Congenital Alveolar Synechiae in a Neonate: Case Report and Review of Literature

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ABSTRACT

Aim: This case report highlights the management of congenital alveolar synechiae (CAS) in a 4-day-old neonate to alleviate the parent’s anxiety and associated problems.

Introduction: Congenital alveolar synechiae is a rare developmental anomaly found in neonates characterized by adherence of both the jaws by the fibrocollagenous bands. It will cause difficulty in feeding, has reduced mouth opening, and also affects the development of the jaws, if not excised at an early age.

Case report: A 4-day-old neonate reported with inability to open the mouth properly. A diagnosis of CAS was made and managed under inhalation general anesthesia.

Conclusion: Early management results in resolution of the feeding difficulty and adequate mouth opening. These cases should be managed as early as possible to prevent possible complications.

Keywords: Alveolar synechiae, Congenital defects, Electrocautery, Jaw development.


Source of support: Nil

Conflict of interest: None

INTRODUCTION

Congenital alveolar adhesion is a rare anomaly found in neonates. These fibrocollagenous adhesions are found in neonates at the time of birth. These adhesions are found in the anterior area attached to floor of mouth or in the posterior region between two jaws. They may be associated with the cleft palate or be present without a cleft.1 The various syndromes like Van Der Woude syndrome, oromandibular limb hypogenesis syndrome, micrognathia, and popliteal pterygium syndrome may be associated with CAS and has been found to be associated with 33 to 43% of the cases.2

These adhesions may cause problems like difficulty in feeding, effect on development of the jaws, etc. In long-standing cases, ankylosis of the temporomandibular joint (TMJ) may occur due to restriction of functional movements in the growing stage. In the literature, there is no classification that exists for the different pattern of alveolar synechiae. The fibrous adhesions have been found either in the posterior region of the mandible or in few case reports in the anterior region of the mandible.

In the present case, the fibrous adhesions present in both posterior and anterior regions of the jaws showed a different pattern of adhesions.

CASE REPORT

A 4-day-old baby girl was brought to the unit of Pedodontics and Preventive Dentistry by her parents with a complaint of inability to open the mouth properly with difficulty in breastfeeding since birth. The family history was noncontributory with nonconsanguineous parents. She had been delivered with normal vaginal delivery at 39 months of pregnancy with birth weight of 2.5 kg. The medical history was noncontributory. On examination, there were fibrous bands of the jaws with two fibrous bands in the right and left posterior regions and one in the anterior region. Mouth opening of 2 to 3 mm was present (Fig. 1). These cases have usually been reported to be associated with cleft palate. A radiographic examination was done to rule this out and also a possibility of a bony fusion between the jaws and any other facial deformity. Computed tomography (CT) examination revealed cleft of secondary palate in the present case (Fig. 2).

The fibrous bands were cauterized with electrocautery under inhalational anesthesia using facemask ventilation (Fig. 3). Lignocaine 1:200,000 was administered before cauterization to prevent postoperative pain. Intraoral examination was done to check for any other associated cleft. Only cleft of secondary palate was evident, so the child was referred to plastic surgery consultation. The baby was breastfed after 12 hours with no complications, and mouth opening was increased to 20 to 25 mm postoperatively (Fig. 4).
DISCUSSION

Isolated CAS is a rare anomaly found in neonates. There are different patterns of fibrous bands present between the jaws. The bands can be of membranes of epithelium, collagenous bands, or other connective tissue like muscles or bone. It may or may not be associated with cleft palate (cleft palate lateral synechiae syndrome) or other syndromes like popliteal pterygium syndrome, Van Der Woude syndrome, oromandibular limb hypogenesis syndrome, oral facial digital syndrome, microglossia, or micrognathia. In the present case, no other facial cleft or syndromes were associated. Gartlan et al\textsuperscript{3} reviewed 50 cases of alveolar synechiae, but only 7 cases of isolated CAS were present and rest of the cases were associated with cleft palate or other syndromes. The rare cases congenital alveolar synechiae without the cleft palate are reported in the literature are presented.\textsuperscript{4-13} (Table 1)

The exact etiology of CAS is unknown, but several hypothesis have been proposed due to interposition of the tongue between the palatal shelves with close contact between the palate and the floor of the mouth predisposing to the formation of intraoral synechiae, remnants of buccopharyngeal membrane, teratogenic drugs like meclizine, alteration in the growth factors interaction, localized ischemia, trauma in late pregnancy, and pressure exerted by amniotic bands on the first branchial arch and these are found to be considered for these synechiae.\textsuperscript{4}
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The management includes excision of the fibrous bands at an early age to allow feeding and development of the TMJ and the orofacial region. Laser, surgical scissors, surgical blade, or bipolar diathermy were used in the literature to excise the bands. Sheets, silastic bite blocks, gauze, and rubber can be used as an interpositional splint material for mouth opening to prevent secondary adhesions of raw areas of the jaws. In the present case, electrocautery was used to cauterize the fibrous bands under inhalational anesthesia and after recovery from the anesthesia, the baby was breastfed without any difficulty in suckling. The mouth opening of the patient was increased to 25 mm postoperatively.

The CAS is a rare occurrence in the neonate causing difficulty in feeding and restriction of mouth opening affecting growth of the TMJ and jaws. Early management of the synechiae helps in feeding and development of the TMJ and orofacial structures.

Table 1: Cases reported in the literature with isolated CAS

<table>
<thead>
<tr>
<th>Sl. no.</th>
<th>Author, year</th>
<th>Age/sex</th>
<th>Type of fibrous bands</th>
<th>Associated facial anomaly/cleft palate</th>
<th>Associated syndromes</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>Szelely, 1941</td>
<td>Unknown</td>
<td>–</td>
<td>Absent</td>
<td>Absent</td>
</tr>
<tr>
<td>2</td>
<td>Randall, 1984</td>
<td>Unknown</td>
<td>–</td>
<td>Absent</td>
<td>Absent</td>
</tr>
<tr>
<td>3</td>
<td>Goodacre and Wallace, 1990</td>
<td>Unknown</td>
<td>–</td>
<td>Absent</td>
<td>Absent</td>
</tr>
<tr>
<td>4</td>
<td>Haydar et al, 2003</td>
<td>8-day-old baby girl</td>
<td>Anterior and posterior alveolar bands</td>
<td>Absent</td>
<td>Absent</td>
</tr>
<tr>
<td>5</td>
<td>Tanrikulu et al, 2005</td>
<td>10-month-old baby girl</td>
<td>Not known</td>
<td>Absent</td>
<td>Absent</td>
</tr>
<tr>
<td>6</td>
<td>Mortazavi and Motamedi, 2007</td>
<td>2-month-old baby girl</td>
<td>–</td>
<td>Absent</td>
<td>Absent</td>
</tr>
<tr>
<td>7</td>
<td>Chiabi et al, 2008</td>
<td>14-day-old baby girl</td>
<td>Complete fibrous synechiae on the gum pads extending to the TMJs</td>
<td>Absent</td>
<td>Absent</td>
</tr>
<tr>
<td>8</td>
<td>Bozdag et al, 2011</td>
<td>1-day-old baby girl</td>
<td>Between the upper and lower gum pads from the right side to nearly half of the left side of the mouth</td>
<td>Absent</td>
<td>Absent</td>
</tr>
<tr>
<td>9</td>
<td>Bali et al, 2010</td>
<td>20-day-old baby boy</td>
<td>Bilateral posterior bands</td>
<td>Absent</td>
<td>Absent</td>
</tr>
<tr>
<td>10</td>
<td>Cerrati et al, 2015</td>
<td>one-day-old baby boy</td>
<td>Bilateral posterior bands</td>
<td>Absent</td>
<td>Absent</td>
</tr>
<tr>
<td>11</td>
<td>Present case</td>
<td>4-day-old baby girl</td>
<td>Anterior and posterior alveolar bands</td>
<td>Absent</td>
<td>Cleft of secondary palate was seen</td>
</tr>
</tbody>
</table>
WHAT THIS ARTICLE ADDS?
This article reports the rare case of isolated CAS and its management in a neonate as anesthesia in neonates is a challenging task.

REFERENCES