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Congenital Epulis of the Newborn (A Case Report)†

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= Abstract =Congenital epulis of the newborn appears at birth as a pedunculated, firm mass on the anterior alveolar ridge. Although more common on the maxillary alveolar ridge, the lesion may occur on the mandibular ridge. In 1871, Neumann first described a "congenital epulis" in a newborn girl. A comprehensive review was published by Fuhr and Krogh in 1972; they cited 113 cases in the literature and described a case of their own. The tumor size varies from a few millimeters to 3 to 4 centimeters in diameter. Rarely, a large congenital epulis sufficient to occupy most of the mouth and cause feeding and respiratory difficulty has been reported.

We present a 12-hour-old Korean girl born with a giant congenital epulis which requires immediate surgical excision for normal nursing and airway compromise.

The histogenetic origin of the tumor cells is ambiguous and the principal theories of origin are odontgenic, myoblastic, fibroblastic, or neurogenic. We also present electromicroscopic and immunohistochemical studies of this tumor in order to search for histogenesis of this kind of tumor.

Key words: ***Giant congenital epulis***

INTRODUCTION

Epulis means "on the gum" and congenital epulis of the newborn is a benign soft tissue tumor of ambiguous origin that is found on the alveolar mucosa at birth. Although the lesion is benign, rarely, a large epulis can cause nursing or respiration problems and require immediate surgical removal.

We present a 12-hour-old Korean girl who has a large congenital epulis measuring 3.5 × 2.5 × 2.0 centimeters in size. The tumor's very large size was sufficient to occupy most of her mouth and cause feeding and respiratory difficulties (Fig. 1). We also present electromicorscopic and immunohistochemical studies in order to search for the histogenesis of this tumor.

CASE REPORT

The infant was born following a full and normal term pregnancy. She was the first sibling of unrelated parents. Her mother's age was 27.

Physical examination: Examination revealed a well nourished, normally developed 2,830 g newborn baby girl. Intraoral examination revealed a soft tissue mass, 3.5 centimeters in maximum diameter, attached to the alveolar process of the maxilla, to the left of the midline. The mass was ovoid, pedunculated, and firmly fixed by a broad pedicle. The mucosa covering the tumor was normal in appearance, as were all the tissues of the oral cavity.

Results of the remainder of the physical examination were within normal limit.

Surgical procedure: The infant was seen in the operating room 12 hours after birth for the removal of the tumor. Under the general anesthesia with endotracheal intubation, the infant was placed in the supine position. The pedicle of the tumor was clamped with small angled vascular clamp and the tumor was excised with No. 15 blade. The pedicle was 1.5 centimeter in length. The wound was closed with 4-0 chromic catgut by continuous double ligation suture.

Transient abdominal distension developed post-

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operatively, but subsided by conservative management 2 days later. The infant was discharged with her mother on the 10th day after delivery. She has been followed up for 6 months without eivdence of recurrence (Fig. 2).

Pathologist's report: Examination of the specimen showed an ovoid mass that measured 3.5 × 2.5 × 2.0 cm in size (Fig. 3). The cut surface was homogenously pinkish gray, and the tumor was well circumscribed.

Microscopically, the tumor was vered by stratified squamous epithelium without rete ridges, and composed of sheets of large, closely packed polygonal or ovoid cells with abundant pale acidophilic granular cytoplasm and one nucleus (Fig. 4). The nuclei were centrally or eccentrically located and relatively small and round or ovoid with one nucleolus. No atypism or mitosis was encountered. Cell membrane were usually distinct. Stroma was scanty with numerous capillaries and a few scattered epithelial rests (Fig. 5,6).

Electron microscopic study revealed numerous intracellular membrane bound granules, presumably autophagic vacuoles containing finely granular structures and small masses of electron dense material.

Junctional complexes between the tumor cells were occasionally encountered (Fig. 7, 8). Immunohistochemical study using peroxidase-antiperoxidase method failed to demonstrate S-100 protein in this tumor.

DISCUSSION

Congenital epulis, first described by Neumann (1871), is usually found in the maxilla and has a predilection for girls. It is usually a pedunculated lesion found in the incisor region, apparently arising on the crest of the alveolar ridges of process. It ranges from a few millimeters to several centimeters in diameter. The large protruding mass in the present case measured 3.5 centimeter in maximum diameter, making it one of the largest ever reported.

There has been little agreement regarding the histogenesis of the tumor. Histologically, it is similar to the granular cell tumor which has been called "granular cell myoblastōma"-so that these epulis tumors have been given this name in the past, and considerable confusion has existed. Custer and Fust (1952) have emphasized the differences between congenital epulis and granular cell myoblastoma.

(1) Uniform structure in congenital epulis, not uniform in granular cell myoblastoma.

(2) Congenital epulis contains no neural components, and no pseudoepitheliomatous hyperplasia.

(3) The prominent vascular component is not found in granular cell myoblastoma.

(4) Congenital epulis occurs only in the newborn.

(5) The typical location of congenital epulis in the incisor region of maxilla or mandible is not a matter of chance.

(6) There is a higher incidence of epulis in female infants. Recent electron microscopic studies support the theory of a neural origin of granular cell myoblastoma. Fisher and Wechsler (1962) observed small, relatively unmyelinated axons, and structures that resembled degenerating myelinated axons in the cytoplasm of some tumor cells. However, Kay and his co-workers (1971), in describing the ultrasturcture of congenital epulis of the newborn, were unable to identify either Schwann's cells or axon fibers. Their electron microscopic studies demonstrated junctional complexes between the cells in congenital epulis and this suggested an epithelial origin for this tumor.

In our case, we also could not find either Schwann's cells or axon fibers and could not demonstrate S-100 protein by immunohistochemical study using peroxidase-antiperoxidase method in this tumor. However, a few epithelial rests were scattered and our electron microscope study of this tumor revealed junctional complexes between some of the granular cells which suggested that they may be of epithelial origin, although the studies were not entirely conclusive.

Although the lesion is benign, when it is very large it can cause nursing or respiration problems and immediate surgical removal is necessary. Bowe (1974) also reported a similar lesion and proposed that the bulk of the mass and its appearance were such that prompt surgical removal seemed desirable.

She has been followed up for 6 months without evidence of recurrence.

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= 국문초록 =

응급수술을 요하였던 신생아의 선천성 거대치육종 치험례

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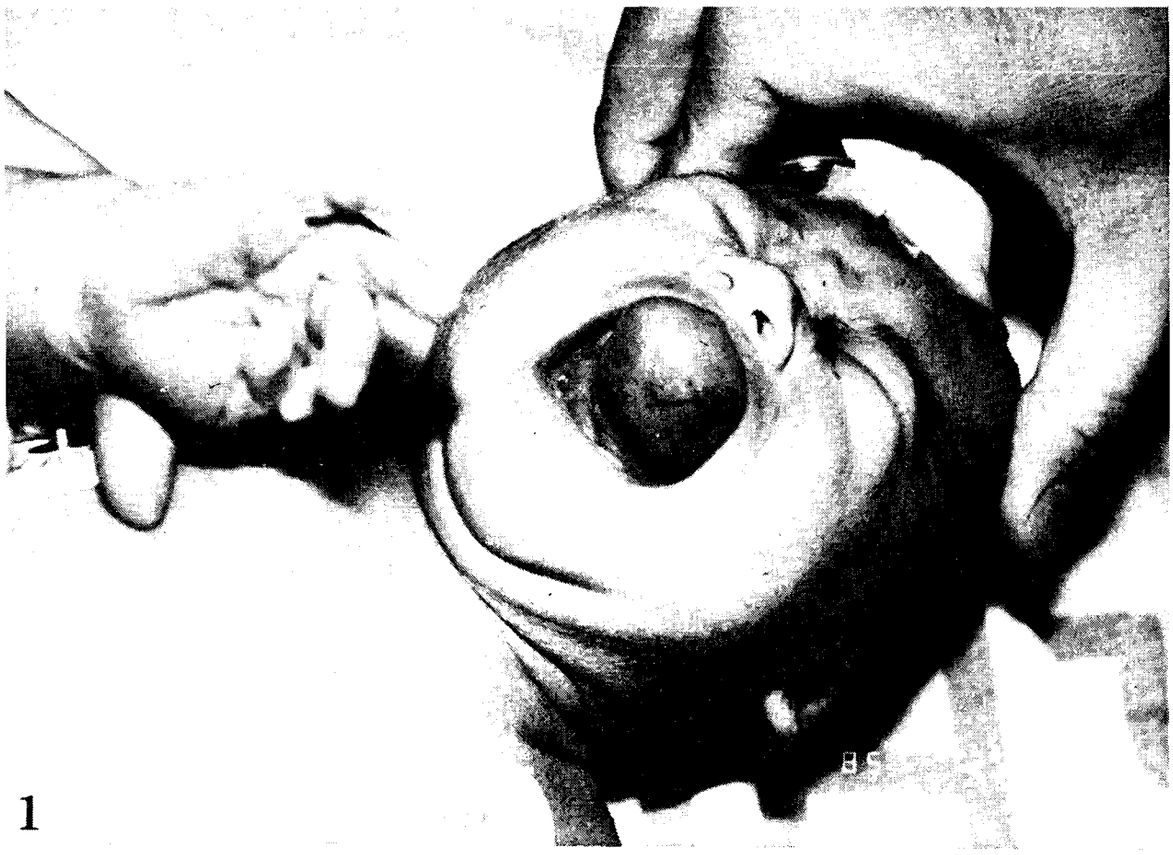
오갑성 · 박철규

선천성 치육종은 치육 특히 그 경계부에서 유경성 혹은 이에 흡사한 상태로 증식한 신생물들을 말한다. Neumann이 1871년에 처음 기술한 이래 세계적으로 약 100여례가 보고된 바 있다. 그러나 신생아의 구강과 호흡기를 폐쇄, 기능장애를 일으킬 정도로 큰 경우는 매우 드물다. 저자들은 생후 12시간 된 신생아에서 정장식이와 호흡에 중대한 지장을 초래한 거대 치육종을 경험한 바, 수술절제한 후 신생아는 곧 정상 호흡과 수유가 가능하였다.

이 선천성 치육종의 발생기전은 상피세포기원설, 근세포기원설과 신경세포기원설 등이 있는 바, PAP 방법을 이용한 면역형광 염색법과 전자현미경등으로 그 발생기전을 찾아보았다.

LEGENDS FOR FIGURES

Fig. 1. A 12-hours old Korean girl suffering from a giant epulis occupying most of her mouth
Fig. 2. 1 month after operation
Fig. 3. Gross appearance of the tumor
Fig. 4. Photomicrograph of the tumor. The tumor was covered by stratified squamous epithelium without rete ridges. H&E X 100
Fig. 5. Photomicrograph of the tumor. The tumor composed of sheets of large, closely packed polygonal or ovoid cells with distinct cytoplasmic border. Stroma was scanty with numerous capillaries. H&E X 100
Fig. 6. Photomicrograph fo the tumor, showing a epithelial rest. H&E X 400
Fig. 7. Electron micrography of tumor, showing numerous intracellular membrane bound granules, presumably autophagic vacuoles containing finely granular structures and small masses of electron dense material (X 8,400)
Fig. 8. Electron micrography showing a junctional complex between the tumor cells (X 22,400)



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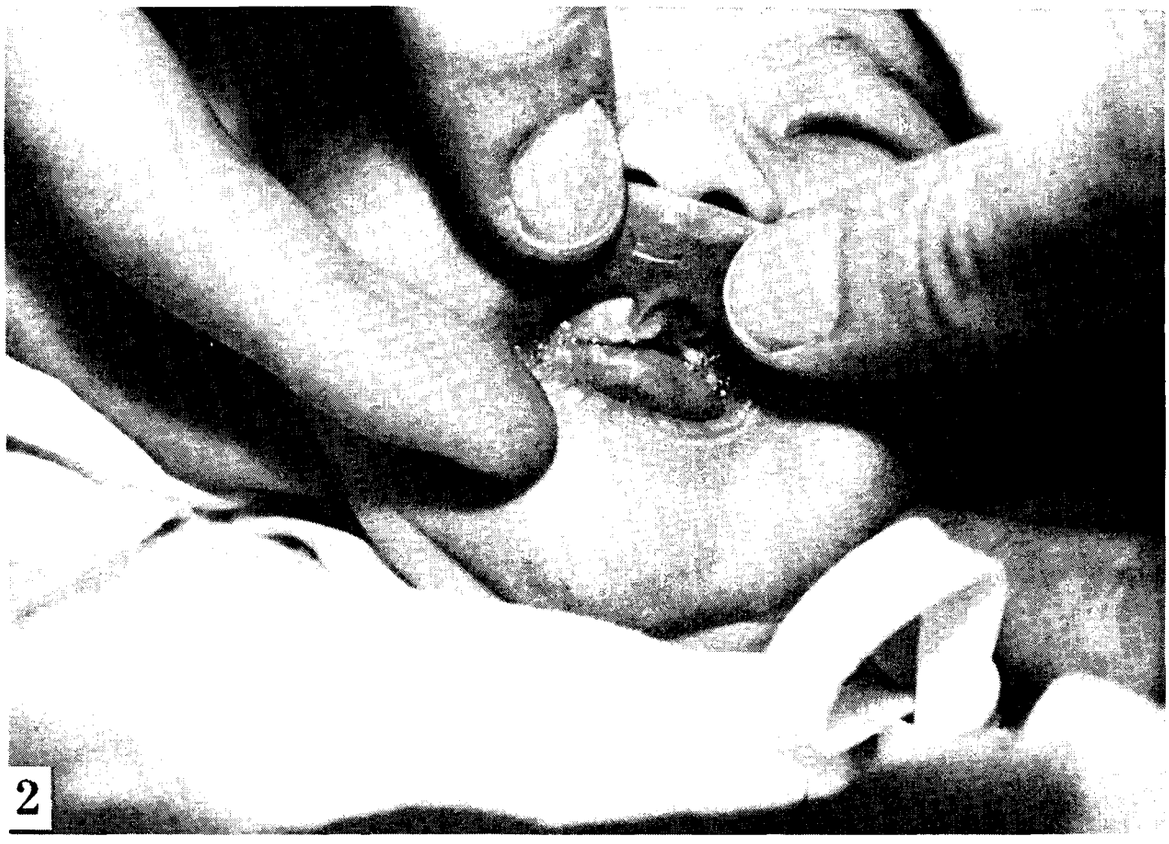

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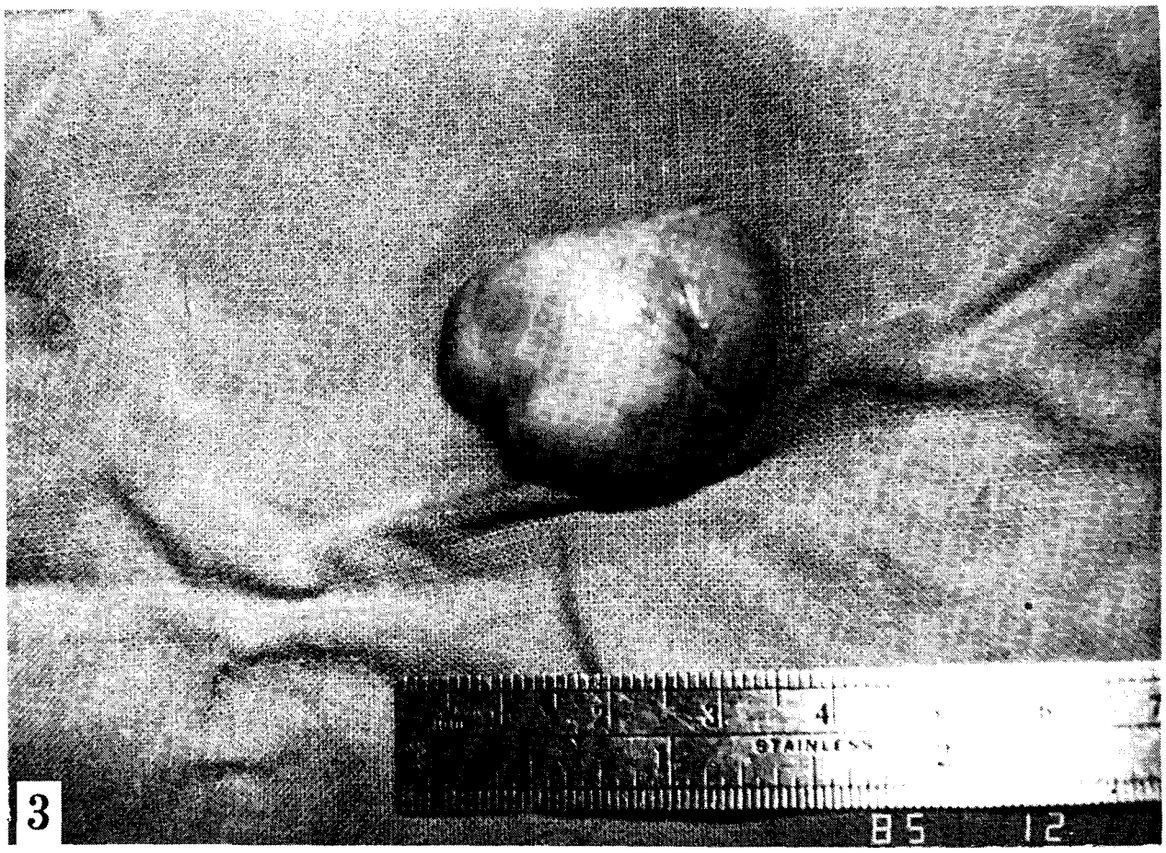
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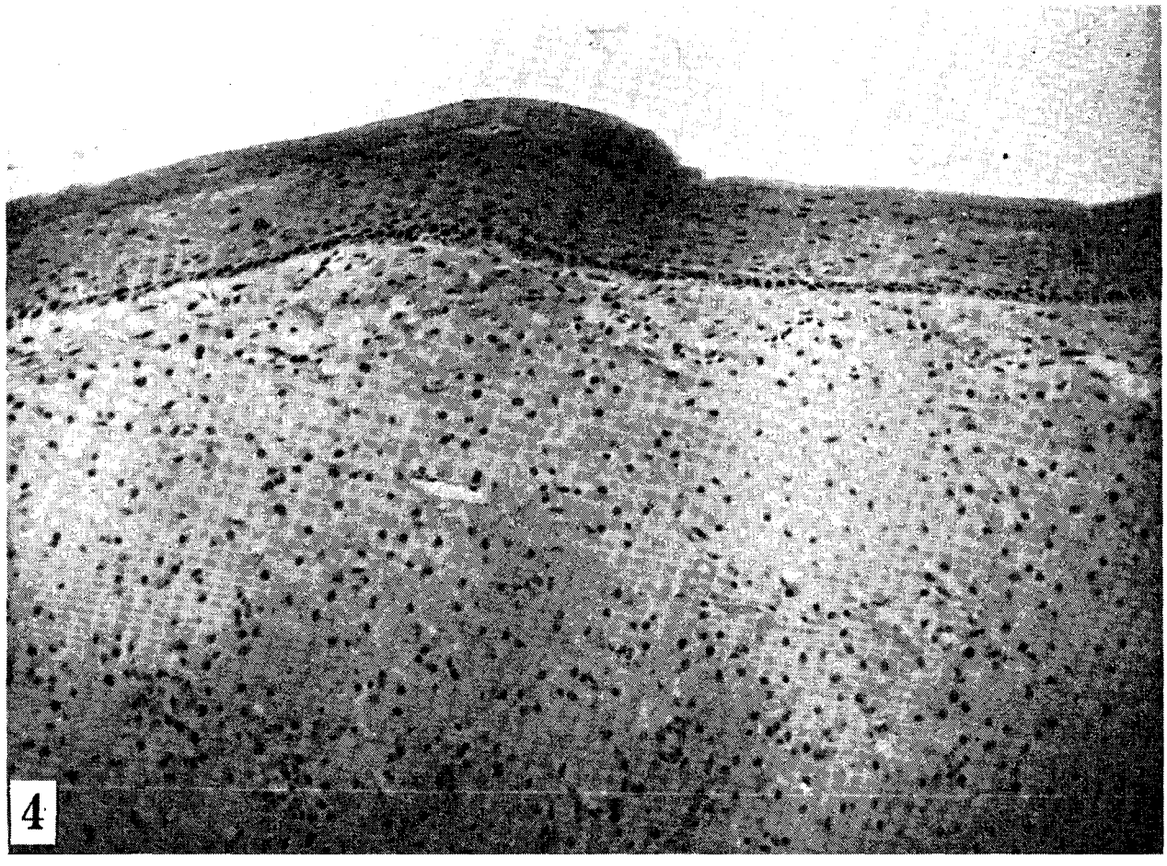
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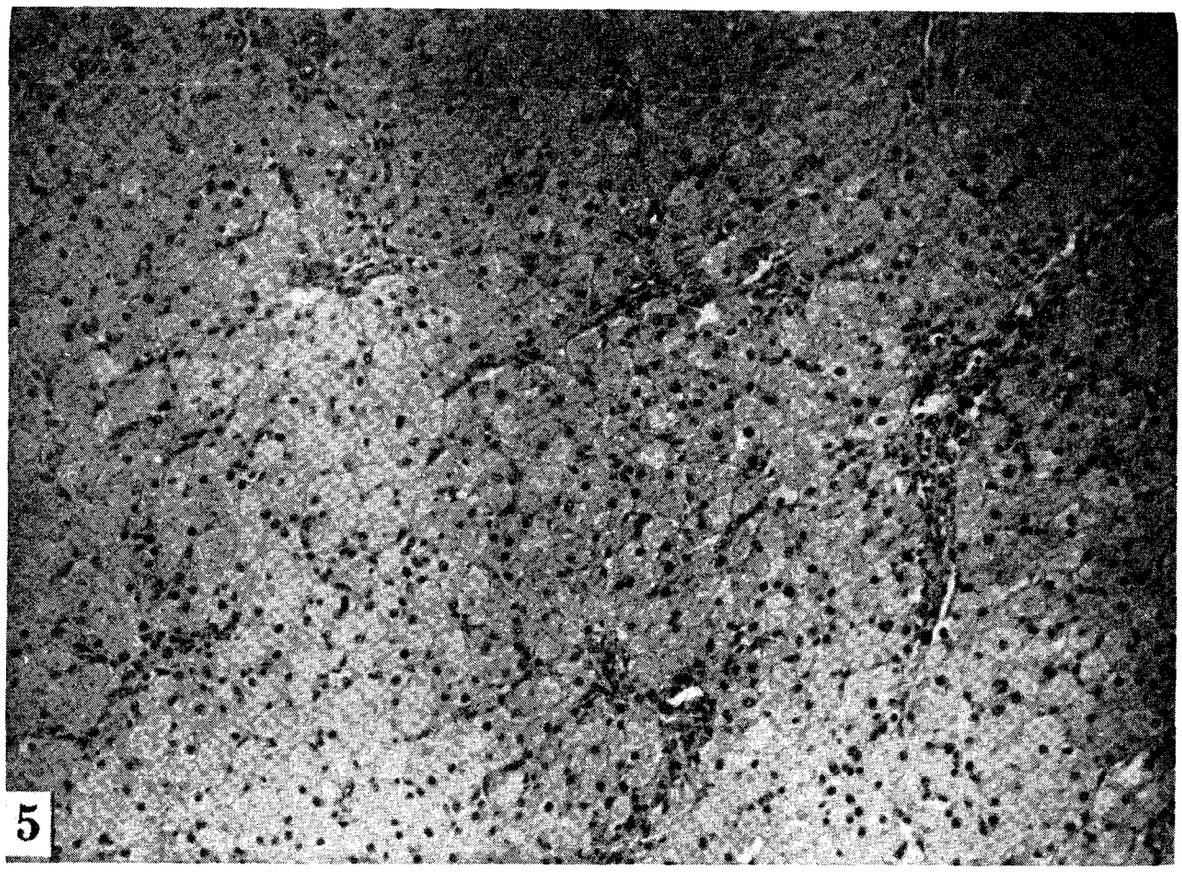

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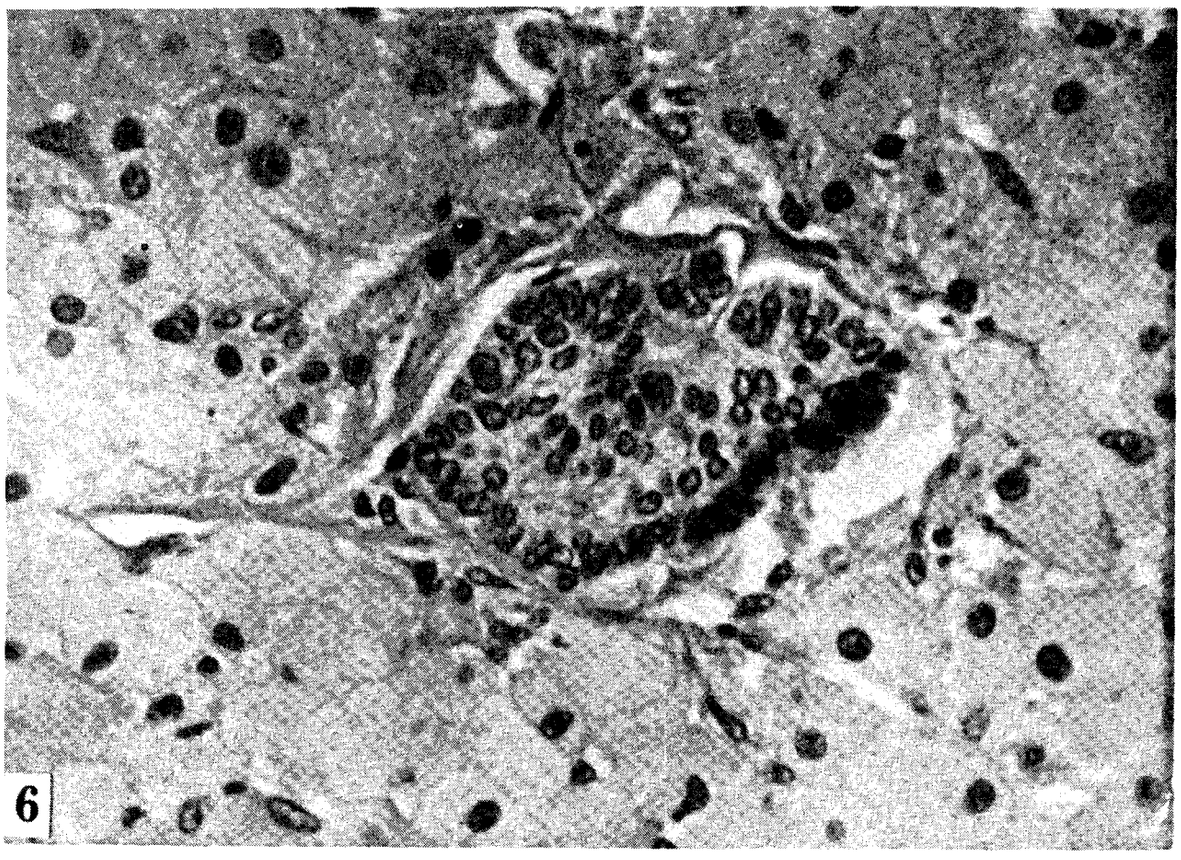

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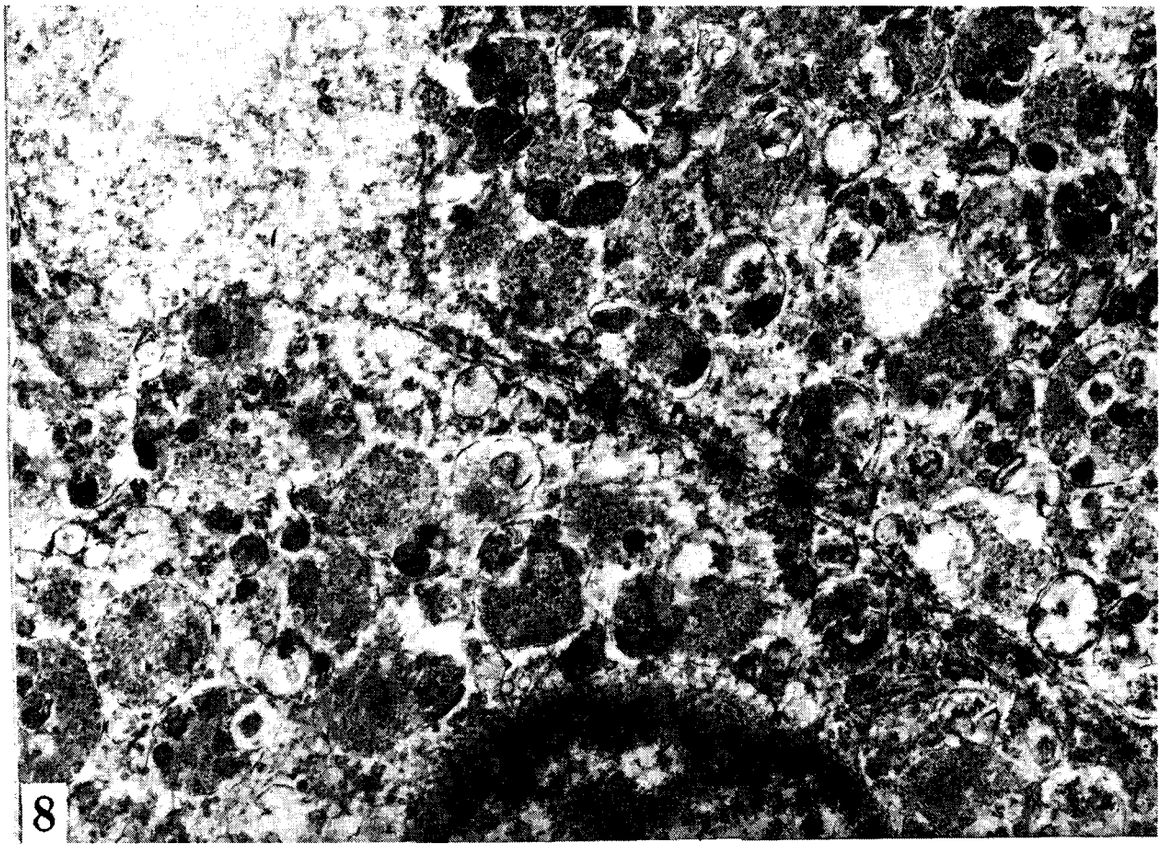

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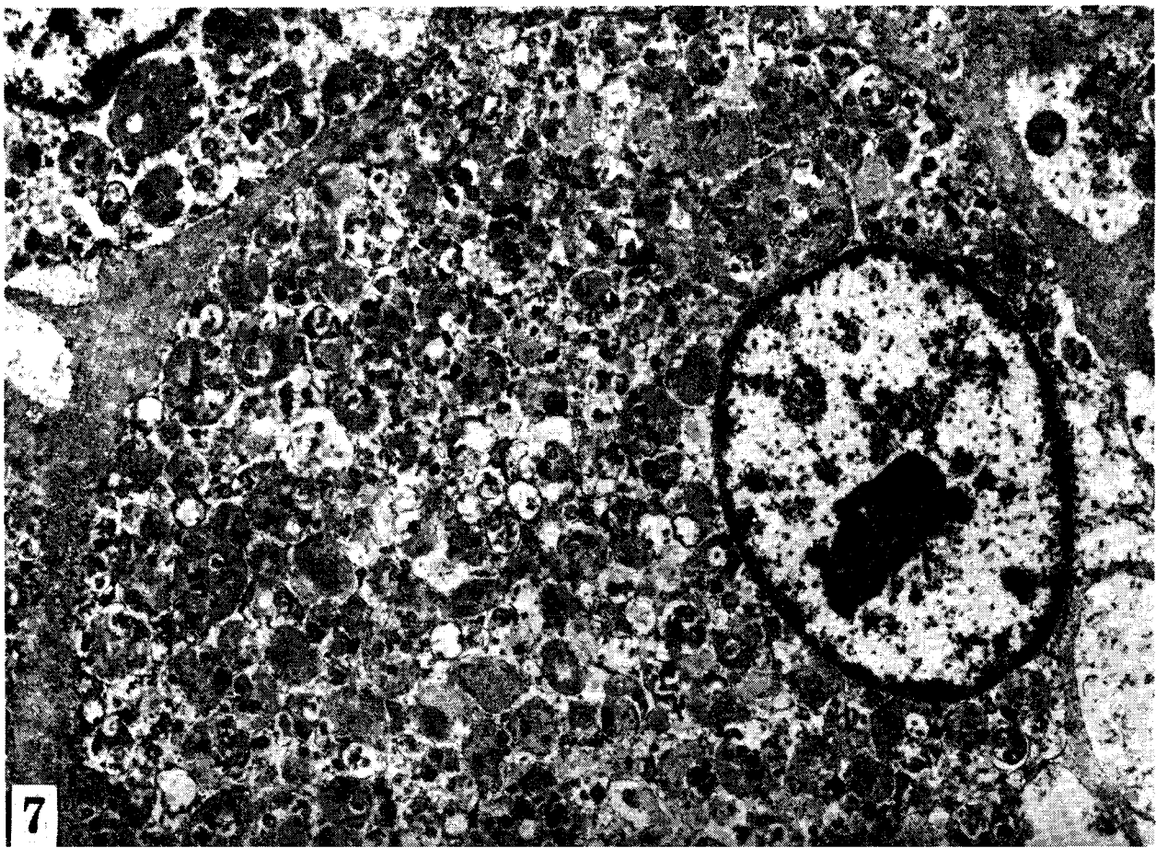

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