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Case report

Claws of cilia: further observation of ciliated epithelium in neurenteric cyst

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Summary. Claw-like projections and related structures on cilium tips in neurenteric cyst epithelium are described. The ciliary claws are about 20 nm in length and just beneath them four parallel electron-dense areas or lines are discernible. Similar structures were also observed in another case of neurenteric cyst. These structures are very similar to those reported previously, and it is suggested that they are commonly present in various ciliated epithelia in Man.

Key words: Neurenteric cyst – Enterogenous cyst – Cilia – Electron microscopy

Introduction

Ciliated cells of epithelia in Man and various animals have been studied for some time. In 1972, Dirksen and Satir reported a new and interesting finding, when they observed a crown of fine hairs, 22.5 nm long and 6 nm thick, on the tip of a cilium in the mouse oviduct. Although similar structures had been observed in various mammalian ciliated cells (Anderson and Hein 1977; Cordier 1975; Jeffery and Reid 1975; Kuhn and Engleman 1978), it is extremely rare in human material, only one having been reported (Foliguet and Puchelle 1986).

A neurenteric or enterogenous cyst is a benign epithelial cyst within or near the spinal canal, containing ciliated cells and goblet cells. They are believed to originate from endodermal tissue prior to its differentiation into oesophagus and trachea (Agnoli et al. 1984; Kinoshita and Tokuda 1973). We previously reported scanning and transmission electron microscopic findings of such a neurenteric cyst (Morita et al. 1990). Although we had observed claw-like projections on the tips of the cilia in this case, we did not publish the data because we assumed that it might be a common feature of a cilium. After a further search of the literature, we discovered that it was an extremely rare finding in human material and had never been reported in a neurenteric cyst. In this report, we describe the ultrastructural details of such claw-like projections in a human neurenteric cyst.

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Material and methods

A detailed case record was reported previously (Morita et al. 1990). Briefly, a 3-year-old boy suffered from an intradural extramedullary cystic mass from C3 to C7 vertebral levels. At surgery, a thin-walled, unilocular cyst ventral to the spinal cord was partially resected. The histopathological diagnosis was of a neurenteric cyst. He returned home symptom-free. Eight years and 3 months later, however, bilateral shoulder pain developed. He was admitted to our department again, and the wall of a large cyst, from C3 to C5, was again resected partially. He returned home without neurological deficit.

The cyst wall, obtained from the second operation, was immediately fixed with 4% glutaraldehyde in 0.1 *M* phosphate buffer, pH 7.4, overnight at 4° C and post-fixed with 1% osmium tetroxide in 0.1 *M* phosphate buffer, pH 7.4, for 90 min, at 4° C and then cut into pieces. They were dehydrated through graded ethanol and embedded in Epon 812. Ultra-thin sections were stained with uranyl acetate and lead citrate, and observed with a transmission electron microscope, JEOL JEM 200CX, at 80 kV. If indicated, the specimens were examined under appropriate tilting with a goniometer.

Results

The cyst epithelium was composed of ciliated cells, secretory cells, non-ciliated microvillous cells, squamous cells, and basal cells (Fig. 1 A). Their detailed ultrastructural findings have been reported previously (Morita et al. 1990). Among the ciliated cells, some of cilia had "claw-like projections" on their tips (Figs. 1 B, 2). These projections were about 20 nm in length and less than 12 nm in width, and sprouted directly from the outer layer of cilium membrane. Some projections appeared to exhibit equivocal periodicity perpendicular to their axes, as re-

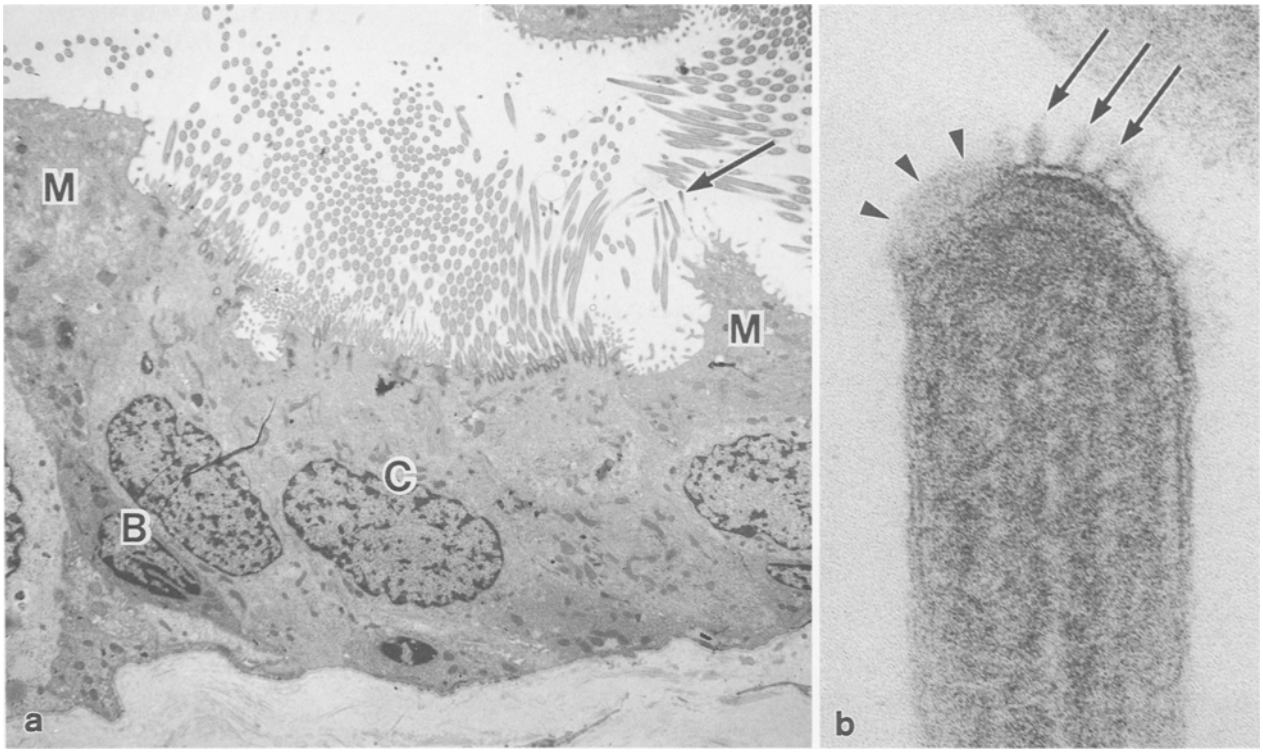

Fig. 1 A, B. Transmission electron microscopic view of cyst epithelium. **A** In this area, ciliated cells (*C*), non-ciliated microvillous cells (*M*), and basal cells (*B*) are observed. *Arrow* indicates the cilium tip demonstrated in B. ×4500. **B** Higher magnification view of a cilium tip with ciliary claws (*arrows*). *Arrowheads* indicate swelling of the ciliary membrane. Such swelling or ballooning of the ciliary membrane was commonly observed on the ciliary tip and shaft as well as bubbles between the cilia. ×250000

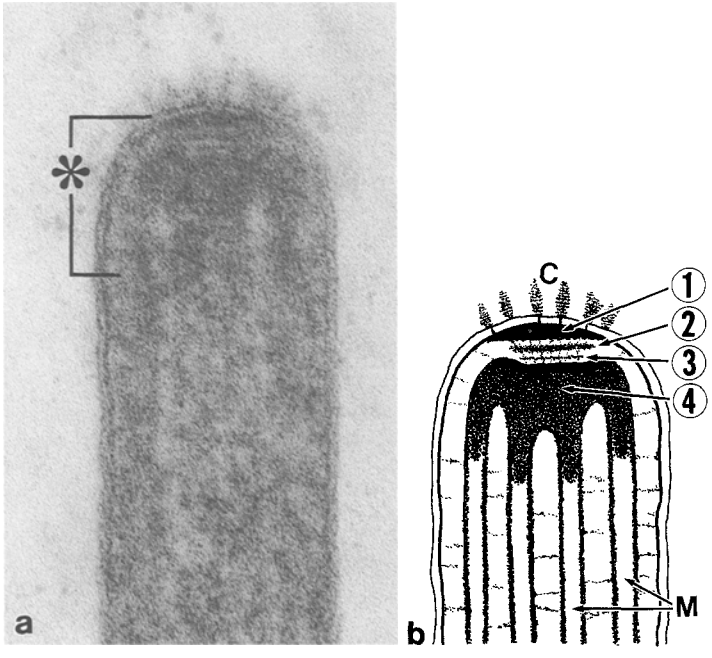

Fig. 2. A Another example of ciliary claws. Multilayered electron-dense areas or lines (*asterisk*) were discernible just beneath the ciliary claws. ×250000. **B** The schematic drawing of a cilium tip. ①, The most distal electron-dense plate which is attached to the innermost layer of cilium membrane; ②, the second electron-dense band; ③, the third electron-dense line; ④, the fourth electron-dense area, where the microtubules (*M*) terminate. The distal end of this area is centrally concave and more electron-dense. *C*, Ciliary claws. Modified from Foliguet and Puchelle (1986)

ported by Kuhn and Engleman (1978) and Foliguet and Puchelle (1986).

Beneath the area bearing these projections, four parallel electron-dense areas or lines were discernible (Figs. 1 B, 2). At the most distal end, an electron-dense plate, 8–10 nm in thickness, was attached to the innermost layer of cilium membrane. At 2–3 nm from this, an electron-dense band of 2–3 nm in thickness and a line of 1–2 nm in thickness was present. Further beneath these areas, there was an electron-dense area of 20–30 nm in thickness, the distal end of which exhibited some increase in density and occasionally showed central concavity. The distal ends of microtubules within the cilium increased in density and fused into the last electron-dense areas. The representative ultrastructure of a cilium tip with ciliary claws is illustrated in Fig. 2 B.

Discussion

A number of specialized structures are known to be present near cilia, one of which is "a crown" or "claw-like projections" on the apical aspect of the cilium tip. Such structures have been observed in various animals and organs, i.e. mouse oviduct (Dirksen and Satir 1972), rat trachea (Jeffery and Reid 1975), thymic cysts of nude mice (Cordier 1975), rabbit oviduct (Anderson and Hein 1977), and trachea and bronchioles of hamster, rat and rabbit (Kuhn and Engleman 1978). The only example in human materials was reported by Foliguet

and Puchelle (1986) in a study of bronchial mucosa. These previously reported structures closely resemble each other. Although their precise function and composition still remain unknown, it has been suggested that these structures facilitate the propelling and transport of mucus (Foliguet and Puchelle 1986; Jeffery and Reid 1975). It has been also reported that they are present only in the fully mature cilia and absent in the developing oviductal and respiratory cilia (Dirksen and Satir 1972; Kuhn and Engleman 1978).

The projections and electron-dense areas of a cilium tip in the present case were identical to those in the previous reports. To be precise, the most distal electron-dense plates in our case were relatively thick and attached to the ciliary membrane. In this respect, the ciliary claws and related structures in our case were very similar to those of mammalian cilia (Anderson and Hein 1977; Dirksen and Satir 1972; Kuhn and Engleman 1978) rather than human tracheal cilia (Foliguet and Puchelle 1986). The significance of this difference remains unknown.

Foliguet and Puchelle (1986) stated that ciliary claws were present in 48% of the cilium tips examined. Thus they were not infrequent. The precise incidence of their appearance in our case could not be determined because of the limited material, but it was not infrequent. We also examined specimens of another case of intraspinal neurenteric cyst. In spite of their initial fixation with formalin, we observed similar claw-like projections and related structures including the electron-dense areas attached to the ciliary membrane (unpublished data). Thus it is suggested that the ciliary claws are not rare in neurenteric cysts.

The neurenteric cyst is not normal tissue but arises as a result of maldevelopment. It might be considered to be more akin to normal tissue than pathologically altered metaplastic or neoplastic tissues. The data of Foliguet and Puchelle (1986) and our own suggest that the ciliary claws and related structures are also common in human material. Ciliated cells have been observed in various epithelia in Man, including respiratory tract, oviduct, developing oesophagus (Sakal et al. 1989), and metaplastic gastric mucosa (Torikata et al. 1989). However, ultrastructural differences have not been well described. The data from the previous mammalian animal studies suggest the possibility of the presence of the ciliary claws and related structures in many epithelia.

Why are they so rarely found in human material? It is possible that little attention has been paid to the tips of cilia. In the present study, some claws and related structures were detectable only under appropriate tilting of the specimen. It is also possible that claw-like projections have been missed due to the thickness of sections, as Foliguet and Puchelle (1986) pointed out. We presume that detailed examination of the electron-dense cilium tips under appropriate tilting, or of thick sections at high accelerating voltage (Foliguet and Puchelle 1986), may reveal claw-like projections more frequently.

It is also still uncertain whether ciliated cells in neurenteric cyst originate from respiratory ciliated cells or developing oesophageal ciliated cells. We favour the respiratory ciliated cell origin (Morita et al. 1990). The presence of ciliary claws in our cases is compatible with tracheal ciliated cells, but this is not necessarily definitive, since the possible occurrence of ciliary claws in the oesophageal epithelium of human fetuses exists. Further careful and detailed ultrastructural studies of various ciliated epithelia of Man are needed.

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