histories of prematurity were not obtained since prematurity is irrelevant as a cause of craniotabes after the age of 3 months.[5] A single hydrocephalic infant was excluded from the survey.

Craniotabes, as elicited by thumb palpation, was considered to be mild when the area of skull which could be depressed fairly easily did not exceed 1 inch in diameter; moderate when the area of skull which could be depressed with ease exceeded 1 inch in diameter but did not exceed 4 inches; and severe when an area of skull exceeding 4 inches could be depressed with extreme ease (having a 'parchment-like' effect).

Clinical rickets was recorded as being present when enlarged wrist or ankle joints and enlarged costochondral junctions were found in association with craniotabes. The same significance was not attached to other signs of rickets such as Harrison's grooves, cranial bossing, bow-legs or knock-knees, when these were present without enlarged wrist or ankle joints and enlarged costochondral junctions.

Kwashiorkor when present was graded in severity according to Dean's classification.[13]

RESULTS

In this survey craniotabes was usually found bilaterally, involving the same areas of skull on each side, just above, and behind and above, the ears over the parietal and temporal regions. When the craniotabes was severe the whole of the parietal bone, the temporal bone, some of the occipital bone (lateral aspects) and the posterior portion of the frontal bone were involved on both sides.

A. *Children aged 3 Months - 1 Year*

234 children with craniotabes were found. The incidence of craniotabes in this practice over a period of 1 month was 76·3% of all children seen in the age group 3 months to 1 year (106 out of 139 children examined).

Of the 234 children, 44 (18·8%) had severe craniotabes, 172 (73·1%) had moderate craniotabes, and in 18 cases (8·1%) the craniotabes was mild (Table I).

There was a high incidence of kwashiorkor—163 out of the 234 children (69·7%). Severe craniotabes occurred more commonly among the children with kwashiorkor (34 out of 163, or 20·9%) than among normally nourished children (10 out of 71 or 14·1%) (Table I).

Where rickets was found, it was in all cases mild to moderate in intensity. There was a total of 33 cases of rickets (14·1% of those children who had craniotabes). Rickets was found far more frequently in children with kwashiorkor and craniotabes (26 out of 163—15·9%) than in normally nourished children with craniotabes (7 out of 71—9·9%). The greatest incidence of rickets was in children with kwashiorkor and severe craniotabes (7 out of 34—20·6%). (Fig. 1 and Table I). The next highest frequency was in children with kwashiorkor and moderate craniotabes (17 out of 114—14·9%). Only 1 case of rickets was found in the group of normally nourished children with severe craniotabes (Table I).

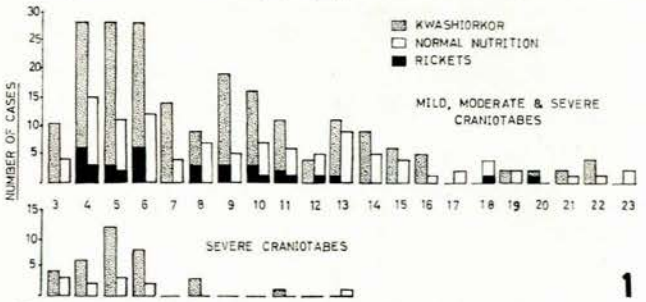

Fig. 1. The distribution by age of the nutritional status of craniotabes and rickets.

Three children in this age group had severe kwashiorkor. In one of these cases the craniotabes was severe and in the other 2 it was moderate. Only the child with severe craniotabes had rickets.

TABLE I. THE NUTRITIONAL STATUS, DEGREE OF CRANIOTABES AND DISTRIBUTION OF RICKETS IN THE TWO AGE GROUPS

Age group 3 months – 1 year

Nutritional status	*No. of cases*	*Craniotabes*			*Cases of rickets*
		Mild	*Mod.*	*Severe*	
Kwashiorkor	15				2
	163		114		17
				34	7
Normal nutrition		3			0
	71		58		6
				10	1
Totals	234	18	172	44	33

Age group 12 months – 23 months

Nutritional status	*No. of cases*	*Craniotabes*			*Cases of rickets*
		Mild	*Mod.*	*Severe*	
Kwashiorkor	10				1
	45		35		1
				0	0
Normal nutrition	11				1
	36		24		1
				1	0
Totals	81	21	59	1	4

B. *Children aged 1 - 2 Years*

81 children with craniotabes were found. The incidence in the practice over a period of 1 month was 58·3% of all children seen in the age group 1 - 2 years (67 out of 115 children examined).

Of the 81 children, 1 had severe craniotabes, and this child was normally nourished. 59 (72·8%) had moderate craniotabes, and in 21 cases (25·9%) the craniotabes was mild (Table I).

There were again more children with kwashiorkor and craniotabes than normally nourished children with craniotabes —45 out of 81 (55·6%) (Table I). Only 4 cases of rickets were found in this age group.

Five children in this age group had severe kwashiorkor. In all these cases craniotabes was moderate, and no rickets was found.

Comparing the 2 age groups (Fig. 1):

1. Craniotabes was found more commonly under 1 year than over 1 year of age.
2. Severe craniotabes was present almost entirely in children under 1 year of age, and most commonly in the age group 3 - 6 months.
3. Rickets was far more frequent in children under 1 year of age than in those over 1 year of age (14·1% and 4·9% respectively).
4. There were more children who had severe kwashiorkor accompanied by craniotabes over 1 year than under 1 year of age, but rickets (1 case) was found in association with severe kwashiorkor only in the younger age group.

DISCUSSION

Craniotabes

Unlike Eliot and Park,[5] who describe a typical rachitic distribution of craniotabes and the fact that craniotabes rarely occurs symmetrically, I found it impossible to distinguish rachitic from non-rachitic craniotabes on the basis of its anatomic distribution; in addition, craniotabes was almost invariably found to be symmetrical.

The high incidence of craniotabes in the present series is difficult to explain. One reason might be the method of thumb palpation employed. There was no means of standardizing the intensity of thumb pressure on the skull to elicit craniotabes; thus I may have included a greater number of cases as exhibiting craniotabes than would have been the case had lighter pressure been used.

A considerable variation in the incidence of craniotabes is reported in the literature. Early authorities such as Moore and Dennis[14] quote an incidence of 62·2% (White children under 1 year); Barenberg and Bloomberg[7] an incidence of 44·6% in the first year of life; and De Buys[15] an incidence of 43·48% in Coloured and an incidence of 33·3% in White (rachitic) infants under 1 year old. Bille[6] quotes a report by Japanese workers who found an incidence of 8·1% in the same age range. More recent reports are those from Bille,[6] who found an incidence of 36·5% during the first year of life, and Jeliffe[16] whose figure is 5% of children between the ages of 3 months and 1 year.

The over-all incidence of craniotabes in children up to 1 year of age in the present series is greater (76·3%) than the figures cited above, and is also greater than the incidences reported in Johannesburg by Griffiths[17] (10·11%) and in Cape Town by Dancaster and Jackson[1] (50%). The fact that the latter authors did not consider craniotabes to be present when it involved an area about 1 inch in diameter (whereas in this series it was classified as 'mild' craniotabes) is probably one of the reasons for the discrepancy. Dancaster and Jackson's series is not completely comparable, however, since only 10 out of the 103 children whom they investigated were 'pure' Africans, the remainder being of 'mixed' origin. It is well known that the dark races are highly susceptible to rickets[4, 5, 18, 19] but there is no indication in the literature that the same applies to craniotabes alone. It may however be relevant that Bille[6] quotes Reiss and Boder (1948) and Boder (1949 and 1952) in studies on newborn infants as finding a predilection in negro infants for congenital craniotabes.

Although the incidence of craniotabes in this series was lower (58·3%) in children between 1 year and 2 years of age than in those under 1 year (76·3%) it is nevertheless very much higher than that reported in previous investigations. Eliot and Park[5] state that craniotabes usually disappears by the 8th month of life but that it may be found occasionally as late as 18 months; Feldman[9] finds that it is rarely present in infants with active rickets over the age of 9 months, and only 1 of his 20 cases of florid rickets over 1 year of age had softening along the lambdiodal suture lines (this has the same significance as craniotabes[4, 5]). In Barenberg and Bloomberg's series,[7] of 26 children who had craniotabes in the age group of 4 months - 1 year, only 1 was 1 year old. In Corner's survey,[20] of 274 children aged 1 - 2 years, craniotabes was found in only 2 cases—an incidence of 1·7%; in both these cases there were also other signs of rickets. The only other series is that of Ito Sukeo,[21] who found 1 case of craniotabes in 623 non-rachitic children aged 1 - 2 years, and 3 cases in 129 rachitic children in the same age range.

The incidence of severe craniotabes in this series in children aged 3 months to 1 year was 18·8%. When this age-group is sub-divided into the 2 intervals 2 - 6 months and 7 - 12 months, as Dancaster and Jackson[1] did in their survey, the incidence in the first age group is 31·7%, which is similar to that found by Dancaster and Jackson (33·3%).* In the second group it is 4·3%, considerably less than their figure of 14·8%.* Barenberg and Bloomberg[7] had 2 groups of patients in their series; 1 group under 4 months of age, where the incidence of marked craniotabes was 27·9%, and another group aged 4 - 12 months where the incidence was 42·8%. The oldest child with severe craniotabes was 7 months. From these series it appears that severe craniotabes occurs more consistently in those children aged less than 6 months.

Rickets and Kwashiorkor

In the present series the highest incidence of rickets occurred in the age group which had most cases of severe craniotabes, namely fourth to sixth month (Fig. 1). However, the incidence of rickets was only slightly higher (18·2%) in the cases of severe craniotabes than in the cases of moderate craniotabes (13·3%). Dancaster and Jackson[1] similarly found that children with severe craniotabes were no more liable to rickets than those with lesser degrees of craniotabes.

Using the clinical signs of rickets as the only indication of its presence will obviously not give a complete picture of its incidence.[1, 5] This (in addition to the fact that rickets in children without craniotabes was excluded from the survey) explains the low incidence of 14·1% (33 cases out of 234) in children under 1 year of age, and 4·9% (4 out

*Derived from their Fig. 1.

of 81) in children aged 1 - 2 years, as compared with the incidence of 50% in Dancaster and Jackson's series,[1] in which radiological means were used to ascertain the presence or absence of rickets; although Griffiths[16] found an even lower incidence in the first year of life (2·04%). Other surveys comparable to the present one are those of Barenberg and Bloomberg,[7] who found an incidence of 23·9%, and Moore and Dennis,[14] where an incidence of 23% of children with craniotabes was found. Bille[6] found no rickets in the 36·5% of his patients who had craniotabes.

The age distribution of rickets in this series was fairly typical,[5, 18] except that some authorities[4, 20, 22] state that, whereas mild rickets occurs in the first few months, marked rickets occurs towards the end of the first and during the second year. The fact that the incidence of rickets fell precipitously in this series after the age of 7 months (unlike the findings in the above series) without the incidence of craniotabes diminishing commensurately, would tend to weaken a correlation between craniotabes and rickets over the age of 7 months. Even if cases of rickets without craniotabes were included, no significant increase would have occurred either under or over 1 year of age (0·7% and 3·4% respectively).

Williams[23] found that nearly all cases admitted to her wards were due to or complicated by some form of malnutrition, yet she found no craniotabes. In the present series, more of those children with craniotabes had kwashiorkor than were normally nourished. Rickets was found commonly in those children with kwashiorkor, and more so in those with severe craniotabes. However, it would seem that the severe craniotabes was related to the kwashiorkor rather than to rickets, because not all the cases of severe craniotabes had clinical rickets and because the incidence of rickets in normally nourished children with severe craniotabes was much lower (see Table I).

If rickets were the cause of all forms of craniotabes in this series, it would raise the incidence of rickets to 70%, which is very much higher than the 50% incidence suspected by Dancaster and Jackson.[1] On the other hand, it would explain why craniotabes occurs so commonly in children with kwashiorkor, since we already know that in this series rickets occurred most commonly in children with kwashiorkor. This is a view accepted by a number of authorities,[2, 5, 9, 19, 21, 23-25] who have shown that rickets can occur frequently in children with kwashiorkor. The earlier view that rickets occurs only under conditions of rapid and normal growth[4, 7, 22, 26, 27] would seem to be no longer valid.

A further factor showing a correlation between craniotabes and kwashiorkor lies in the accepted view[9, 28] that osteoporosis occurs in kwashiorkor. Feldman[9] considers that, as a result of there being insufficient protein present in the skeleton, onto which calcium and phosphate can be precipitated, osteoporosis occurs. Jones and Dean[28] found that in protein malnutrition the bones were smaller, less well calcified and had diminished trabecular pattern.

The histological concept of craniotabes being osteoporotic in nature is certainly not new. According to Barenberg and Bloomberg[7] both Pomner (1885) and Wieland (1910) showed that craniotabes in the newborn was osteoporotic; and Moore and Dennis[14] quote Hess as expressing the view that craniotabes before the fifth or sixth month of life is caused by congenital osteoporosis.

If it is accepted that osteoporosis in general is a feature of malnutrition, it is suggested that where there is no other explanation for the presence of craniotabes, its presence may be a manifestation in the cranium of osteoporosis of malnutrition.

SUMMARY

In a survey of African children in general practice the incidence of craniotabes was found to be 76·3% in those children between 3 months and 1 year old, and 58·3% in those between 1 and 2 years of age.

In both age groups there were more children with kwashiorkor and craniotabes than normally nourished children with craniotabes. Severe craniotabes occurred almost exclusively in children under 1 year of age, and most commonly in those children with kwashiorkor. Only 1 case of severe craniotabes occurred in a patient under 1 year of age with severe kwashiorkor, the majority of cases of severe kwashiorkor being over 1 year of age.

The incidence of rickets among children with craniotabes in the age group 3 months to 1 year was 14·1%, and 4·9% in the age group 1 - 2 years. Rickets was found more commonly in those children with kwashiorkor. Despite the fact that rickets occurred most commonly in the group of children with kwashiorkor and severe craniotabes, it is suggested that the severe craniotabes is correlated with kwashiorkor rather than with rickets.

It is suggested that craniotabes is a manifestation in the cranium of osteoporosis of kwashiorkor, and may account for the high incidence in this series.

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REFERENCES

1. Dancaster, C. P. and Jackson, W. P. U. (1960): S. Afr. Med. J., **34**, 776.
2. *Idem* (1961): *Ibid.*, **35**, 890.
3. Ellis, R. W. B. and Ellis, A. E. (1944): Arch. Dis. Childh., **19**, 47.
4. Warkany, J. *in* Nelson, W. E., ed. (1954): *Textbook of Pediatrics*, 6th ed., p. 325. Philadelphia: Saunders.
5. Eliot, M. M. and Park, E. A. *in* McQuarrie, ed. (1945): *Brenneman's Practice of Pediatrics*, vol. 1, chapt. 36. Hagerstown: W. F. Prior.
6. Bille, B. S. V. (1955): Acta paediat. (Uppsala), **44**, 185.
7. Barenberg, L. N. and Bloomberg, M. W. (1924): Amer. J. Dis. Child., **28**, 716.
8. Dancaster, C. P. (1959): S. Afr. Med. J., **33**, 503.
9. Feldman, N. (1952): 'A clinical study of florid rickets in non-European infants in Johannesburg', M.D. thesis, University of the Witwatersrand.
10. Moore, C. U. (1924): J. Amer. Med. Assoc., **83**, 1469.
11. Zarfl, M., Dalyell, E. J. and Mackay, H. M. M. (1919): *Studies of Rickets in Vienna*, part 3, no. 2, p. 115. Spec. Rep. Ser. Med. Res. Coun. (Lond.), 77.
12. Arena, J. M., Sarazen, P. and Baylin, G. J. (1951): Pediatrics, **8**, 788.
13. Dean, R. F. A. (1960): J. Pediat., **56**, 675.
14. Moore, C. U. and Dennis, H. G. (1925): Amer. J. Dis. Child., **30**, 683.
15. De Buys, L. R. (1924): *Ibid.*, **27**, 149.
16. Jeliffe, D. B. (1951): Trans. Roy. Soc. Trop. Med. Hyg., **45**, 119.
17. Griffiths, J. (1962): 'The urban Bantu infant: growth, mortality and morbidity', M.D. thesis, University of the Witwatersrand.
18. Follis, R. H., Park, E. A. and Jackson, D. (1952): Bull. Johns Hopk. Hosp., **91**, 480.
19. Gillman, J. and Gillman, T. (1951): *Perspectives in Human Malnutrition*, p. 393. New York: Grune & Stratton.
20. Corner, B. D. (1944): Arch. Dis. Childh., **9**, 68.
21. Sukeo, I. (1956): Tohoku J. Exp. Med., **64**, suppl. 4, 25.
22. Findlay, L. and Ferguson, M. (1918): *A Study of Social and Economic Factors in the Causation of Rickets* Spec. Rep. Ser. Med. Res. Coun. (Lond.), 20.
23. Williams, C. D. (1946): Arch. Dis. Childh., **21**, 37.
24. Stransky, E. and Dizon-Santos-Ocampo, P. O. (1958): J. Trop. Pediat., **4**, 17.
25. Snyman, J. D. and Murray, A. B. (1961): S. Afr. Med. J., **35**, 595.
26. Smith, D. R. (1958): N.Z. Med. J., **57**, 594.
27. Griffel, B. and Winter, S. T. (1958): J. Trop. Pediat., **4**, 13.
28. Jones, P. R. M. and Dean, R. F. A. (1959): J. Pediat., **54**, 176.