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Case Report (Pages: 5195-5198)

First Report of A Unique Presentation of Hypertrophic Pyloric Stenosis Following Type I Esophageal Atresia; A Case Report

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Abstract

Combination of congenital esophageal atresia and subsequent hypertrophic pyloric stenosis is a rare condition which occurs in early infancy. The underlying etiology and pathophysiology of this association still remains unclear. In this paper we report a unique case of hypertrophic pyloric stenosis, for the first time, which occurred in an infant who underwent surgery for type I esophageal atresia. Therefore, we intend to highlight the role of poor nutrition intake in the incidence of this condition.

Key Words: Esophageal Atresia, Hypertrophic pyloric stenosis, Infant, Nutrition.

*Please cite this article as: Salimi A, Iranikhah A, Rashidi Nia Sh, Eftekhari SS, Shahmoradi S. First Report of A Unique Presentation of Hypertrophic Pyloric Stenosis Following Type I Esophageal Atresia; A Case Report. Int J Pediatr 2017; 5(6): 5195-5198. DOI: **10.22038/ijp.2017.23442.1972**

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Received date: Mar.24, 2017; Accepted date: Apr.22, 2017

1- INTRODUCTION

Congenital esophageal atresia (EA) takes account the most common gastrointestinal atresia (1) with an incidence rate of one in 2,500-4,000 live births (2). Isolated EA is a serious subset of EA cases which usually presents as long-gap type (3) and occurs approximately in 1 out of 40,000 live births (4). Infantile hypertrophic pyloric stenosis is another gastrointestinal anomaly which accounts the most common cause of obstruction within the early months of life with the incidence rate of about two cases per 1000 live births (5).

Nevertheless, the coincidence of these two conditions has been challenged in the literature and the incidence rate of 1-10% is reported (2, 6). Notwithstanding the condition is well established in the literature, no consensus is raised so far, regarding the underlying pathophysiology. In this study, for the first time, we aimed to report an outstanding association of hypertrophic pyloric stenosis following surgical repair of type I esophageal atresia.

2- CASE REPORT

A 53 days, 2,700 grams boy was born in our university hospital of Hazrat Masumeh in Qom province of Iran through cesarean delivery at term. Upon birth, the diagnosis of esophageal atresia was raised following physical examinations and radiological evaluation. The patient, then, underwent surgery and a type I esophageal atresia according to Ladd's classification (7) was confirmed with no other concomitant anomalies. Eventually a feeding gastrostomy and cervical esophagostomy were provided and the patient has discharged with a planned dietary regimen of mixed formula and breast milk after he got stable and ensuring an acceptable food passage.

Fifty-two days later, the infant was readmitted to our hospital with a complaint of milk return from gastrostomy (**Figure.1**). The parents declared that this condition had been progressively started a few days earlier and the son became lethargic. With suspicion of hypertrophic pyloric stenosis, the ultrasonographic evaluation, was affirming (**Figure.2**), and the surgery confirmed the diagnosis, for which an open pyloromyotomy was carried out (**Figure.3**). The post-operative course was uneventful and the infant tolerated feeding within the first 24 hours.

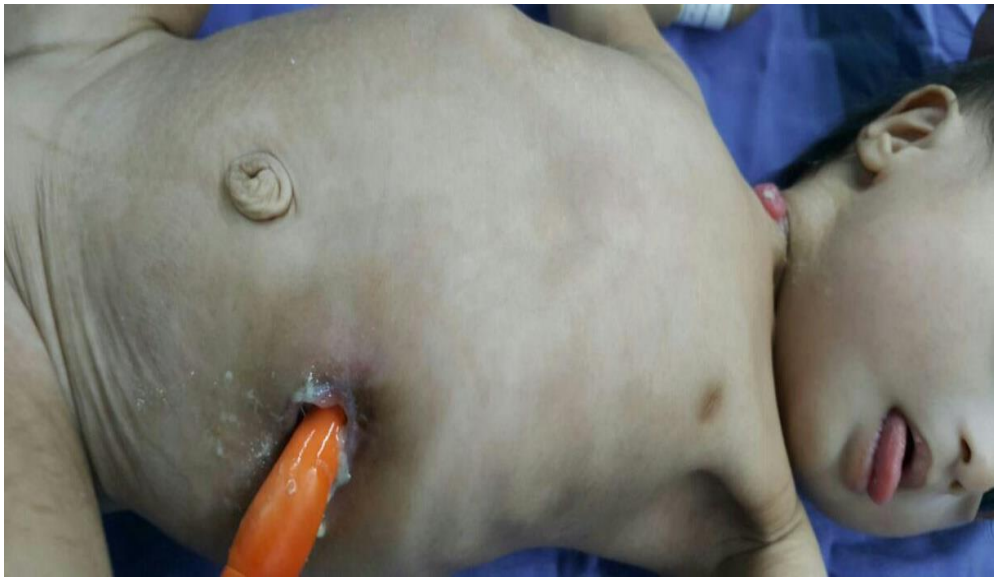

Fig.1: Return of stomach contents through the gastrostomy tube.

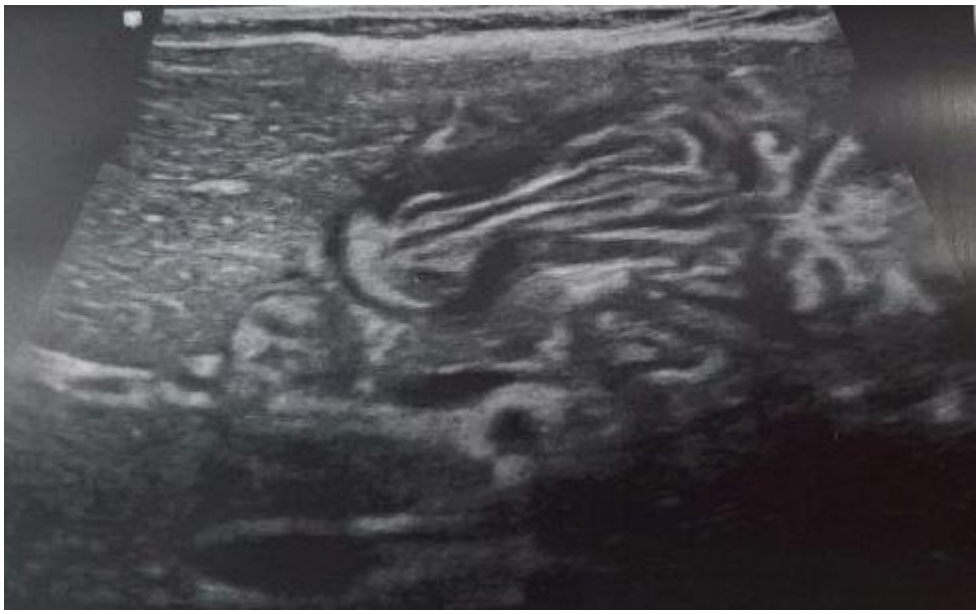

Fig.2: Ultrasonographic result, indicative of hypertrophic pyloric stenosis.

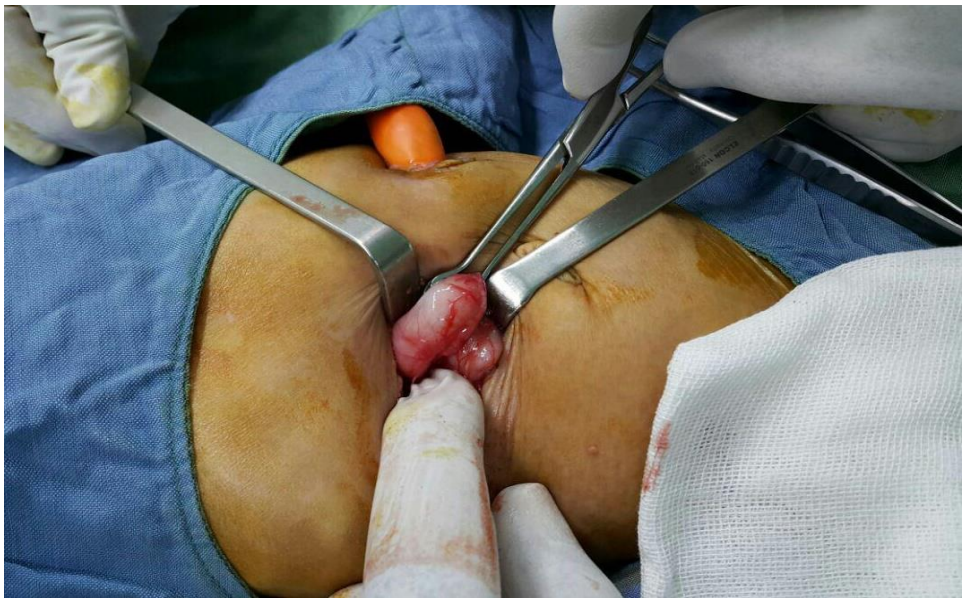

Fig.3: Intra-operative photograph demonstrating the hypertrophied pyloric muscle.

4- DISCUSSION

Infantile hypertrophic pyloric stenosis is a common surgical condition in early infancy, in which various etiological factors are involving and there is no real consensus in this regard (5). Observational findings, nevertheless, are in favor of metabolic disturbances and even those of potentially occur within early infancy (8, 9). In terms of the association between esophageal atresia and subsequent hypertrophic pyloric stenosis, the interesting thing for us is the post-operative course of these patients. All previously reported cases of this association are mentioned the occurrence of Infantile Hypertrophic Pyloric Stenosis

(IHPS) following a while as the treatment course of EA (10-14). So it appears that a period of insufficient nutrition intake in early infancy would be leading cause of IHPS through potential metabolic disturbances. However, undiagnosed pyloric stenosis as a concomitant disorder with EA is supposed in the literature (14).

5- CONCLUSION

In this paper we reported a unique combination of esophageal atresia with hypertrophic pyloric stenosis. To the best of our knowledge, this is the first report in this context, in which the pyloric stenosis occurred following a type I esophageal atresia and well presents the impression of insufficient nutrition intake as an underlying etiology of this condition. Nonetheless, further investigations as cohort studies with ample sample size are strongly recommended.

6- CONFLICT OF INTEREST: None.

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