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Case Report

A case of thyroglossal duct cyst on the oral floor of a neonate

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ABSTRACT

Thyroglossal duct cysts are congenital cysts originating from thyroglossal duct remnants from the embryonic stage. Midline cervical cysts are the most common, while cysts in the oral cavity are rare. In the present report, we summarize our experience of a case of thyroglossal duct cyst in the oral floor in an 11-day-old infant; the cyst required enucleation because of feeding and respiratory difficulties in the infant. The patient presented with a protrusion in the oral floor at birth, and was admitted to the neonatal intensive care unit at our hospital at the age of 1 day because of being unable to feed orally. Based on the subsequent presentation of sleep apnoea, the patient underwent a detailed examination of the protrusion in the oral floor at our department where we performed enucleation of the cyst from the oral cavity at the age of 11 days. Following surgery, the patient exhibited functional suckling and the respiratory difficulties improved. To date, 1 year post-surgery, there has not been a recurrence of symptoms and progress is favourable.

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1. Introduction

Thyroglossal duct cysts are congenital cysts originating from the remnants of the thyroglossal glands formed during the embryonic state. They are histopathologically most commonly characterized as cyst walls lined by a ciliated columnar epithelium [1]. They commonly appear as a mass in the midline cervical area and rarely occur in the oral cavity [2,3]. A thyroglossal duct cyst is especially difficult to diagnose when it appears in the sublingual area or on the oral floor area because it is often difficult to differentiate from other cysts such as dermoid cysts, epidermoid cysts, ranula and cystic hygromas [4,5]. In the present report, we summarize our experience of a case of thyroglossal duct cyst in the oral floor of a neonate.

☆ Asian AOMS: Asian Association of Oral and Maxillofacial Surgeons; ASOMP: Asian Society of Oral and Maxillofacial Pathology; JSOP: Japanese Society of Oral Pathology; JSOMS: Japanese Society of Oral and Maxillofacial Surgeons; JSOM: Japanese Society of Oral Medicine; JAMI: Japanese Academy of Maxillofacial Implants.

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2. Case report

2.1. Clinical history

A 1-day-old female patient was admitted to our hospital because of a protrusion in the oral floor. There was nothing notable in her family history. The patient's mother did not present with any abnormalities during pregnancy. The patient was born at a gestational age of 39 weeks 1 day. Birth weight was 3.038 g, and the Apgar score was 9/10. Owing to the observation of a protrusion in the oral floor and feeding difficulty resulting from an inability to suck, the patient was fed by tube and was transported to the neonatal intensive care unit at our hospital at the age of 1 day. Because of the continued inability to feed orally following admission, tube feeding was performed. However, the patient presented with occasional sleep apnoea, as well as a temporary decline in percutaneous oxygen saturation to <80%. We therefore performed detailed examination of the protrusion in the oral floor.

2.2. Present illness

The extraoral findings showed no apparent swelling in the neck or submental region. The intraoral findings showed an elastic,



Fig. 1. Intraoral image at the initial examination. An elastic, well-defined protrusion is in the midline of the oral floor, and the tongue is elevated.

well-defined protrusion in the midline of the oral floor and elevation of the tongue (Fig. 1).

In the magnetic resonance imaging of the oral floor, the T1-weighted image demonstrated low signal intensity, while the T2-weighted images demonstrated a well-defined mass with a maximum diameter of 25 mm presenting with a uniform high signal intensity (Fig. 2). The clinical diagnosis was a cyst in the oral floor.

2.3. Treatment and progress

Based on consultation with a neonatologist, we planned an early surgery. At the age of 11 days, we enucleated the cyst from the oral cavity under general anaesthesia. We made a vertical incision directly above the lesion along the lingual frenulum, exposed the

cyst wall and bluntly detached the cyst from the surrounding tissue. Detachment was easy because of the absence of adhesion with the surrounding tissue, and no association was observed between the cyst and the sublingual gland or the submandibular gland. We also observed a fistula-like duct posterior to the lesion, and proceeded as far along the duct as possible before ligating and severing the duct, and then enucleating the lesion (Fig. 3). We then placed a Penrose drain in the wound and concluded the operation. Postoperative swelling on the oral floor was mild, and percutaneous oxygen saturation improved to 99–100%; therefore, we removed the Penrose drain on day 2 following surgery. We initiated suck training using an artificial nipple on day 3, and by day 7 the patient was able to feed orally at all times. In addition, the apnoea observed before surgery disappeared, and the patient was discharged with remission of symptoms on day 11. To date, 1 year post-surgery, no recurrence has been observed and the patient's progress is favourable.

2.4. Histopathological findings

The enucleated specimen was 24 mm × 25 mm × 25 mm in size with a smooth surface, and was filled with a milky, highly viscous fluid (Fig. 4). Histopathological findings showed that the cyst was covered alternately by a ciliated columnar epithelium and a mucus-producing high columnar epithelium. Mucus had accumulated in the lumen, while fibrous connective tissue was observed below the epithelium (Fig. 5). The final histopathological diagnosis was thyroglossal duct cyst.

3. Discussion

Thyroglossal duct cysts are congenital cysts originating from the remnants of the thyroglossal glands formed during the embryonic state. They are histopathologically most commonly characterized

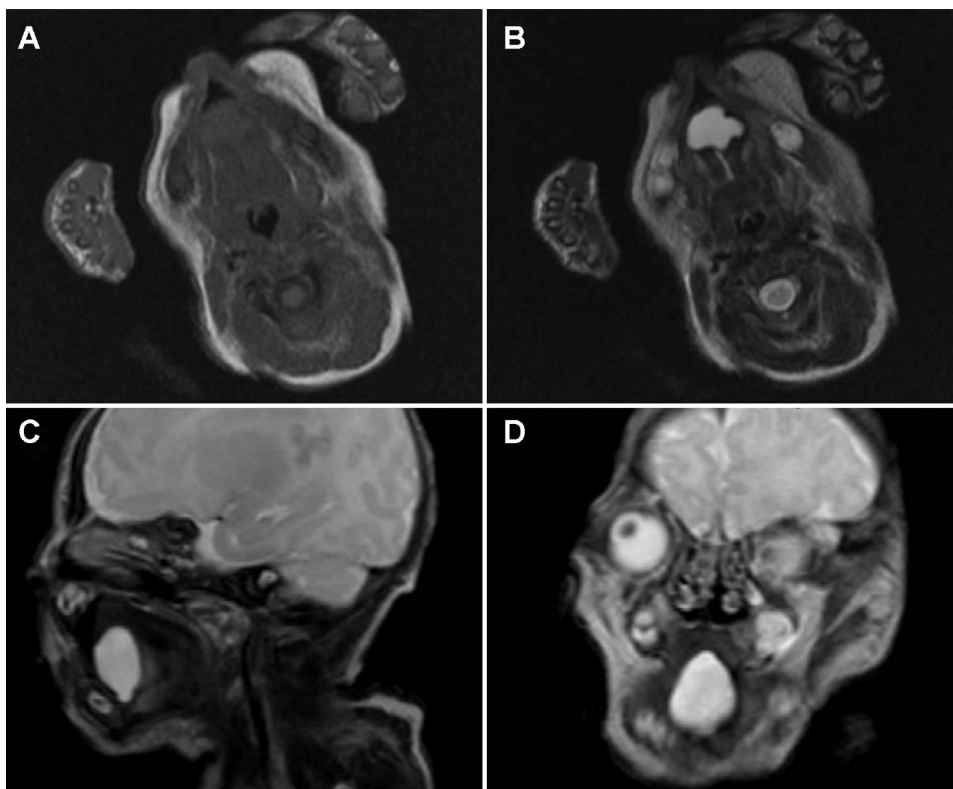


Fig. 2. Magnetic resonance images. (A) Horizontal section, T1-weighted image. (B) Horizontal section, T2-weighted image. (C) Coronal section, T2-weighted image. (D) Sagittal section, T2-weighted image. The T1-weighted image demonstrates a low signal intensity, while the T2-weighted images demonstrate a high signal intensity. A well-defined mass with a maximum diameter of 25 mm is observed.

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