

CASE REPORT



Ectopic lamellar Pacinian corpuscle within the thymus. Atypical or abnormal location?

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Abstract

Lamellar (Pacinian) corpuscle is a cutaneous encapsulated sensory corpuscle, mainly functioning as a rapidly adapting low-threshold mechanoreceptor with characteristic "onion skin"-like appearance. Even though the Pacinian corpuscle is typically located in the skin, histomorphologically and confirmed by using immunohistochemical methods, we have identified it within the interstitium of the thymus of a newborn with congenital heart defect. To the best of our knowledge, this is the first such account ever to be published. The structure of the thymic Pacinian corpuscle was the same as the Pacinian corpuscle in the skin. The ectopic thymic Pacinian corpuscle can be hypothetically explained as the consequence of impaired migration of neural crest cells, since neural crest-derived cells play an important role in the development of the Pacinian corpuscle, as well as the thymus and heart. In general, the occurrence of ectopic Pacinian corpuscle in different organs is rare. In the scientific literature, there are reports of its sporadic presence in the pancreas, in the lymph nodes, inside the prostate and the urinary bladder wall. Our report presents the first described case of the Pacinian corpuscle in a heterotopic location in the thymus. Similar to other incidental findings of this anatomic structure, explanation of its ectopic development, as well as its local function remains only speculative.

Keywords: Pacinian corpuscle, thymus, ectopy, neural crest-derived cells, immunohistochemistry.

Introduction

Discovering an ectopic tissue within any organ is always a histological peculiarity. Those findings, which cannot be unambiguously explained in terms of embryonic development, are always the most remarkable. A perfect example is the occurrence of acid-secreting gastric mucosa, pancreatic tissue or large intestinal tissue within the Meckel's diverticulum of the small intestine, usually near the ileocecal valve [1, 2]. Although this finding is relatively common, it still belongs in this category of "enigmatic" ectopically localized tissue. Similarly, interesting is the presence of thyroid gland tissue within the liver [3] or within adrenal gland [4], or bronchial tissue in the heart [5]. These findings cannot be explained by impaired cell migration outside the migration pathway of progenitor cells during embryonic development. Those anomalies, which are characterized by proliferation of histologically normal tissue in abnormal/ectopic locations, can be termed "choristomas", e.g., the presence of a plate of hyaline cartilage inside the tongue [6], or the presence of mature thymic tissue inside the lungs [7].

Lamellar (Pacinian) corpuscle (*corpusculum lamellosum*, Vater-Pacinian corpuscle) is a cutaneous encapsulated sensory corpuscle, mainly functioning as a rapidly adapting low-threshold mechanoreceptor with characteristic "onion skin"-like appearance. Pacinian corpuscles are normally localized in the deep layer of the dermis (reticular dermis)

and hypodermis of the hairless (glabrous) mammalian skin [8].

Aim

This article discusses a curious case not yet described in the through database available scientific literature – histologically and immunohistochemically verified Pacinian corpuscle within the thymic tissue of a newborn. Over the last 10 years, the pathologists participating in this study have examined more than 1500 juvenile thymuses, as an integral part of their routine practice of surgical pathology. The thymuses came from newborns and children who had their thymuses removed during cardiac surgery for various congenital heart defects. Despite the quantity of thymuses examined, we have never encountered the Pacinian corpuscle inside the thymus before. Thus, the article focuses on the possible embryonic explanation for the presence of the Pacinian corpuscle within the thymus and also provides an overview of manuscripts published to date discussing the accidental findings of this structure in different organs.

Case presentation

A female newborn diagnosed with tetralogy of Fallot underwent surgical correction on the third week after birth at Children's Cardiac Center, National Institute of Cardiovascular Diseases, Bratislava, Slovakia. Subtotal resection of the thymus was part of the surgical procedure.

The thymus was fixed in formalin, routinely processed in paraffin, and 5 µm thick slices were cut and stained with Hematoxylin–Eosin for histological evaluation. Immunohistochemistry was performed with the FLEX system, with application of prediluted ready-to-use antibodies against S100, cluster of differentiation (CD) 34, CD56 and synaptophysin (Agilent, Santa Clara, CA, USA), according to the manufacturer's instructions. The study was performed in congruence with the Bratislava University Hospital Ethics Committee guidelines.

In the period of 17 years, over 1500 thymuses have been histologically evaluated at the Institute of Pathological Anatomy, Faculty of Medicine, Comenius University in Bratislava, Slovakia. The presented case is the first and the only with the finding of a lamellar Pacinian corpuscle. In the specimen with normal histomorphological structure of a female newborn thymus, a formation of oval shape, with the longest diameter up to 0.5 mm and concentric lamellar composition, was noticed in the interstitium in close relation to blood vessels and identified as a Pacinian lamellar corpuscle (Figure 1). The microscopic structure of the thymic Pacinian corpuscle was similar to those localized in the skin with one inner core, outer core and the external fibrous capsule. Using immunohistochemical (IHC) methods, we clearly described the main components (Figure 2): (i) synaptophysin-positive nerve fiber was present in the center of the inner core; (ii) neural cell adhesion molecule (CD56)-positive Schwann-like cells attached directly to the nerve fiber, and forming the central

portion of the inner core; (iii) multilayered S100 antigen-positive Schwann cells forming the inner core; (iv) CD34-positive endothelial cells of blood capillaries present at the outer surface of the inner core and among connective tissue cells of the outer core.

The patient has no history of familial heritable disease or neurofibromatosis and six months after heart surgery is cardially compensated and doing well.

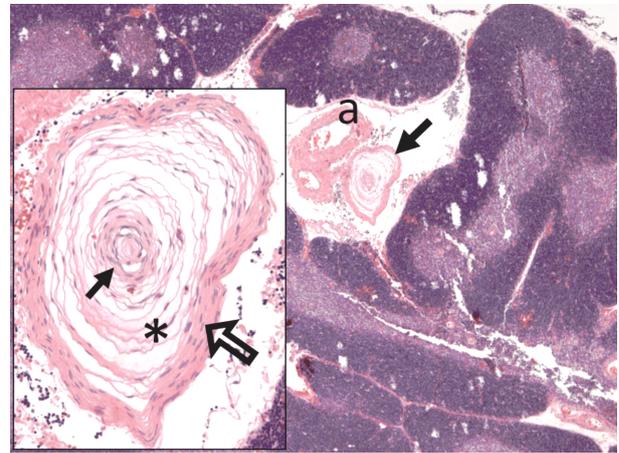


Figure 1 – Human thymus with normal histological structure; next to an artery (a), there is a formation (arrow) with typical appearance of a lamellar (Pacinian) corpuscle (insertion) with the inner core (short arrow), outer core (*) and the external fibrous capsule (open arrow). Hematoxylin–Eosin staining, $\times 25$ (inset, $\times 100$).

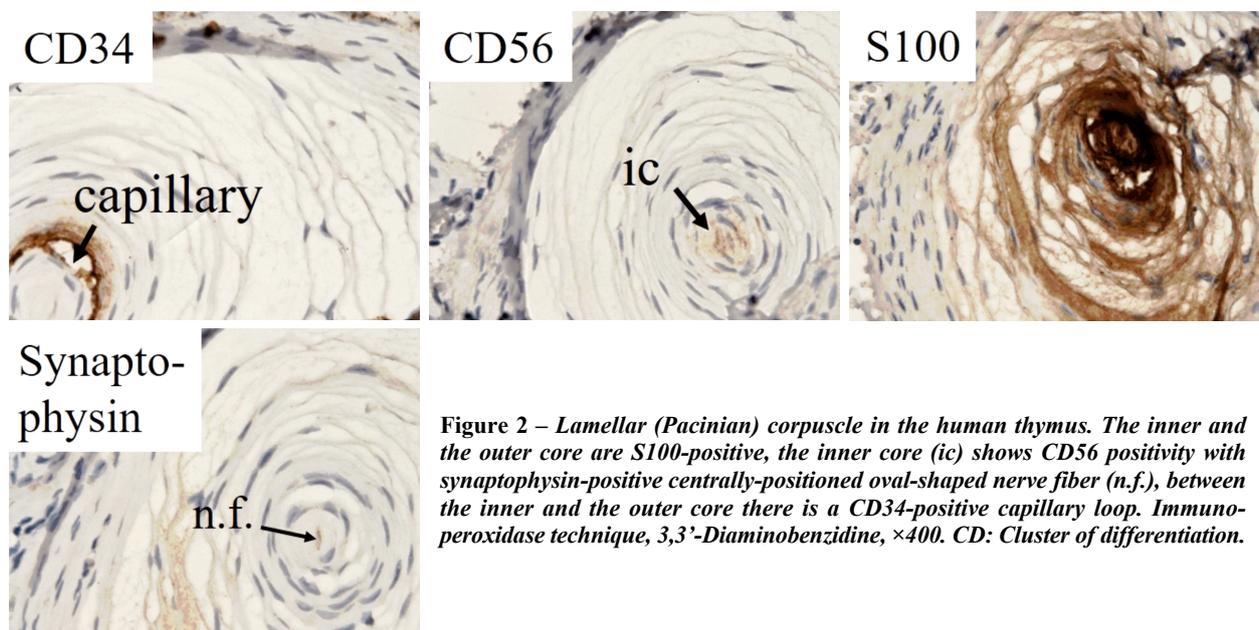


Figure 2 – Lamellar (Pacinian) corpuscle in the human thymus. The inner and the outer core are S100-positive, the inner core (ic) shows CD56 positivity with synaptophysin-positive centrally-positioned oval-shaped nerve fiber (n.f.), between the inner and the outer core there is a CD34-positive capillary loop. Immunoperoxidase technique, 3,3'-Diaminobenzidine, $\times 400$. CD: Cluster of differentiation.

Discussions

Pacinian corpuscles are encapsulated mechanoreceptors and play a role in the sensation of pressure, vibration and touch. Normally, they are functionally connected to myelinated nerve fibers that originate from sensory neurons of the dorsal root ganglia [9]. Even though the Pacinian corpuscle is typically located in the skin, histomorphologically and confirmed by using IHC methods, we have

identified it within the interstitium of the thymus. To the best of our knowledge, this is the first such account ever to be published. The size, structure and IHC profile of the thymic Pacinian corpuscle was the same as the Pacinian corpuscle in the skin or pancreas [8, 10].

The occurrence of ectopic Pacinian corpuscle is rare. In the scientific literature, there are reports of its sporadic presence in the pancreas [9, 11], in the lymph node [12–15], inside the prostate [16, 17] and the urinary bladder

wall [18]. In animals, the Pacinian corpuscle was found in the mesentery of cats [19] and nerve endings similar to the Pacinian corpuscles were also described in the lungs of Indian black rats [20].

Hypothetically, it could be assumed that thymic Pacinian corpuscles play a role in monitoring of the pressure changes within the mediastinum. The significance of such assumption is related to a possibility that vibration and pressure can influence thymic function. However, considering the sporadic nature of such case not yet described in the literature, it is more probable that it is merely an accidental ectopic Pacinian corpuscle within the thymic tissue with no specific function. In spite of that, there is one plausible embryonic explanation for the presence of the Pacinian corpuscle especially within the thymus of a patient diagnosed with tetralogy of Fallot. During the prenatal development, multipotent neural crest-derived cells differentiate firstly into Schwann cell precursors, which subsequently form a population of adult Schwann cells of the inner core of the Pacinian corpuscle and also partially differentiate into so-called endoneurial fibroblasts of the outer core [10, 21]. The same multipotent neural crest-derived cells are important in the development of the connective tissue components of the thymus [22–24] and are also essential in the coordination of the outflow tract remodeling of the developing heart [25, 26]. Abnormalities in the migration of neural crest cells have been implicated in the pathogenesis of multiple conotruncal defects, including tetralogy of Fallot [27]. Therefore, the ectopic Pacinian corpuscle can be hypothetically explained as the consequence of impaired (halted) migration of neural crest cells (a new form of “neurocristopathy” [28]), since neural crest-derived cells play an important role in the development of the Pacinian corpuscle, as well as the thymus and heart.

Feito *et al.* [14] described the rare sporadic presence of Pacinian corpuscles in lymph nodes of different locations. Interestingly, they identified these structures near blood vessels, which was also the case of our finding of Pacinian corpuscle in the thymus. We also identified the presence of capillaries inside of the corpuscle [29], most probably functioning in maintenance of their metabolism, rather than as a manometer monitoring changes in local blood supply [30].

☐ Conclusions

Our report presents the first described case of the Pacinian corpuscle in a heterotopic location in the thymus. Similar to other incidental findings of this anatomic structure, explanation of its ectopic development, as well as its local function remains only speculative.

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