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CORONAL CLEFT OF VERTEBRAE, A VARIANT OF NORMAL ENCHONDRAL OSSIFICATION

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In 157 spontaneously aborted human embryos and fetuses the incidence and evolution of coronal cleft formation was investigated radiologically and histologically. Complete clefts were formed in nine cases and incomplete clefts in 16 cases. There was a predominance of males. It is concluded that coronal clefts are variations of normal enchondral ossification and that the notochord plays no role in its pathogenesis.

Coronal clefts seen on radiographs of newborn should not be interpreted as a malformation. Thus no preventive or treatment regimen is indicated.

Key words: intervertebral disc, notochord; ossification, physiologic; spine abnormalities; spine, embryology

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A vertical radiolucent band in the vertebral body seen in a lateral projection, the so-called coronal cleft, is recognized fairly frequently in the perinatal period. Although it is now generally accepted that this condition represents a normal variant of the process of ossification without any clinical significance, its onset and development warrant further elucidation.

Most authors (Cohen et al. 1956, Wollin & Elliott 1961, Rowley 1955, Schmorl & Junghanns 1975, Ehrenhaft 1943) think that the cleft is occupied by the notochordal tissue, and that consequently its persistence disturbs the normal development of the vertebral body. In most cases, however, the diagnosis was first made radiologically, and histologic sections were done subsequently without proper comparison with the normal development of the human spine.

In the course of radiologic and histologic investigations of the prenatal development of the vertebral column, we were able to identify various patterns of ossification of the vertebral bodies. On radiographs we identified coronal clefts, incomplete clefts such as two ossification centres connected with a transverse osseous bar, and cranial or caudal halves of clefts. On histologic sections, besides finding the different clefts we observed in many cases another pattern of ossification; an isolated area of cartilaginous tissue inside the ossified nucleus which could by closer examination be suspected radiologically as an ill-defined spot of rarefaction. We were never able to see any evidence of persistence of the notochord nor of a lateral expansion of this tube-like structure. The absence of notochordal abnormalities suggests that the various patterns of ossification must results from other factors such as blood vessel distribution inside the vertebral body.

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MATERIALS AND METHODS

From our collection of 266 spontaneously aborted human embryos and fetuses, 157 fetuses showing radiological evidence of ossification or calcification of the vertebral bodies were selected for this study.

Their crown-rump length ranged from 45 mm to 490 mm and their estimated age of gestation varied from 9½ weeks to full-term.

The specimens were fixed in 10% formalin. The vertebral columns were dissected, radiographed, decalcified with 12% EDTA, and then cut either in a cross, frontal or sagittal plane. The sections were stained with Hematoxyline Phloxine Saffron, Toluidine Blue, Mann-Dominici or Goldner's methods.

RESULTS

Radiological investigation

Coronal clefts were found in nine cases (5.7%, seven males and two females). In a lateral projection the clefts were located in the midline or slightly dorsal to it (Figure 1). Incomplete clefts such as two ossification centres connected by a transverse osseous bar, a cranial or caudal half of such a cleft, or a very narrow radiolucent line were detected in 16 cases (10.2%, 12 males and four females) (Figure 2). The location of the incomplete clefts was the same as that of the complete clefts. A majority of these clefts were seen in the thoracic and lumbar vertebrae. Cartilaginous islands observed inside an ossified nucleus of the body on histologic sections were rather difficult to identify radiologically. Only retrospectively could these areas be recognized as sites of decreased density.

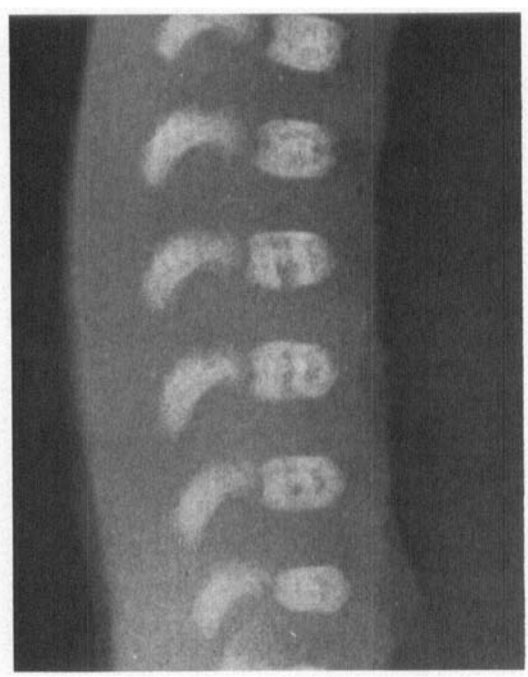


Figure 1. Coronal cleft seen in the lateral projection of a 20-week-old fetus.

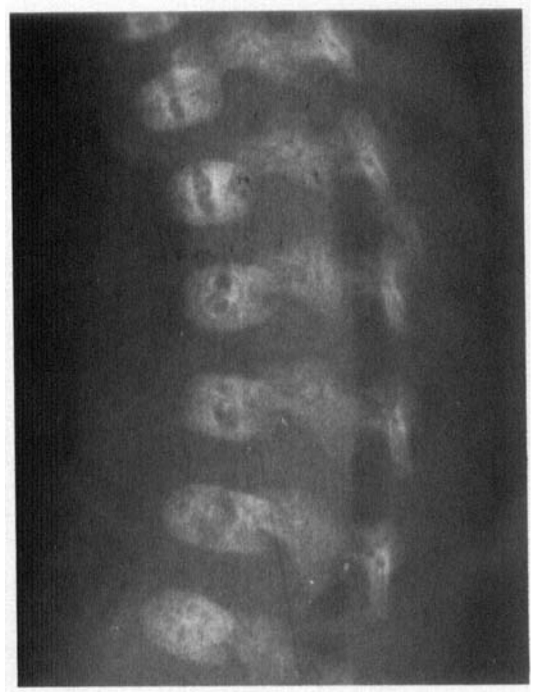


Figure 2. Incomplete clefts seen in the lateral projection of a 21-week-old fetus. Note the transverse osseous bar seen in most of the vertebrae.

Histological investigation

Before describing the coronal clefts, it is important to document the regressive changes of the notochord in the course of ossification of the vertebral body. During the stage of chondrification, the presence of notochordal cells is gradually reduced to the intervertebral disc spaces. The remnant of the notochord, the perichordal sheath containing a few or no notochordal cells, remains in a slightly ventral portion of the vertebral body,

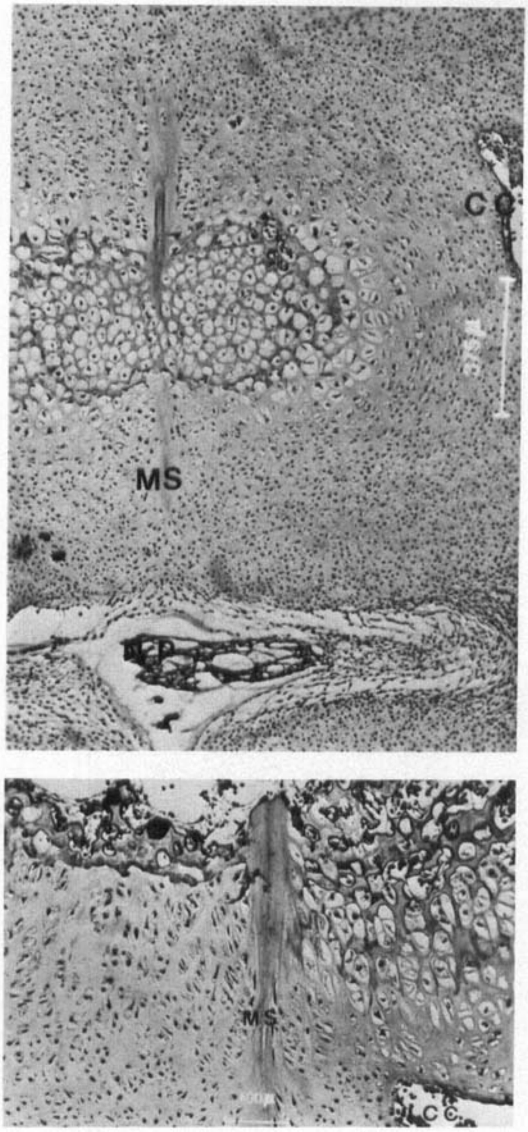


Figure 3. Sagittal section showing enchondral ossification and a mucous streak. a) Hypertrophy of cartilage cells and beginning of calcification in a 12-week-old specimen. Goldner. b) Resorption of calcified cartilage and beginning of ossification. The mucous streak (M.S.) interferes little in the process. 14-week-old embryo. Goldner. C.C. cartilage canal. N.P. nucleus pulposus.

and it is called the mucous streak. Histologically this is a homogeneous or a very fine fibrillar structure, resembling the matrix of fibro-cartilage. At this stage, however, it can be easily distinguished from the cartilage by its more eosinophilic staining. Shortly after the central ovally shaped portion of the body has undergone calcification at 8–10 weeks, ossification follows calcification (Figure 3a, b). As ossification progresses, the mucous streak is gradually broken up by the process of ossification in the central portion (ossification centre), whereas at the periphery (remaining cartilaginous part of the body) the streak gradually becomes part of the cartilaginous matrix; by 20 weeks no remnants of the notochord persist in the vertebral body. The process of ossification spreads radially from one calcified central portion.

In some cases, however, ossification develops, or progresses more rapidly in two areas, ventrally and dorsally, even if calcification or early ossification has begun as a single centre. A 13-week-old fetus shows the latter type of ossification (Figure 4). Ossification has started as

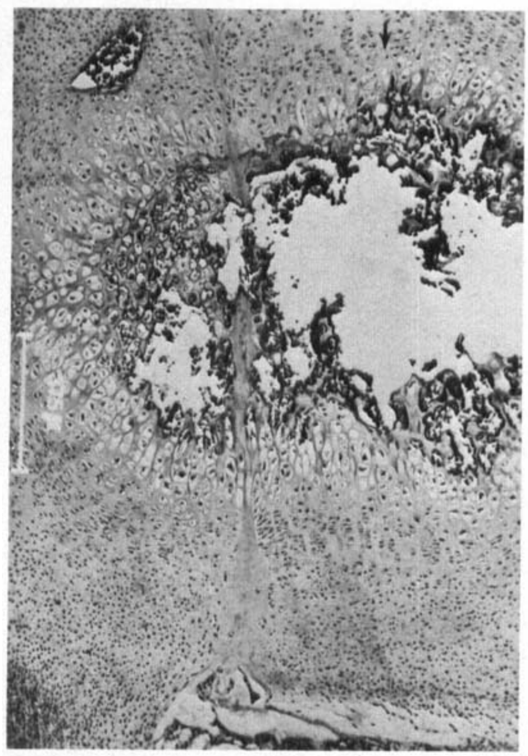


Figure 4. Sagittal section of a 13-week-old embryo showing a slightly anterior position of the mucous streak and a delay in hypertrophic changes (arrow) post posterior to it. Goldner.

Table 1. Radiological assessment

	Cervical 553	Dorsal 1569	Lumbar 785	Sacral 391	Coccygeal 1	Total 3299 bodies
Complete	–	6	4	1	–	11
Incomplete	1	15	38	1	–	55

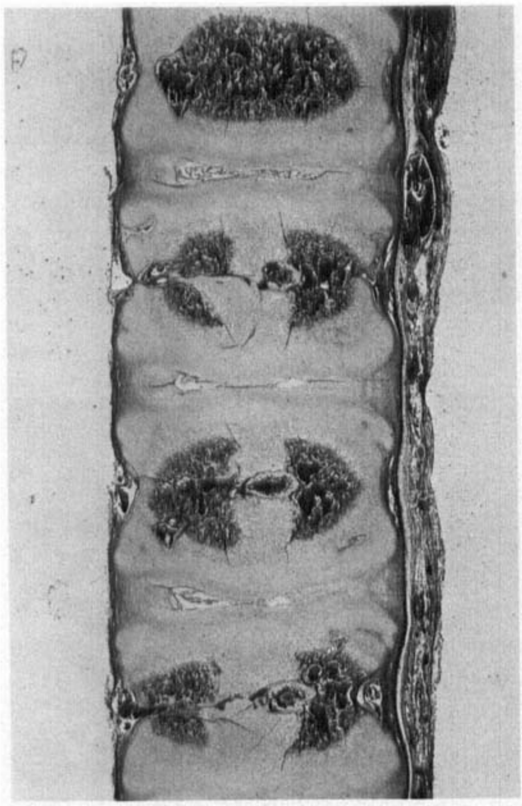
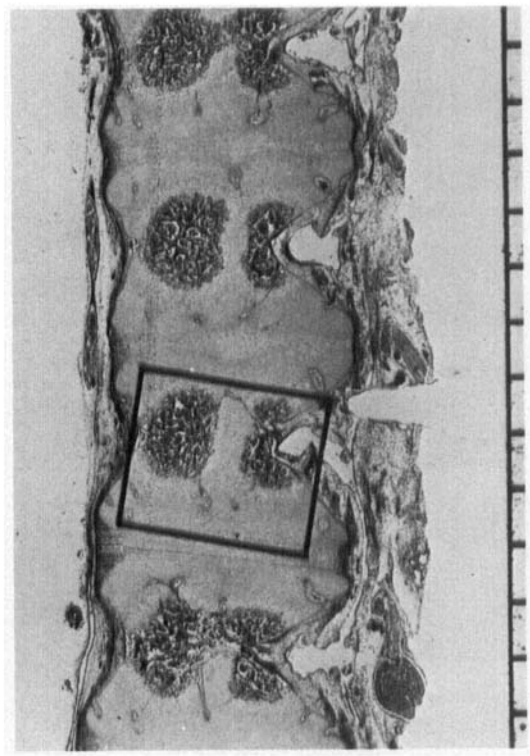


Figure 5. A 21-week-old fetus showing a vascular connection between the centres surrounded by bone. Due to the level of the section, the connection is incomplete in two vertebrae. Sagittal section. Goldner ×9.



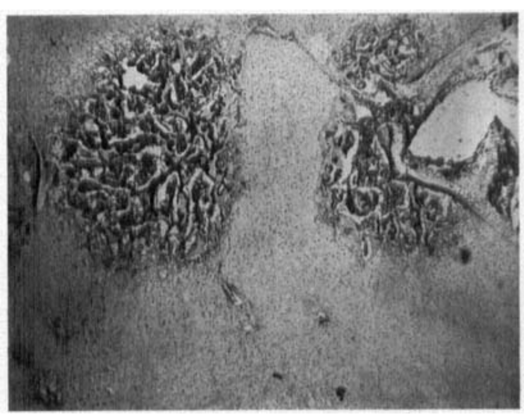


Figure 6. a) Sagittal section of a 20-week-old fetus showing two complete and two incomplete cartilaginous septa. The slightly posterior position of some septa is evident. H.P.S. b) Few hypertrophic chondrocytes are seen in the septum close to the ossifying centres. ×40.

one ossification centre, but enchondral ossification, as evidenced by hypertrophic cells, is observed mostly ventrally and dorsally. In the central portion, hypertrophic or degenerative changes of chondrocytes are retarded. This suggests that two nuclei of ossification may develop in this vertebral body.

The notochordal remnants (the mucous streak and some degenerated notochordal cells) are more ventrally situated than the site where enchondral ossification is delayed. Evidently, this site does not correspond to the location of the notochordal tissue.

The radiolucent band in the vertebral body represents histologically an area of cartilaginous tissue which divides the osseous nucleus into the ventral and dorsal ossification centres. Serial sections, however, show that even if a complete cleft is clearly recognized radiologically, there is a transverse vascular channel surrounded by bone, connecting the two ossification centres in the central portion of the body (Figure 5). In this sagittal section, endochondral bone formation is observed to progress normally at the periphery of the two ossification centres, but is delayed between both centres. In fact, it is less active at the vertical boundaries between the ossification centres and at the transverse vascular channel. Chondrocytes which constitute the septum show different features. In the mid-portion of the septum, most of chondrocytes are similar to resting ones. Toward the ossification centres, there are some hypertrophic chondrocytes which contain elongated nuclei. Degenerated chondrocytes are seen in the calcified matrix. The activity of these cells indicate a process of a delayed endochondral

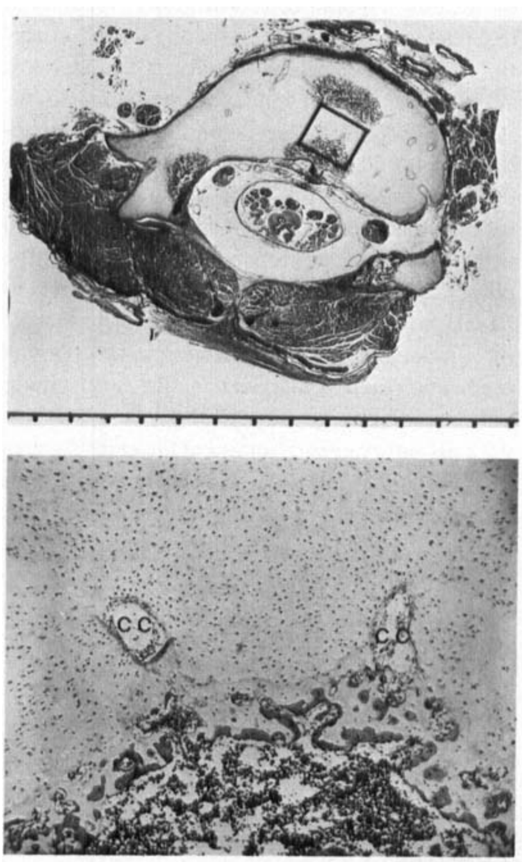


Figure 7. a) Coronal section of a 17-week-old fetus. Note again the posterior position of the septum. Goldner. b) The process of enchondral ossification is less active at the sides facing the other centre. C.C. cartilage canal. ×60.

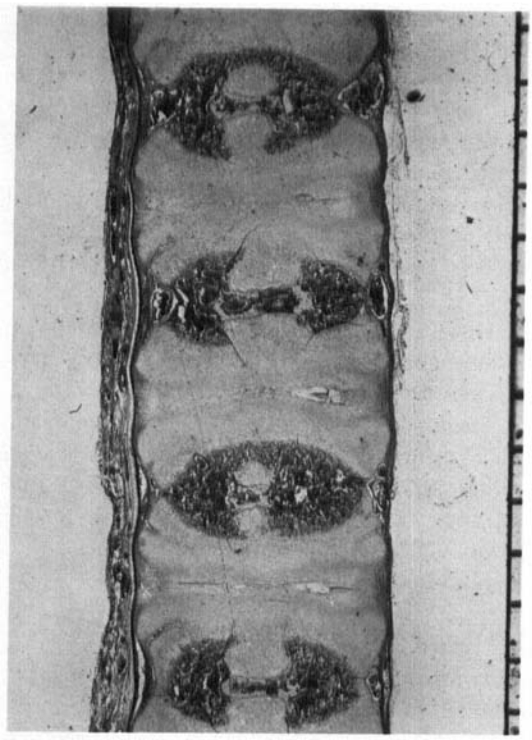


Figure 8. The progress of enchondral ossification in incomplete bars is more pronounced at the periphery; it leads to the formation of cartilaginous islands. H.P.S.

bone formation. No notochordal derivatives can be seen (Figure 6a, b).

A cross section shows endochondral bone formation taking place at the periphery of the two ossification centres. It seems to be delayed at the sites where the centres face each other. The ventral ossification centre is larger than the dorsal one. A thorough search did not reveal remnants of notochordal tissue in none of the serial sections (Figure 7a, b).

Histological findings of incomplete clefts resemble those of the complete clefts. They show various patterns of delayed ossification.

Enchondral ossification in cases of incomplete clefts progressed faster at the periphery leading to the formation of cartilaginous islands (Figure 8).

DISCUSSION

A coronal cleft vertebra is known as a transient, normal variant of ossification of the vertebral body. Although the word "cleft" is still used, it is rather unsuitable since it is not a true cleft, but a layer of normal cartilaginous tissue around the central vascular channel interposed between two ossification centres. The dominance in males has been confirmed by this study. According to Cohen et al. (1956) the incidence of coronal cleft is 5%. Both facts were also confirmed. However, the incidence of cleft vertebrate would be much higher if complete clefts were included.

The pathogenesis has been attributed to the persistence of notochordal tissue. In fact, Schmorl & Junghanns (1975) thought that the persistence of the notochord disturbs vertebral development. Cohen et al. (1956), Wollin & Elliott (1961), and Meyer-Burgdorf & Klose-Gerlich (1935) stated that clefts were occupied by the notochordal tissue. No explanation has ever been forwarded as to why cleft formation secondary to persistence of notochordal tissue occurs only in one plane. The results of our study do not permit to support the notochordal theory. We could not observe any evidence of persisting notochordal tissue. Also, our previous study (Tanaka & Uhthoff 1981) showed that, in the early stage of development vertebral malformation was not associated with abnormalities of the notochord. Schmorl & Junghanns' (1975) schema of the pathogenesis of vertebral malformations, including coronal clefts, appeals for didactic reasons. However, it lacks a scientific foundation. Not a single instance of notochordal remnants inside the clefts was observed in any of the serial sections of our material. Only cartilage and vascular mesenchyme were present in the cartilaginous septum. Here chondrocytes show no specific features other than changes compatible with a delayed process of endochondral bone formation. Cohen et al. (1956) concluded that "... the variability of the cartilage cells in the band and the absence of an obvious source of origin (e.g. perichondrium, zone of reserve cells) are significant points suggesting notochordal derivation of these cells". Wollin & Elliott (1961) mentioned that "microscopically the section showed a central core of irregularly arranged cartilage cells in a very loose matrix of connective tissue mucin, indicating notochordal derivation". These descriptions do not identify notochordal tissue convincingly. We believe that only a mucous streak or notochordal cells, as seen in the intervertebral disc, constitute a reliable evidence of the notochordal persistence at this stage. The mucous streak, however, lying slightly anterior to the site of a cleft, undergoes normal endochondral ossification and disappears in the cartilaginous part of the body by about 20th week. Notochordal cells were never observed in the cleft. As seen in Figures 3b and 4a, the mucous streak may cause a slight delay of the maturation of cartilage cells locally but never inhibits the process of endochondral ossification as evidenced by the presence of hypertrophic cells in intimate contact with the streak. On radiographs, taken in a lateral projection, the clefts are located in the midportion of the vertebra or slightly posterior to it. To the contrary, the notochord or, in the later stage, the mucous streak is located in the anterior portion of the body. Evidently, the location of the clefts does not correspond to that of the notochord.

Finally, the observation that clefts occur mostly in the thoracic and lumbar vertebrae is difficult to reconcile with the fact that the notochord extends over the whole vertebral column.

The variations from the patterns of ossification suggest the following process: calcification of cartilage starts in the centrum, but ossification develops in two areas, the dorsal and ventral portions of the body. Serial sections, permitting the reconstruction of the third dimension showed that none of the clefts was complete. We believe that in cases of coronal clefts endochondral bone formation progresses around two ossification centres which are connected by a vessel giving it a dumbbell shape. In some cases, the two ossifying centres merge first at the periphery, so that a cartilaginous island remains in the middle.

These concepts are also compatible with the fact that the coronal cleft clinically disappears shortly after birth (Cohen et al. 1956, Schmorl & Junghanns 1975).

The shape of the ossified area seems to be determined by vascular distribution in fetuses. Thus different patterns of vascular supply are likely responsible for the different patterns of ossification.

Our study permits to refute the theory that the notochord is responsible for the development of cleft formation; it confirms the current thinking that coronal clefts do not lead to any disturbance of the vertebral development.

CONCLUSIONS

In a collection of 266 human embryos and fetuses, coronal clefts were found in nine cases. However, in none of these cases was a complete separation between the two ossification centres present. A bony bridge containing a blood vessel always connected the two centres.

Incomplete clefts were observed in 16 cases radiologically or histologically.

No remnants of the notochordal tissue could be recognized in the cartilaginous septum. After the stage of chondrification, the notochordal cells were confined to the intervertebral disc spaces. The mucous streak disappeared around the 20th week. Coronal clefts are variations of normal enchondral ossification, they are never complete and they are most probably due to different patterns of blood vessel distribution.

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