

## **The incidence of cervico-thoracic spina bifida occulta in South African negroes**

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### **INTRODUCTION**

During the last twenty years interest has been shown in the geographical and racial incidence of congenital malformations, and reports of a number of epidemiological investigations have appeared (Stevenson, Johnston, Stewart & Golding, 1966). A recent report of a World Health Organization Scientific Group (1970), summarizing the evidence regarding the frequency of spina bifida, concludes that 'there is little doubt that there are real variations in the frequencies of spina bifida in different communities'. The published material dealing with the incidence of spina bifida occulta is almost exclusively concerned with the occurrence of this anomaly in the lumbo-sacral region of the spine, and there is relatively little information on its frequency in the cervico-thoracic region.

The radiologist is favourably placed to detect the presence of an asymptomatic skeletal malformation in the cervico-thoracic region because this area is invariably visualized in routine postero-anterior chest radiographs. Every radiologist has had occasion to note, as an incidental finding when examining a chest film, the presence of congenital anomalies such as cervical ribs or spina bifida defects in the lower cervical or upper thoracic spine.

Because we had formed a clinical impression that the latter anomaly was relatively frequent in chest radiographs of African patients, we decided to assess its incidence statistically. This paper records and discusses the results.

### **MATERIAL AND METHOD**

In the course of the routine work of an X-ray department serving a general teaching hospital handling African patients, each author independently recorded every consecutive chest examination seen, care being taken not to include in the count patients seen by both examiners or the same patient examined on different occasions.

The chest films of 5363 patients were analysed. Approximately half of these were males and half were females. On all the radiographs the spinous processes of at least C5 to T4 were visualized.

All the patients were Bantu-speaking negroes and were mostly members of either the Nguni or Sotho ethnic groups. Their ages ranged from 9 years to 68 years, but the majority were in the third, fourth and fifth decades of life. When a spina bifida

Table 1. *Incidence of cervico-thoracic spina bifida defects in 5363 patients*

Number of vertebrae affected	Number of patients	Percentage of total patients	Vertebral level	Males	Females	Total patients	Percentage incidence at each level
One	81	60.5	C6	0	2	2	2.5
			C7	15	12	27	33.3
			T1	30	17	47	58.0
			T2	2	2	4	4.9
			T3	1	0	1	1.2
Two	33	24.6	C6, C7	3	2	5	15
			C7, T2	6	12	18	54
			T1, T2	4	5	9	27
			C7, T3	1	0	1	3
Three	17	12.6	C6, C7, T1	3	4	7	41
			C7, T1, T2	5	3	8	47
			T1, T2, T3	2	0	2	11
Four	2	1.5	C6, C7, T1, T2	0	1	1	—
			C7, T1, T2, T3	1	0	1	—
Five	1	0.8	C6, C7, T1, T2, T3	0	1	1	—
Totals	134	100	—	73	61	134	—
			Percentage	54.5%	45.5%	100%	

anomaly was noted in the cervico-thoracic region, the name, age and sex of the patient were recorded, with the number and levels of the vertebral segments involved.

#### RESULTS

In every instance the finding of a spina bifida occulta in the chest radiograph was incidental and unrelated to the patient's clinical symptoms or the reasons for which the chest examination was requested. None of the patients had symptoms relevant to the anomaly and indeed none was aware of its presence as far as we could tell.

In the 5363 radiographs, one or more spina bifida defects in either lower cervical (C6, C7) or upper thoracic (T1, T2, T3) vertebrae were observed 134 times, an incidence of 2.49%.

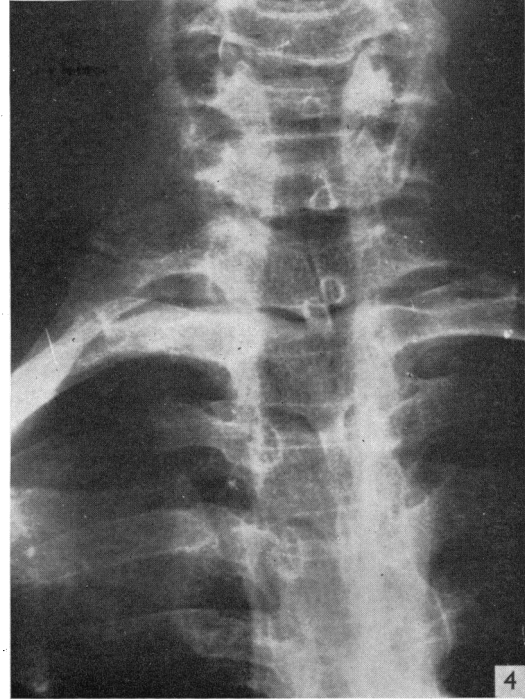
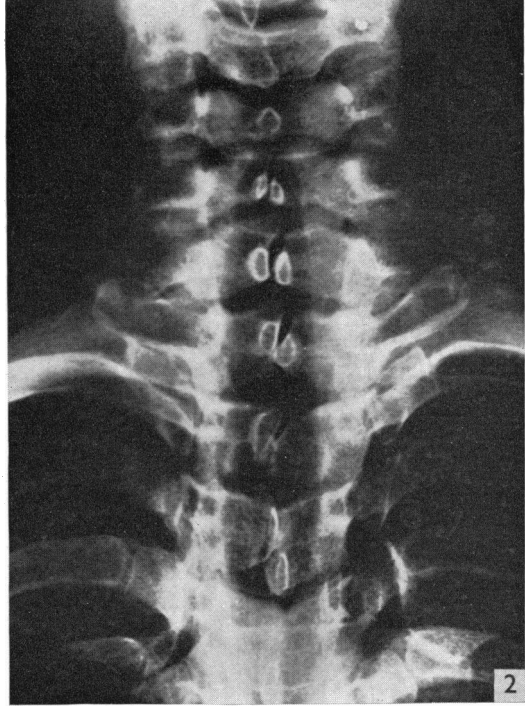
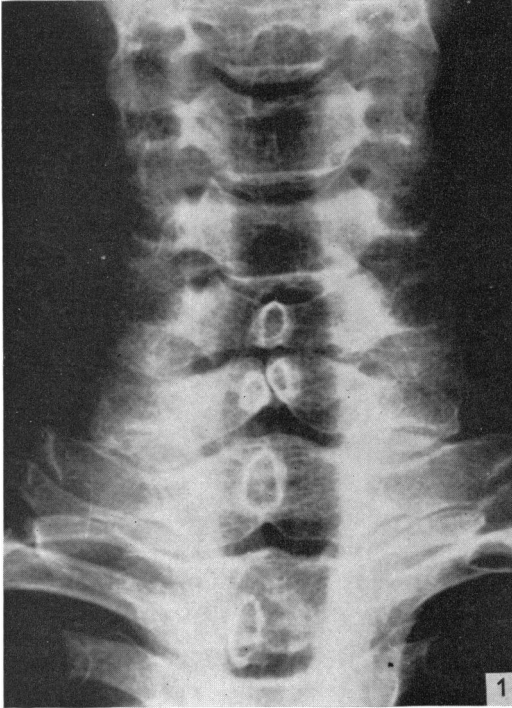
There was no significant difference in sex incidence, 54.5% of the 134 patients

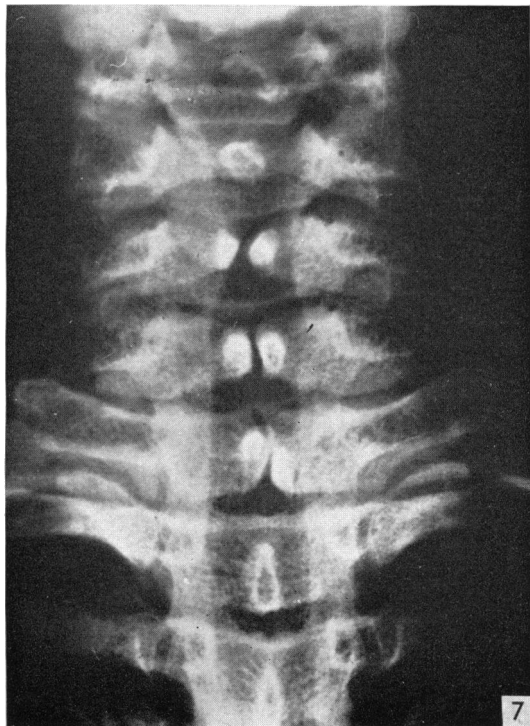
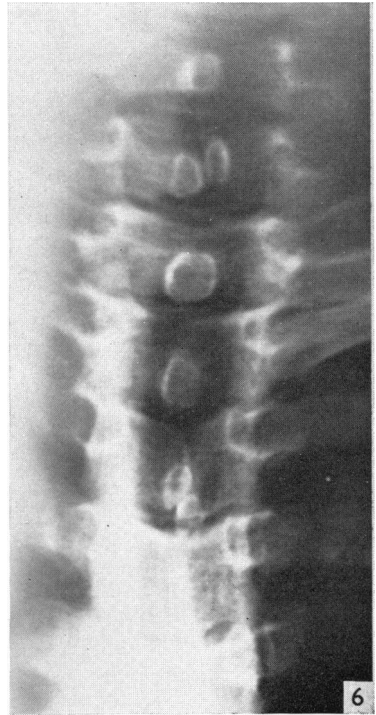
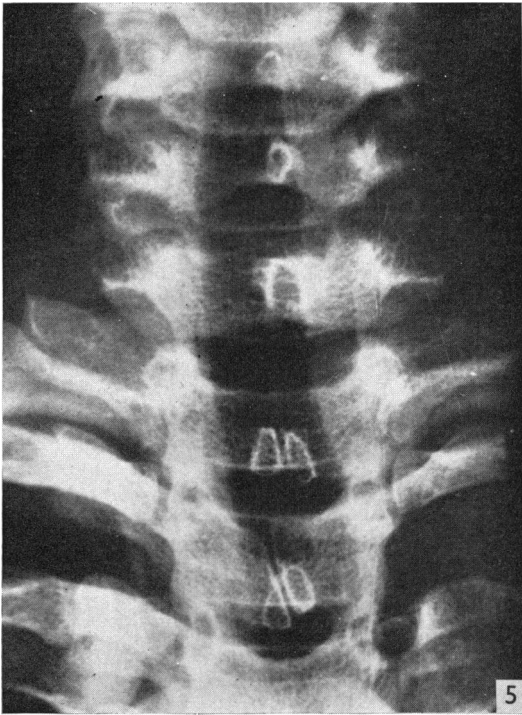
Fig. 1. Close-up view of a characteristic appearance of spina bifida occulta of the seventh cervical vertebra as seen in a routine chest radiograph. Note the oblique translucent cleft separating the two densely corticated halves of the non-fused spinous process. The axial appearance of normal spinous processes is well shown at the levels above and below the anomaly.

Fig. 2. Five vertebrae adjacent to the cervico-thoracic spinal junction exhibit the typical defects of spina bifida occulta. The affected vertebrae are C6, C7, T1, T2 and T3. This is the most extensive degree of malformation found in this series.

Fig. 3. Spina bifida occulta of the first thoracic vertebra. This was the commonest site of single vertebral involvement.

Fig. 4. The association of spina bifida with other developmental anomalies of transitional type in the cervico-thoracic region was not uncommon. Here a spina bifida defect of C7 is associated with a large bony cervical rib on the right and a rudimentary one on the left.





being males and 45.5% females out of approximately equal total numbers of patients of each sex.

In Table 1 the incidence of the anomaly at single and multiple spinal levels is given. Of the 134 patients with spina bifida occulta at the cervico-thoracic transition 81 (60.5%) had only one vertebra affected (Fig. 1). Thirty-three patients showed the defect in two vertebrae, 17 had three vertebrae involved and 2 had four. One patient had five contiguous vertebrae affected (Fig. 2).

In the 81 patients in whom one vertebra only was affected, the commonest site was the first thoracic vertebra (58.0%; Fig. 3), and the next most frequent was the seventh cervical (33.3%; Fig. 1). Other developmental anomalies of transitional type, such as bony cervical ribs and rudimentary first thoracic ribs, were sometimes found at the cervico-thoracic junction in association with spina bifida defects (Fig. 4).

When two vertebrae were affected the most common combination – C7 and T1 – occurred in more than half (54%). Fig. 5 illustrates spina bifida defects in the first and second thoracic vertebrae. Whenever more than one vertebra was affected it was found that the involved vertebrae were adjacent to each other (Table 1), with a single remarkable exception. In this instance the patient, an adult male, showed typical defects in the seventh cervical and third thoracic vertebrae. The intervening first and second thoracic vertebrae had intact posterior neural arches (Fig. 6).

Of the 17 patients who had defects in three vertebrae (Fig. 7) the commonest involvement (47%) was C7, T1 and T2. Only three patients were discovered with four or more defective vertebrae. Two of these each had defects in four vertebrae, the levels involved being C6, C7, T1, T2 and C7, T1, T2, T3 respectively, and the third, who had the most extensive involvement seen in this series, showed spina bifida of the five vertebrae C6, C7, T1, T2, T3 (Fig. 2). To our knowledge spina bifida defects involving four and five vertebrae in the cervico-thoracic region have not previously been described.

## DISCUSSION

### *Definition and sites of spina bifida occulta*

Spina bifida occulta is a midline defect in the posterior part of the vertebral arch due to a failure of fusion of its two halves, without any externally visible malformation of the spinal cord, its nerves or meninges. The anomaly is most commonly found at the areas of transition of one vertebral region into another, namely the occipito-cervical, cervico-thoracic, thoraco-lumbar and lumbo-sacral areas. Köhler (1956), Schmorl & Junghanns (1959), Bucy (1960) and Epstein (1969) are agreed that the lumbo-sacral region is the most frequent site of the malformation.

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Fig. 5. Spina bifida clefts in T1 and T2. These two vertebrae were involved in 27% of patients who had the anomaly at two spinal levels in the cervico-thoracic region.

Fig. 6. The only instance where two anomalous vertebrae were not adjacent to each other. Spina bifida defects are present in C7 and T3, and the intervening vertebrae, T1 and T2, are normal.

Fig. 7. An example of spina bifida occulta involving three vertebrae in the cervico-thoracic region, in this instance C6, C7 and T1. Note the variation in the degree of separation and direction of obliquity of the non-fused elements of the vertebral arch.

Sutherland (1922), at the Mayo Clinic, found 621 cases of spina bifida in approximately 12000 radiographs of the spine, and nearly 95 % of these were at the lumbosacral junction. S1 and S2 were involved in 70 % of cases and L5 in 24.5 %. Wheeler (1920) found the order of frequency in individual vertebrae to be: S1, L5, C1.

The thoraco-lumbar junction is regarded as the next site in order of frequency after the occipito-cervical region, and the cervico-thoracic junction is the least frequent site of all. Schmorl & Junghanns state 'cleavage of the spinous processes is seen only rarely in the lower cervical area.'

In summary, spina bifida occulta occurs at transitional sites of the axial skeleton in the following order of frequency: lumbosacral, occipito-cervical, thoraco-lumbar and cervico-thoracic.

#### *Incidence of cervico-thoracic spina bifida in whites*

Cockshott (1958) stated that before going to Nigeria from Scotland he rarely saw a spina bifida occulta defect at the level of the thoracic inlet. Kerley (1950) has given the only figure of incidence for a British population group that we have been able to find. He estimated the frequency of incomplete fusion of the arch of the seventh cervical vertebra and occasionally of the first thoracic vertebra at one in 600 in over 10000 chest radiographs. This gives an incidence of 0.166 %. The difference between this figure and the incidence of 2.49 % in our series, tested by  $\chi^2$ , is highly significant ( $P < 0.001$ ).

Walker & Bucy (1934) found in Chicago whites an incidence of spina bifida occulta in the sixth and seventh cervical vertebrae of 0.02 % and 0.04 % respectively, and an incidence of 0.09 %, 0.10 % and 0.05 % for the first, second and third thoracic vertebrae respectively. No data have been found comparing the incidence of the anomaly in North American whites and negroes. Lanier (1939) studied the presacral vertebrae of 100 American white and 100 American negro males. In no instance was a spina bifida defect found in either group in the cervico-thoracic region.

#### *Incidence of cervico-thoracic spina bifida in African and other populations*

Our figure of 2.49 % is similar to data for other African groups.

Whittaker (1957) examined 1226 postero-anterior radiographs of chests of African patients in the King George VI Hospital, Nairobi, and found 16 cases of spina bifida occulta, giving an incidence of 1.3 %. C7 was involved in ten instances, T1 in four and C7 plus T1 together in two. Cockshott (1958) examined 1000 consecutive adult chest films at the University College Hospital, Ibadan, and found an incidence of spina bifida of 2.2 %. The levels affected were: C7 in 9 patients, T1 in 17 patients and T2 in 6 patients. In eight instances two adjacent vertebral arches showed the defect, and in one instance three adjacent vertebrae were affected. The majority of the patients were Yorubas and two-thirds were males.

Some information is available from the study of African skeletal material but the number of specimens in each series is relatively small. Shore (1930) examined 82 vertebral columns from the skeletons of South African Negroes in the Anthropological Museum of the Anatomy Department of the University of the Witwatersrand.

Table 2. Summary of comparative data on incidence of cervico-thoracic spina bifida in various populations

Author	Population studied	Material	Number studied	Vertebral levels affected	Incidence of spina bifida occulta (%)
Shore (1930)	South African negroes	Skeletons	82	C7, T1, T2, T3	3.6
Stewart (1932)	Alaskan Eskimos	Skeletons	217	C3	0.46
Allbrook (1955)	East African negroes	Skeletons	206	C7, T1, T3	2.9
Walker & Bucy (1934)	North American whites	Radiographs	4088	C6	0.02
			5381	C7	0.04
			5406	T1	0.09
			4989	T2	0.10
			4234	T3	0.05
Kerley (1950)	British whites	Radiographs	10000	C7, T1	0.16
Whittaker (1957)	East African negroes	Radiographs	1226	C7, T1	1.3
Cockshott (1958)	West African negroes	Radiographs	1000	C7, T1, T2	2.2
Levy & Freed (1972)	South African negroes	Radiographs	5363	C6, C7, T1, T2, T3	2.49

He found spina bifida defects in the cervico-thoracic region in three out of the 82 columns, an incidence of 3.6%. The affected vertebrae were C7 in one subject, T1 in one subject and T1, T2 and T3 together in the third. Allbrook (1955) studied 206 vertebral columns of East African ethnic origin. Spina bifida occulta was present in the cervico-thoracic area in 2.9% of cases, distributed as follows: C7, 2 cases; T1, 3 cases; T3, 1 case.

Stewart (1932) found one example of spina bifida in the cervical region (C3) of 217 vertebral columns of Alaskan Eskimos (0.46%). There was no instance of the anomaly occurring in the upper thoracic region.

In Table 2 the comparative data available on the incidence of spina bifida occulta in the cervico-thoracic region of the spine in various population groups is summarized, including the results of the present investigation.

Three conclusions emerge from this study:

1. The finding of an incidence of spina bifida occulta in the cervico-thoracic region of 2.49% in South African negroes is in close agreement with the figures of Cockshott and Whittaker for West and East Africa respectively and suggests that there is a relatively high incidence of this malformation generally among the indigenous peoples of the African continent.

2. The study confirms the validity of our clinical impression of a higher incidence in South African Bantu-speaking negroes as compared with available data giving the incidence in whites.

3. Finally our investigation suggests that not only do South African negroes have one of the highest incidences of this anomaly that has so far been demonstrated, but among them multiple levels of involvement in the cervico-thoracic region are more numerous and extensive than in other populations.

## SUMMARY

The incidence of spina bifida defects in the cervico-thoracic region of the vertebral column was assessed in the chest radiographs of 5363 male and female South African negroes aged 9 to 68 years.

The anomaly was found in 2.49% compared with an incidence of from 0.02 to 0.16% in Caucasian populations reported in the literature. The available evidence suggests that the negroes of South, East and West Africa have a higher incidence of this anomaly than the whites of Europe and North America.

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## ADDENDUM

Since this investigation was closed a second case of involvement of five vertebrae has been seen. The affected levels were C7, T1, T2, T3, T4.

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