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RESEARCH ARTICLE

FEEDING APPLIANCE FABRICATION FOR A COLPOCEPHALY ASSOCIATED WITH CLEFT PALATE CHILD: A RARE CASE REPORT

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ABSTRACT

Colpocephaly is an abnormal enlargement of occipital horn of lateral ventricle associated with several abnormalities. Various etiologies have been reported including intrauterine/ perinatal injuries, genetic disorder and an error of morphogenesis. Here a case of colpocephaly associated with cleft palate and hypoplastic corpus callosum is reported. A feeding appliance covering the palatal defect is fabricated.

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INTRODUCTION

Abnormal enlargement of occipital horn of lateral ventricle is defined as colpocephaly, which is associated with normal frontal horns. Benda (1940) first indentified the "failure of decrease in the size of the primitive brain vesicles" in a mentally retarded boy with microcephaly, epilepcy,abscent corpus callosum, micro- and macrogyria, heteropic grey matter and he named this syndrome as vesiculocephaly (Benda, 1940). Yakovlev and Wadsworth (1946) coined the term colpocephalyand described it as a persistence of fetal cerebral ventricles in postnatal life (Yakovlev, 1946). Greek word 'kolpos' means hollow.

Case report: One and half year old boy came to the department of pedodontiafor feeding plate fabrication).Clinical findings were observed like, microcephaly, protruded premaxilla (Fig.1), complete cleft involving both hard and soft palate (Fig.2). Family history was not significant and no history of seizure. Level of glucose (fasting), urea (blood), creatinine (serum) was normal in report.

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The magnetic resonance imaging of brain demonstrated dilatation of left occipital horn of lateral ventricle, hypoplastic corpus callosoum, hypoxic-ischemic encephalopathy involving both temporo-parietal and left occipital lobar cortex, extra-axial cerebro spinal fluid spaces, widened cortical sulci with loss of volume of subcortical white matter. No abnormal findings were obtained in sleep electroencephalogram and chest radiograph. After proper examination, primary impression (Fig.3) was taken by impression compound for special tray fabrication. Special tray (Fig.4) was prepared by self cure acrylic resin and final impression (Fig.5) was taken by elastomeric impression material. Final model (Fig.6) was made by blocking all undercuts. After that, feeding plate (Fig.7) was fabricated by autopolymerizing resin and properly polished to avoid injury to oral mucosa. Finally, feeding plate was inserted into patient's mouth by using customized headgear (Fig.8). The headgear was used to prevent accidental swallowing of the appliance. The patient was feed comfortably after inserting the appliance (Fig.9). Proper instruction was given to the parents about insertion, removal and cleaning of the appliance. A regular follow up was carried out after 24 hours, 1 week and 1 month.

DISCUSSION

Since identifying the first (1940) case approximately 50 cases have been reported (Seyfettin *et al*., 2012).

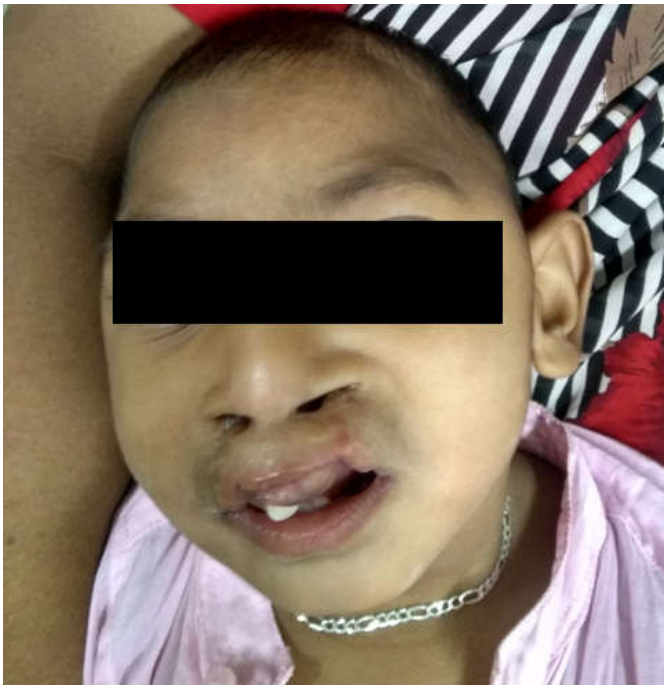

Fig.1. Showing microcephaly and protruded pre maxilla

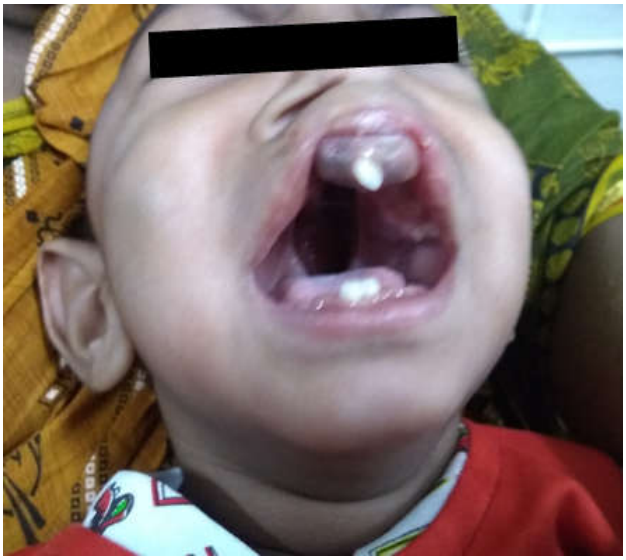

Fig.2. Showing cleft involving both soft palate and hard palate

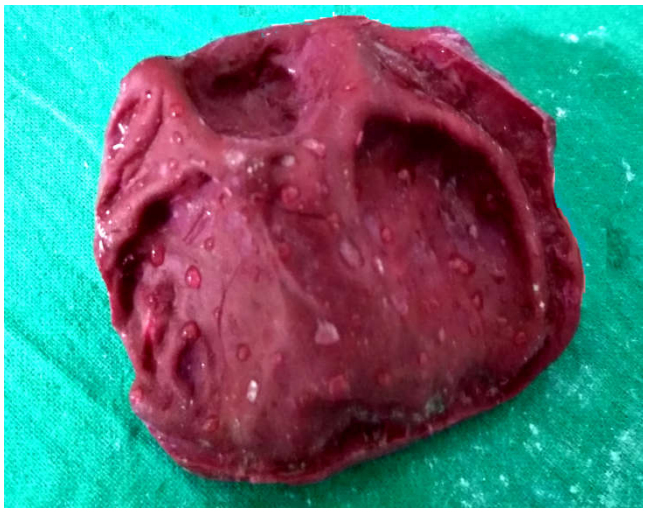

Fig. 3. Showing primary impression

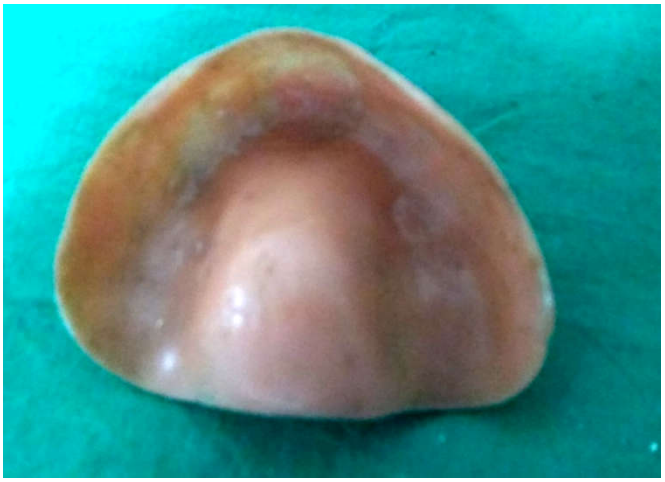

Fig.4. Showing special tray

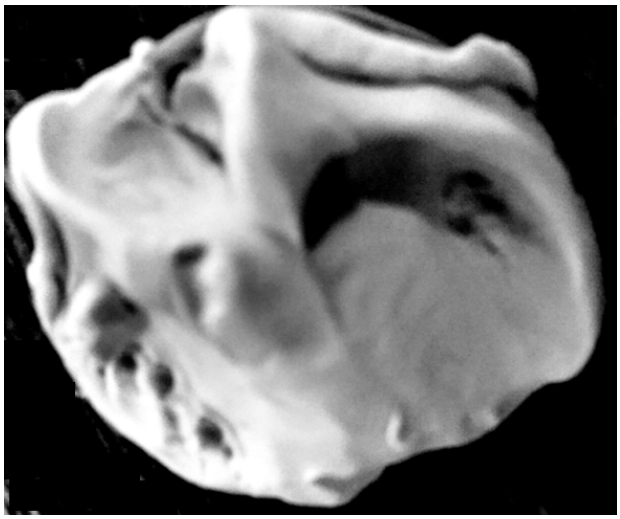

Fig.5. Showing final impression

Fig.6. Showing master cast

Colpocephaly is not a specific congenital malformation of central nervous system as suggested by various authors.It is a multiple disorder and occurs due to different etiology including a) intrauterine infection like toxoplasmosis b) perinatal ischemic encephalopathy c) maternal drug ingestion during early pregnancy like corticosteroid, theophyline and salbutamol (Surasak Puvabanditsin *et al*., ?).

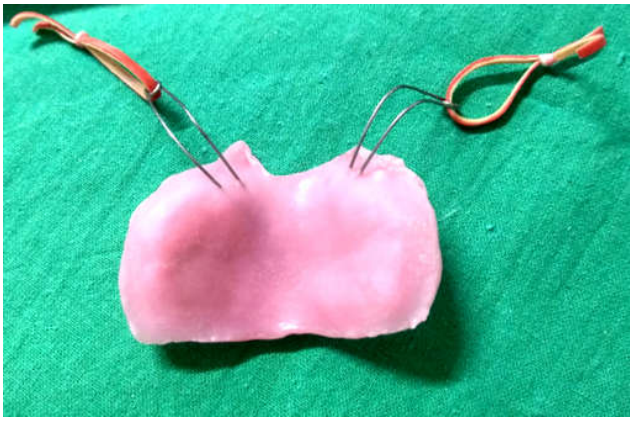

Fig.7. Showing feeding plate

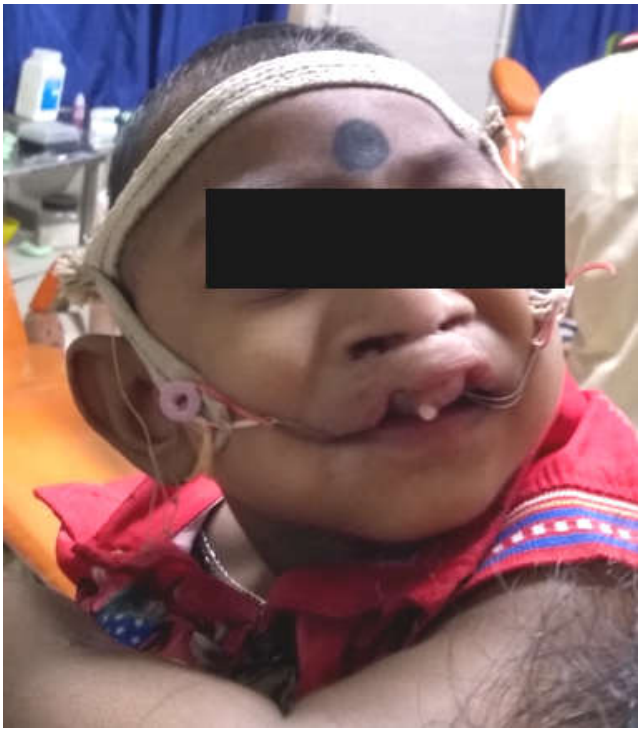

Fig.8. Appliance placed in to the patient's mouth with customized headgear

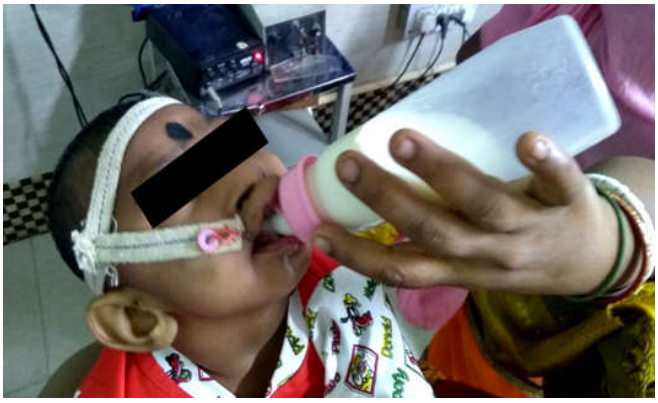

Fig.9. Patient successfully feed after inserting the appliance

Three cases of familial occurrences of colpocephaly have been reported (Herskowitz et al., 1985). Some cases were identified based on genetic origin with an autosomal or X-linked recessive. Ventricular enlargement of colpocephaly was caused due to developmental arresting of white matter between middle of second to fifth month of fetal life (Cerullo *et al*., 2000).

An embryological mechanism was proposed that lateral ventricle arise as large cavities of telencephalic vesicle. Normally ventricular size decreases after formation of foramen Magendie which decompresses the ventricles. The unknown causes may interfere with this process and express abnormal enlargement of occipital horn. Periventricular leukomalacia may cause abnormally enlarged occipital horn. Radiological investigations are commonly used to identify Colpocephaly. It may be present isolated or may be associated with multiple conditions likelinear nevus sebaceous syndrome, lissencephaly type 1Marden-Walker syndrome, tourette syndrome, Aicardi syndrome, trisomy 8 mosaic, trisomy 9 mosaic, Norman-Roberts syndrome, hemimegalencephaly Zellweger syndrome, Nijmegen breakage syndrome, Chudley-McCullough syndrome (Garg, 1982). Colpocephaly most commonly confused with hydrocephalus and arachnoid cyst. Diagnosis is important because increased CSF pressure causes hydrocephalus and it requires surgery (shunting).

No treatment requires in asymptomatic but surgery should be done in symptomatic arachnoid cystic lesions. The clinical findings of colpocephaly are affected with different etiology and associated conditions. Most common central nervous system malformations associated with colpocephaly are agenesis of corpus callosum; neuronal migration disorders (lissencephaly, pachygyria), macrogyria, microgyria, schizencephaly, enlargement of cisterna magna, optic nerve hypoplasia, meningomyelocele (Seyfettin, 2012) and cleft palate. Other associated anomalies included micrognathia, hypoplastic nails, simian creases, Pieree-Robin syndrome and neurofibromatosis (Noorani, 1988). Colpocephaly is usually seen bilaterally but unilaterally cases also have been found. Unilateral colpocephaly is an important finding in hemimegalencephaly with enlarged hemisphere[9]. An uncommon form of familial procencephaly is another cause of unilateral colpocephaly (Aguglia *et al*., 2004). A very rare case of colpocephaly associated with macrocephaly is described in literature. Increased intra cranial pressure was found in this case and Ventriculo-peritoneal shunts were used to drain the fluid into peritoneum (Patnaik *et al*., 2012). Reported colpocephaly commonly associated with mental retardation, seizure, motor and visual abnormalities. In literature approximately thirty six cases have been reported and three out of these reporting caseshad completely normal neurological and motor development[4].Stem cell therapy is considered a very promising treatment for colpocephaly. Oligodentroglial cells can be used to produce myelin and alleviate the symptoms of colpocephaly.

Conclusion

Usually colpocephaly is nonfatal. There is no known definitive treatment for colpocephaly but specific treatment depends on associated conditions. Anticonvulsant medication can prevent seizure complications. Physical therapy is used to prevent contractures in patients with limited mobility. Prognosis depends on associated conditions and degree of abnormal brain development. In our case, patient has a large cleft involving both hard palate and soft palate. Feeding plate not only helps in feeding but it also helps in proper positioning of tongue away from palate and it promotes growth of palatal shelves towards each others. In our case, feeding was done comfortably and improvement of body weight was seen after wearing feeding plate.

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