Autoamputation of Congenital Hairy Polyp in Neonate with Stridor and Respiratory Failure

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Autoamputation of Congenital Hairy Polyp in Neonate with Stridor and Respiratory Failure

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Abstract

Hairy polyp of the oronasopharynx is a rare developmental malformation. It is usually seen as a pedunculated tumor in the neonate. It is classified as a dermoid since it is derived from the ectoderm and mesoderm. We describe a neonate with a hairy polyp originating from the right lateral pharyngeal wall causing significant respiratory distress with cyanosis and stridor immediately after birth. Symptoms in the neonate disappeared following autoamputation of the mass. To our knowledge, this is the third case described in literature, with full recovery following autoamputation of a hairy polyp.

Introduction

Hairy polyps (HP) are rare congenital malformations of the oronasopharynx derived from two germinal layers, ectoderm and mesoderm. They are benign pedunculated masses with limited growth potential, seen predominantly female neonates. There is no documentation of malignant transformation. They typically present with polyhydramnios antenatally and shortly after birth with respiratory distress or feeding difficulty, depending on the size and location of the polyp. Here we describe a full term female neonate, presenting with respiratory distress and stridor immediately after birth due to pharyngeal HP, and a full recovery following autoamputation of the mass. To our knowledge, this is the third case whose symptoms resolved after autoamputation of the pharyngeal HP.

Case Report(s)

This female child, weighing 2.6 kg, was born to second degree consanguineous Pakistani parents, at 37 weeks of gestation, by Cesarian delivery without any complications in the intranatal period. The pregnancy was notable only for the increased amniotic fluid observed at the time of delivery. Antenatally there was no maternal illness, history of any drug ingestion, exposure to radiation or family history of any congenital anomalies. The first pregnancy had ended as Intrauterine Fetal Death at 26 weeks gestation. The APGAR scores were 8 and 9 at 1 and 5 minutes respectively.

Soon after delivery, the baby was noted to have grunting, suprasternal recession and cyanosis. While intubating, a mass was noted in the oral cavity making the intubation difficult. However the distress settled and the child became pink. It was then realized that the baby needed an oral airway to maintain normal saturation. A detailed examination of the mass was attempted; the mass appeared sausage shaped, pale non-tender, with hair noted on its surface. A CT scan was done which revealed a large heterogenous mass arising from the right lateral pharyngeal wall partly obliterating the oro-pharyngeal air column, measuring 28x12x16 mm, showing irregular dense calcification superiorly with adjacent fat density and hypodense cystic component inferiorly (Figure 2). There was no extension of the mass intracranially. A naso-gastric tube was inserted into the stomach and blood gas analysis and X-ray chest were done which was essentially normal. An ENT consultation was sought and a surgical excision of the mass was planned. The operation had to be postponed because the baby developed aspiration pneumonia for which she was started on antibiotics.

Meanwhile an MRI was done, which confirmed the CT findings. The periphery of heterogenous mass was hyperintense on T1 and T2 imaging; while the central region was hypointense, with no contrast enhancement. There was no extension to the nasal cavity or intracranially. The contour was well delineated from the anterior, inferior and posterior surfaces; features suggestive of a mature teratoma, probably a dermoid cyst.

The day before the surgery, the baby was noted to have repeated cough. A detailed oral cavity examination revealed a grey round shaped mass detached in the oral cavity. The surgeon examined her oral cavity and found no remant of the mass; so the operation was deferred. An X-ray was repeated to confirm the same.

Discussion

The HP are uncommon, benign, congenital malformations of the oronasopharynx. Arnold first
described the hairy polyp in 1870, and classified them into four categories, namely epignathi, teratomas, teratoids and dermoids (hairy polyps). According to his classification hairy polyps are the lowest order of teratoma, and also known as dermoids to describe their covering skin. There is a distinct female preponderance noted among the cases reported; however there is no evidence of any genetic inheritance. The embryogenesis of hairy polyp remains unclear with several theories being proposed. The first theory suggests that they represent early totipotent tissue remnants that consequently lost the organized growth pattern of normal tissue. Other authors suggest that they are midline ectopic remnants of the primitive streak (dermoid, teratoma). Heffner et al. proposed that hairy polyps were displacement of first or second branchial arch tissue. Approximately 10% cases are associated with first and second branchial arch malformations and a similar percent with cleft palate. There are reports of various congenital malformations associated such as cleft lip and palate, facial hemihypertrophy, agenesis of ear pinnae and uvula, left carotid artery agenesis and ankyloglossia which may explain the pathophysiology of the HP. HP most commonly arise from the nasopharynx, followed by the oropharynx, as noted by the oropharynx. Polyhydramnios is usually noted during pregnancy due to the polyp obstructing the fetal swallowing mechanism. Prenatal diagnosis did not have a significant impact on the diagnosis and treatment of these lesions. They typically present in the immediate postnatal period as an asymptomatic sausage shaped mass or with features of upper airway and pharyngeal obstruction. The airway obstruction symptoms include respiratory distress, stridor, which in some cases may be intermittent, and cyanosis. They may also present with feeding difficulty, drooling, repeated coughing and gagging. In addition, they may present with Eustachian tube dysfunction, hemoptysis, asphyxia and persistent nasal discharge. The present case had significant distress which settled following intubation and continued to remain distress-free even following extubation within a few hours with an oral airway kept in-situ. There was drooling of saliva which required regular oral suctioning; feeding was commenced via naso-gastric tube and the baby tolerated feeding.

HP might be overlooked in endotracheal intubation because the mass is usually mobile, soft and pedunculated. Budenz reported a case of pharyngeal hairy polyp obscured by physical examination and intubation, but diagnosed on brain MRI. We noticed the mass while intubating in this case. As the full extent of the mass could not be ascertained we proceeded for a CT scan and the size and extent of the mass was determined by the CT scan. The baby could be extubated and an airway was inserted which maintained the patency of the respiratory path and resolved the respiratory distress. However due to the presence of airway there was continuous drooling of saliva which required regular suctioning.

Radiological investigations are crucial to determine the extent of the mass, to rule out an intracranial extension, to differentiate it from other masses and to determine the presence of any other associated anomalies. Bony anomalies and mid-line defects are best identified by CT scanning; while intracranial extension and sagittal images are best demonstrated by MRI. In this case, the lesion was initially evaluated by CT scan, due to non-availability of MRI in our center, and later MRI was done outside for further evaluation. The differential diagnosis of a neonatal nasopharyngeal mass includes teratoma, meningoencephalocele, nasal glioma, neuroblastoma, haemangioma, thymic-thyroglossal or a lingual cyst. Due to the non-availability of the surgeon, the operation was initially postponed; feeding was commenced via naso-gastric tube. Unfortunately, the baby developed aspiration pneumonia leading to postponement of the surgery further.

Pedunculated HP can be excised by surgery from the pedicle base, while suture ligation and circumferential excision is carried out for sessile tumours. Surgical excision was planned but, the lesion had undergone spontaneous autoamputation. The autoamputation of the mass might be due to torsion of the mass resulting in ischaemic necrosis. This is the third reported case of autoamputation of hairy polyp with full recovery.

**Conclusion**

Oropharyngeal hairy polyps should be considered in any cases with polyhydramnios in the antenatal period with history of stridor and respiratory distress postnatally. Such masses need to be meticulously examined especially in its relation to intracranial structures. Significant morbidity and mortality can be avoided with early diagnosis and multidisciplinary management.

**References**


Illustrations

Illustration 1

Figure 1: X ray showing a heterogenous cystic mass in the oropharynx (arrow)

Illustration 2

Figure 2: Sagittal CT Scan showing oropharyngeal mass (arrow)
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