



## A case of seckel syndrome with human bocavirus infection and respiratory failure

### Human bocavirüs enfeksiyonu olan ve solunum yetmezliği gelişen seckel sendromu olgusu

Seher Erdogan, Arzu Oto, Mehmet Bosnak

Department of Pediatric Critical Care, Gaziantep University School of Medicine, Gaziantep, Turkey

#### Abstract

Human bocavirus (HBoV) is a newly described member of the family parvovirus. We report an 18 -month-old male patient under observation with a diagnosis of Seckel syndrome and presenting to our emergency department with respiratory distress was admitted to the pediatric intensive care unit. Oxygen saturation with non-rebreather mask was 93%. The patient was intubated due to respiratory insufficiency and respiration on a mechanical ventilator. HBoV was isolated in a nasopharyngeal specimen using PCR. Antimicrobial and antiviral therapy was started. The patient's blood pressure fell, dopamine infusion was started. Hypotension persisted, and noradrenalin was added to treatment. Hypotension persisted, and the patient died on the 4<sup>th</sup> day of hospitalization. The purpose of this case report is to emphasize the need to remember HBoV infections in cases of respiratory failure, particularly under the age of 5, and that although the condition is rare in the literature, intensive care may still be required.

**Keywords:** Seckel Syndrome; Human Bocavirus; Child.

#### Öz

Human Bocavirüs(HBoV), Parvovirüs ailesinin yeni bir üyesidir. Burada, Seckel sendromu tanısı ile izlenen, solunum sıkıntısı nedeniyle acil servise başvuran ve çocuk yoğun bakım ünitesine yatırılan 18 aylık erkek hasta sunulmuştur. Oksijen saturasyonu ,rezervuarlı geri solunum maske ile %93'tü. Solunum yetmezliği nedeniyle hasta entübe edilerek mekanik ventilatörde solutuldu. Nazofarıngeal aspirat örneğinde PCR yöntemi ile HBoV izole edildi. Antimikrobiyal ve antiviral tedavi başlandı. İzlemede hastanın kan basıncı düştü, dopamin infüzyonu başlandı. Hipotansiyonun devam etmesi üzerine tedaviye noradrenalin eklendi. Kan basıncı düşüklüğü devam eden hasta yatışının 4. gününde kaybedildi.

Bu olguyu sunmaktaki amacımız; özellikle 5 yaşın altında solunum yetmezliği olan hastalarda Human Bocavirüs enfeksiyonlarının da hatırlanması gerektiğini, literatürde nadir bir durum olarak belirtilmesine karşın, yoğun bakım ihtiyacının olabileceğini vurgulamaktır.

**Anahtar Kelimeler:** Seckel Sendromu; Human Bocavirüs; Çocuk.

Received/Başvuru: 17.11.2015  
Accepted/Kabul: 14.12.2015

#### Correspondence/İletişim

Seher Erdogan  
Department of Pediatric Critical  
Care, Gaziantep University School of  
Medicine, Gaziantep, Turkey  
E-mail: seher70@gmail.com

#### How to cite this article/Atıf için

Erdogan S, Oto A, Bosnak M.  
A case of seckel syndrome with  
human bocavirus infection and  
respiratory failure. J Turgut Ozal  
Med Cent 2016;23(4):439-42.

## INTRODUCTION

Human bocavirus (HBoV), a newly described member of the family parvovirus, was first isolated from nasopharyngeal specimens from children with lower airway infection in Sweden in 2005 (1). HBoV activity persists throughout the year, with no seasonal characteristics, and is seen at higher levels in males. It is more common in children aged under 2 years hospitalized with diagnoses of bronchiolitis and asthma attack. Other clinical symptoms seen in patients include cough, fever, sore throat, rhinorrhea, headache and nausea (2). Seckel syndrome is an extremely rare inherited disorder characterized by growth delays prior the birth resulting in low birth weight. Growth delays continue after birth, resulting in short stature (3).

## CASE REPORT

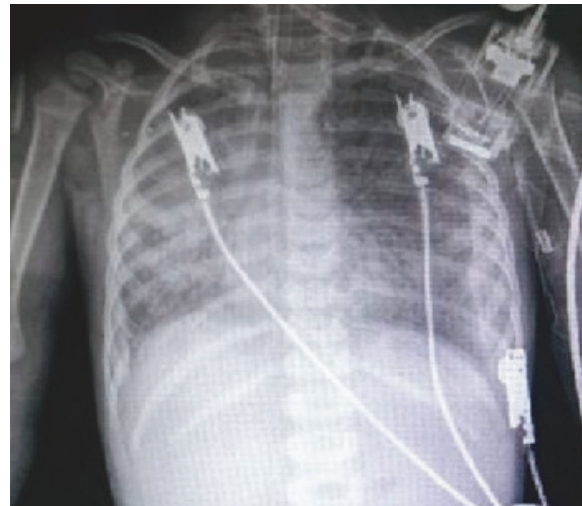
An 18-month-old male patient under observation with a diagnosis of Seckel syndrome and presenting to our emergency department with fever, respiratory distress and cough was admitted to the pediatric intensive care unit with informed consent from the family. The symptoms had commenced 2 days previously. The patient had been started on oral antibiotic and bronchodilator therapy at the health center to which he had originally presented, but the symptoms had persisted. He had been born by normal spontaneous vaginal delivery, on term, weighing 2600 gr, and had received mother's milk and supplements. Second-degree consanguinity was present between the parents. His three other siblings were healthy.

He had begun sitting up unsupported 1 month previously, but could not yet speak. At physical examination, body weight was 8.5 kg (<3 p), height 73 cm (<3 p) and head circumference 43 cm (<3 p). His general condition was average, and he was conscious but restless.

An appearance involving a narrow forehead, large eyes, a beak-like nose, a high palate and micrognathia was present. His body temperature was 37.9 °C, respiration rate 50/min, heart rate 155/min and blood pressure 105/55 mmHg. Inter-, sub- and supracostal contractions were present. Coarse rales and rhonchi were heard in both lungs at auscultation. 2/6 systolic murmur was present in the mesocardium. Oxygen saturation with non-rebreather mask with a 5 lt/min flow reservoir was 93%. At laboratory examination, his hemoglobin level was 11.2 gr/dl, white cell count 19.400/mm<sup>3</sup>, platelet 236.000/mm<sup>3</sup> and C-reactive protein 12.3 mg/dl. Blood gas analysis results were pH 7.04, PaCO<sub>2</sub> 72.4 mmHg, PaO<sub>2</sub> 32.9 mmHg, SaO<sub>2</sub> 54.1% and HCO<sub>3</sub> 19.2 mmol/lt. Areas of peribronchial and interstitial infiltration were present at pulmonary radiology (Figure 1).

The patient was intubated due to respiratory insufficiency and respirated on a mechanical ventilator in synchronized intermittent mandatory ventilation-pressure support mode (SIMV-PS). The mechanical ventilator parameters were set at PIP: 22 cm H<sub>2</sub>O,

PEEP:5 cm H<sub>2</sub>O, F: 30/min and FiO<sub>2</sub> :0.7. Sedation analgesia was established with midazolam (1 mcg/kg/min) and fentanyl (1 mcg/kg/hour), and non-muscular block with vecuronium (0.1 mg/kg/hour). Treatment was initiated; its consisted of antimicrobial drug treatment with ceftriaxone (100 mg per kg, twice daily), and clarithromycin (15 mg per kg, twice daily). Blood and urine cultures were negative. The patients bronchoalveolar lavage was negative for influenza viruses, respiratory syncytial virus, human metapneumovirus, parainfluenza viruses, adenoviruses and enteroviruses, herpes simplex virus, Chlamydia pneumonia, Pneumocystis jirovecii. Serologically, the patient was tested negative for CMV, EBV and adenoviruses. Echocardiography revealed second degree tricuspid insufficiency. Pulmonary arterial pressure was 45 mmHg. Human bocavirus was isolated in a nasopharyngeal specimen using PCR. Antiviral therapy was started with oseltamivir. During observation the patient's blood pressure fell, intravenous fluid support was increased and dopamine infusion (10 mcg/kg/min) was started. Hypotension persisted, and 0.05 mcg/kg/min noradrenalin was added to treatment and gradually increased to 1 mcg/kg/min. Hypotension persisted, and the patient died on the 4<sup>th</sup> day of hospitalization.



**Figure 1.** Areas of peribronchial and interstitial infiltration were present on PA radiographs

The patient was intubated due to respiratory insufficiency and respirated on a mechanical ventilator in synchronized intermittent mandatory ventilation-pressure support mode (SIMV-PS). The mechanical ventilator parameters were set at PIP: 22 cm H<sub>2</sub>O, PEEP:5 cm H<sub>2</sub>O, F: 30/min and FiO<sub>2</sub> :0.7. Sedation analgesia was established with midazolam (1 mcg/kg/min) and fentanyl (1 mcg/kg/hour), and non-muscular block with vecuronium (0.1 mg/kg/hour). Treatment was initiated; its consisted of antimicrobial drug treatment with ceftriaxone (100 mg per kg, twice daily), and clarithromycin (15 mg per kg, twice daily). Blood and urine cultures were negative. The patients bronchoalveolar lavage was negative for influenza

viruses, respiratory syncytial virus, human metapneumovirus, parainfluenza viruses, adenoviruses and enteroviruses, herpes simplex virus, Chlamydia pneumoniae, Pneumocystis jirovecii. Serologically, the patient was tested negative for CMV, EBV and adenoviruses. Echocardiography revealed second degree tricuspid insufficiency. Pulmonary arterial pressure was 45 mmHg. Human bocavirus was isolated in a nasopharyngeal specimen using PCR. Antiviral therapy was started with oseltamivir. During observation the patient's blood pressure fell, intravenous fluid support was increased and dopamine infusion (10 mcg/kg/min) was started. Hypotension persisted, and 0.05 mcg/kg/min noradrenalin was added to treatment and gradually increased to 1 mcg/kg/min. Hypotension persisted, and the patient died on the 4<sup>th</sup> day of hospitalization.

## DISCUSSION

Seckel syndrome is a rare, clinically and genetically heterogeneous inherited developmental disorder. It is characterized by proportionate short stature, and facial appearance with a flat brow, a beak-like nose, low ears and micrognathia. Other findings include low birth weight, bone age retardation, clinodactyly in the fifth finger, 11 pairs of ribs, dislocation of the radial head and mental retardation (3). Low birth weight, motor-mental retardation, proportionate short stature, a beak-like nose, a high palate and low ears were also present in our case. Our patient had previously been clinically diagnosed with Seckel syndrome and was under monitoring due to motor-mental retardation. Immunodeficiency have not been reported to date in Seckel syndrome.

In 2005, Swedish researchers showed the existence of a new virus, HBoV, in nasopharyngeal aspirate specimens taken from children exhibiting findings of lower airway infection. Since these patients were not infected with any other viral pathogen, HBoV was considered a cause of lower airway infections, rather than a coincidental finding. Bocavirus is a virus with no envelope, 18-26 nm in diameter and 4-6 kb long, containing a linear single-helix DNA genome. A prevalence of 2-21.5% has been reported in various studies, particularly in children under the age of 3 with lower respiratory tract infections (4-6).

HBoV positivity was determined in 8 (6.7%) out of 120 patients under the age of 5 and diagnosed with lower airway infection in a study by Demirci in 2009. The mean age of these patients was 9 months (7).

Although HBoV infections are reported to exhibit no seasonal variation, Lu et al. reported that 75% cases occurred in February and March in a study from Thailand (8).

Respiratory tract viruses frequently cause co-infections. HBoV infections are reported to cause high levels of co-infection with respiratory syncytial virus (RSV), influenza virus, parainfluenza virus (PIV) human metapneumovirus (HMPV) and adenoviruses (9).

Bastien et al. (10) reported that 50% of patients with bocavirus infection were hospitalized and that 89% of these were children under the age of 5. They recommended that greater care be taken regarding bocavirus infections in children under 5.

Following the discovery of the first HBoV, three more bocaviruses were found in stool specimens, and comorbidity was shown with gastrointestinal disease. The disease resembles other respiratory tract diseases with findings such as cough, fever, sore throat, tachypnea and wheezing. It may rarely also cause respiratory failure. One study reported oxygen requirement in 73% of patients with HBoV (11). When our patient was admitted to the intensive care department he was started on 5 lt/min oxygen therapy with a non-rebreather mask with a reservoir. Despite this, O<sub>2</sub> saturation was measured at 93%. Respiratory insufficiency developed during monitoring and the patient was given mechanical ventilation support.

A study from France in 2006 reported a mean SaO<sub>2</sub> value of 93%, leukocyte numbers 13,200 mm<sup>3</sup> and CRP 13.5 mg/dl in 26 patients aged under 5 hospitalized due to lower airway infection and with HBoV infection. Body temperature exceeded 38 °C and cough symptoms were present in 50% of cases; 46% were diagnosed with bronchiolitis, 28% with asthma, 15% with upper airway infection and 11% with pneumonia. Duration of hospitalization ranged from 1 to 6 days (12). Peribronchial and interstitial infiltration and hyperinflation may be seen on pulmonary radiographs. White blood cell and CRP values were also elevated in our case. Areas of widespread infiltration were present on radiographs.

The purpose of this case report is to emphasize the need to remember HBoV infections in cases of respiratory insufficiency, particularly under the age of 5, and that although the condition is rare in the literature, intensive care may still be required.

## REFERENCES

1. Allender T, Tammi MT, Eriksson M, Bjerkner A, Tiveljung-Lindell A, Andersson B. Cloning of a human parvovirus by molecular screening of respiratory tract samples. *Proc Natl Acad Sci U S A* 2005;102(36):12891-6.
2. Yaman G. Human Bocavirüs. *Van Tıp Dergisi* 2006;13:109-12.
3. Jones K. Smith's recognisable patterns of human malformation. Sixth Edition:WB Saunders, Philadelphia 2005:108-10.
4. Arnold JC, Singh KK, Spector SA, Sawyer MH. Human bocavirus:prevalence and clinical spectrum at a children's hospital. *Clin Infect Dis* 2006;43:283-8.
5. Fry AM, Lu X, Chittaganpitch M, Peret T, Fischer J, Dowell SF, et al. Human Bocavirus: a novel parvovirus epidemiologically associated with pneumonia requiring hospitalization in Thailand. *JID* 2007;195:1038-45.
6. Liu WK, Chen DH, Liu Q, Liang HX, Yang ZF, Qin S, Zhou R, et al. Detection of human bocavirus from children and adults with acute respiratory tract illness in Guangzhou, southern China. *BMC Infect Dis* 2011;(14)11:345.

7. Demirci P. Human bocavirüs prevalence in children under 5 years of age with lower respiratory tract infection. Thesis. Dr. Lütfi Kırdar Kartal Training and Research Hospital, Department of Child Health and Diseases. İstanbul, 2009.
8. Lu X, Peret TC, Fry AM, Chittaganpitch M, Mackay IM, Olsen SJ, et al. Human Bocavirus associated pneumonia in rural Thailand. International Conference on Emerging Infectious Diseases, March 19-22, 2006, Atlanta, GA. Poster Session 53, Page 149.
9. Yüksel H, Türkeli A. New Viral Pathogenes among Pediatric Patients with pneumonia. Güncel Göğüs Hastalıkları Serisi 2014;2:106-16.
10. Bastien N, Brandt K, Dust K, Ward D, Li Y. Human Bocavirus infection, Canada, Emerg Infect Dis 2006;12:848-50.
11. Parlakay AÖ, Kara A. New Respiratuar Virus. Turkish Pediatric Journal 2010;53:5965.
12. Foulongne V, Rodiere M, Segondy M. Human Bocavirus in children. Emerg Infect Dis 2006;12:862-3.