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Clinical Record

Compound odontoma causing airway obstruction of the newborn: a case report

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Abstract
A newborn presented with acute airway obstruction secondary to a compound odontoma of the hard palate/nasal floor. This is the first case recorded in the literature. We review the pathology of compound odontomas and discuss the management of this rare and interesting case.

Key words: Odontoma; Palate, Hard; Airway Obstruction; Infant, Newborn

Introduction

Odontogenic tumours are a complex group of lesions with diverse histology and clinical behaviour. These tumours are very rare with an incidence estimated to be between 0.0002 per cent to 0.1 per cent.[1] However, the odontoma is one of the most common of the group, comprising approximately 70 per cent of all odontogenic tumours.[2–4]

Odontomas are classified as either compound or complex. Both types are composed of enamel, dentin, cementum, and pulp tissues. These odontogenic tumours can be found anywhere in the dental arches. The majority of compound odontomas are located in the anterior region of the maxilla or mandible and are usually diagnosed in the second decade of life.[3,5]

We present a newborn with acute airway obstruction resulting from a compound odontoma of the hard palate/nasal floor. We review the pathology of these rare tumours. We discuss the management of this unique case and discuss the differential diagnosis of airway obstruction in the newborn.

Case report

An aboriginal baby boy, delivered at term, developed significant airway obstruction shortly after birth. This improved with the insertion of a Guedel airway. The nasal passages could not be cannulated and bilateral choanal atresia was suspected.

He was transferred to a regional hospital and attempts were made to dilate the choanae and insert stents. Despite these measures, the infant continued to have airway difficulties and an oral airway was inserted. A computed tomography (CT) scan of the palate and nasal passages was performed. This revealed a lesion containing tooth-like structures occupying the floor of the right nasal cavity extending into the hard palate (Figure 1).

The patient was transferred to the Royal Children's Hospital in Brisbane, the nearest tertiary paediatric centre. It was decided that the lesion should be excised. In the operating theatre the lesion was found to involve most of the floor of the nasal passages and hard palate. Joint transnasal and transpalatal mobilization and total excision of the lesion was performed. The soft palate was resutured into position with a resultant hard palate defect.

The excised lesion was sent for histopathological analysis and diagnosis. The submitted specimen measured 31 mm × 23 mm × up to 15 mm in diameter. When the tissue was cut, several discrete hard areas were seen within tissue of a hard rubbery texture.

Microscopic examination showed several denticles recapitulating the structure of teeth (Figures 2a and 2b). These were pyramidal to bell-shaped arrangements with layers of epithelium and matrix overlying loose fibrous connective tissue similar to the dental papilla. The odontoblasts formed a single layer of tall pseudostratified columnar cells adjacent to a layer of dentin. A brightly eosinophilic layer of mineralized enamel separated the tall columnar ameloblasts from the dentin (Figure 3). A stellate reticulum covered some of the structures. A small amount of cementum, similar to bone, was present adjacent to one of the denticles. These hard tooth-like structures were surrounded by bone and loose fibrous stroma in which there were normal appearing mucoserous glands. Benign squamous epithelium covered the surface. There were no histological features to suggest a teratoma. The microscopic features were typical of a compound odontoma.

The patient made an excellent post-operative recovery only requiring a nasopharyngeal airway and oxygen for the first 24 hours. He was bottle fed using an obturator as used by cleft palate babies.

At twelve months, his cleft palate was repaired by the plastic surgical team. There has been no evidence of disease recurrence in that time.

Discussion

Odontomas are considered to be developmental anomalies resulting from the growth of completely differentiated epithelial and mesenchymal cells that give rise to ameloblasts and odontoblasts.[2,3] The aetiology of the odontoma is still unknown. Several theories have been proposed for the pathogenesis of this tumour, including trauma, infection, family history and genetic mutation.[3,5]

There are three recognized subcategories of odontomas: ameloblastic odontoma, complex odontoma and compound odontoma. The complex and compound odontomas represent the most common odontogenic tumours.

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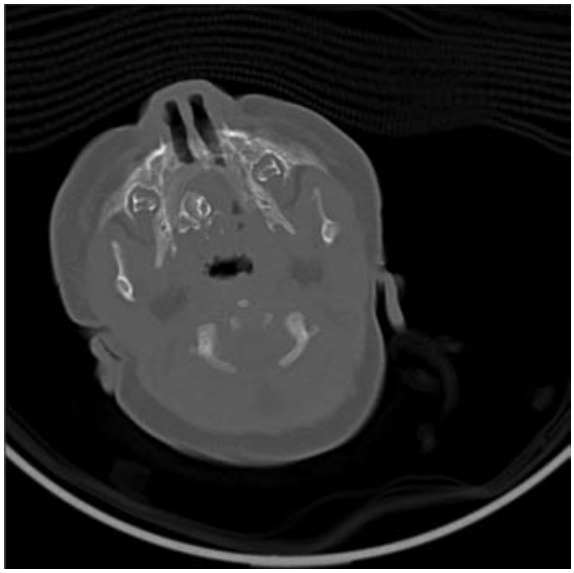

FIG. 1

Axial cut computed tomography scan showing a soft tissue density mass extending from the right nasal cavity into the nasopharynx. This mass appears to be causing some lateral bowing of the right lateral nasal wall and some displacement towards the left of the nasal septum. There are calcified densities within the mass which resemble teeth.

The complex and compound odontomas are non-aggressive lesions and usually remain quite small and asymptomatic.[3,6]

The classification by the World Health Organization defines the compound odontoma as a malformation in which all the dental tissue is represented in a more orderly pattern than in the complex odontoma.[7]

Compound odontomas are formed by tooth-like structures which resemble pulp tissue in the central portion surrounded by a dentin shell and partially covered by enamel. Histological examination often shows the presence of enamel matrix, dentin, pulp tissue and cementum.[3]

Odontomas are usually discovered on radiographs either incidentally or in search of a cause for an unerupted permanent tooth.[6] A presumptive diagnosis of compound odontoma is usually determined by radiographic appearance alone and is seldom confused with any other lesion. It usually presents as a radiopaque mass of multiple, small calcified structures with an anatomical similarity to normal teeth. These radiopaque toothlets often exhibit a thin, radiolucent rim around the periphery.[3,5,8]

Surgical excision is the recommended treatment by most practitioners. The compound odontoma is a well encapsulated lesion and easily enucleated from the surrounding bone with little probability of recurrence following its removal.[1–4,9]

Most compound odontomas are diagnosed in the second and third decades of life with equal sex distribution.[3,7,8,10] They are usually situated adjacent to teeth and roots of teeth. Compound odontomas commonly occur in the incisor-canine region of the maxilla.[1,3,6] The presence of a compound odontoma at birth and not in the vicinity of teeth is very rare. This case is unique as this is the first reported case of a compound odontoma presenting in a newborn and because of the variation from the usual location, with the presentation of life threatening airway obstruction.

As the newborn is an obligate nasal breather during the first few weeks of life, obstruction of the nasal cavity causes acute airway compromise. Structural defects such as severe nasal

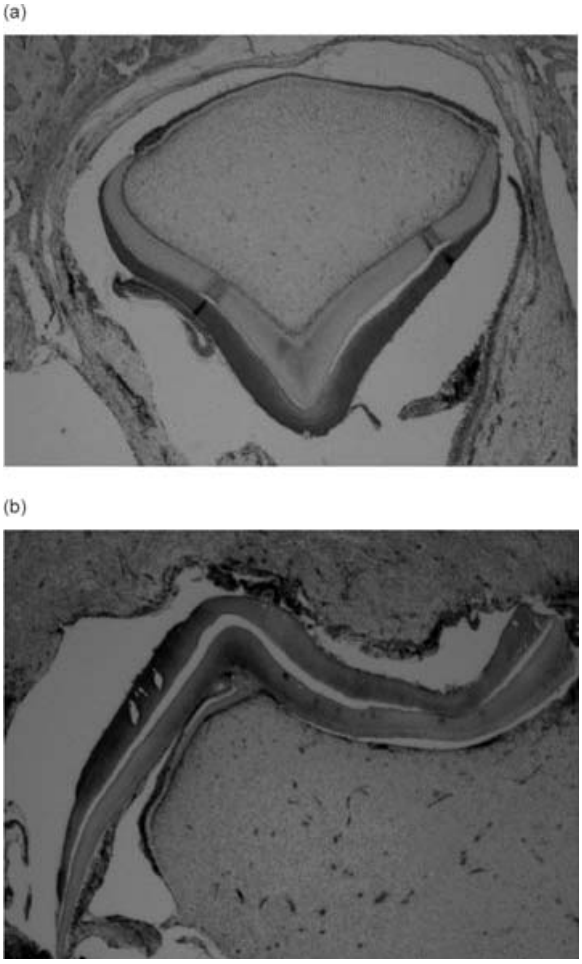

FIG. 2

Two denticles are shown (a) and (b). Although these resemble normal tooth structures, their shapes are simplified and they are smaller than normal teeth. Some artifactual clefting is present (H&E, ×20).

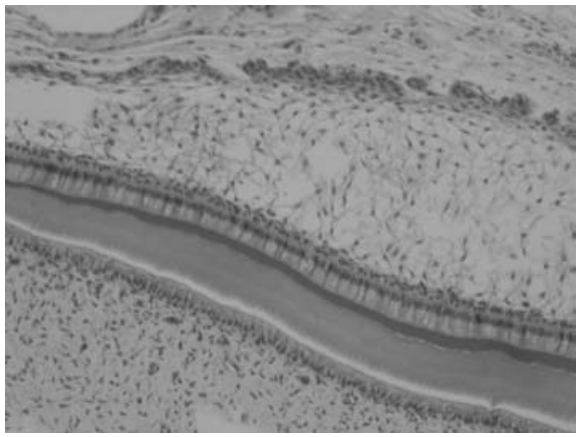

FIG. 3

Higher power view demonstrating dental pulp (lower left), covered by a layer of odontoblasts. A layer of dentin (light grey) and enamel (dark grey) separate the odontoblasts from the layer of ameloblasts. The loose connective tissue above the ameloblastic layer represents the stellate reticulum (H&E, ×20).

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septal deformities, affecting 6 per cent of babies at birth from injury/trauma during the birth process, choanal atresia, or upper respiratory infection, are the more common causes of neonatal respiratory impairment. Congenital tumours are unusual causes of acute nasal obstruction. These tumours include nasal dermoids, encephaloceles, meningoencephaloceles, chordomas, teratomas, craniopharyngiomas and nasoalveolar and nasopharyngeal (Tornwaldt's) cysts.

- **Odontomas are developmental anomalies resulting from the growth of completely differentiated epithelial and mesenchymal cells**
- **Odontomas are classified as benign odontogenic tumours and are subdivided into complex or compound odontomas morphologically**
- **Compound odontomas are rare and usually present in the second and third decades of life**
- **Compound odontomas are commonly situated in the incisor-canine region of the maxilla**
- **A significant variation in age of the patient and in the location of the tumour merits reporting of this case**

Complete or near-complete nasal obstruction presents in the first few minutes of life when the newborn's Apgar score drops and the child becomes cyanotic with the loss of placental oxygen exchange. Opening the baby's mouth to suction the airway results in successful resumption of breathing, only to have the respiration again stop when the baby's mouth closes. Most midwives would recognise the problem and insert a Guedel airway. Choanal atresia is generally the first differential diagnosis and the nasal airway is probed by a catheter to try to establish that diagnosis.

This child had the immediate presumptive diagnosis of choanal atresia when nasal catheters could not be passed into the newborn's nasopharynx. The compound odontoma was only discovered to be the true cause of the nasal obstruction when the CT of the facial bones was performed to determine the predominance of bony or membranous choanal atretic pathology.

The cause for acute nasal obstruction requires expeditious investigation. In such cases we recommend transfer to a specialist unit for investigation and treatment.

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