



Case report

Peripheral facial paralysis in a child with Crimean-Congo hemorrhagic fever

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ABSTRACT

A patient with Crimean-Congo hemorrhagic fever (CCHF), a tick-borne zoonotic disease caused by the Nairovirus is presented in this paper, as an unusual cause of unilateral peripheral facial paralysis. He was 10 years old and admitted to hospital with a 1-day history of fever, frontal headache, fatigue, nausea, vomiting, malaise, arthralgia and myalgia. Real-time PCR confirmed the diagnosis of CCHF. Facial paralysis (grade IV based on the House-Brackman classification) was developed on fifth day of hospitalization. A tick occluding the external auditory canal was seen during the otological examination. The right tympanic membrane was perforated at the postero-inferior quadrant.

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Crimean-Congo hemorrhagic fever (CCHF) is a tick-borne zoonotic disease caused by the Nairovirus of the *Bunyaviridae* family. The virus is transmitted to humans via a bite from the Ixodesticks or by direct contact with blood or tissues of infected humans or viremic livestock. CCHF is a severe disease in humans, with a high mortality rate (2.8–70%) [1,2]. In humans, ticks infest both hairy and non-hairy parts of the body and have also been found in the ear canal [3,4]. Insects and arachnoid foreign bodies in the external auditory canal can make patients feel great distress as a result of otalgia, tinnitus, and hearing loss [5].

Unilateral acute peripheral facial paralysis is a frequent, benign, neurologic disorder, which has a population incidence of 20–25/100,000, and its etiology remains unknown and many factors are thought to be responsible [6]. We present the case of a 10-year-old boy with a tick found in the external auditory canal who was treated for CCHF. He was also diagnosed as having unilateral peripheral facial paralysis on the 5th day of hospitalization.

1. Case report

A 10-year-old boy who lives in a northeastern Anatolian city was admitted to the local emergency service during the second week of June with a 1-day history of fever, frontal headache, fatigue, nausea, vomiting, malaise, arthralgia and myalgia and he was then referred to our emergency department. He was the son of a farmer in a rural area. We made the diagnosis of CCHF and

hospitalised him for further study and began treating him with oral ribavirin and ceftriaxon along with additional supportive medical therapy. Real-time PCR confirmed our diagnosis. We asked for a consult with the otorhinolaryngology clinic on the fifth day of his hospitalisation when we noticed his fascial asymmetry. His left eye was partially closes (Fig. 1a), his mouth was asymmetric (Fig. 1b), and he was lisping though we described him as grade IV based on the House-Brackman classification. Otherwise, his neurologic examination was normal. We found a tick occluding the external auditory canal during the otological examination. The right tympanic membrane was perforated at the postero-inferior quadrant (Fig. 2). The tick was completely removed with a fine cup-forceps under the light microscope. The tick was a *Hyalomma marginatum*, which is the most frequent type found in Turkey and also a vector for CCHF. The pure threshold audiogram and TPOAE tests showed that the boy suffered no hearing loss. No ipsilateral or contralateral stapes reflex was detected by acoustic impedance-metry. We started steroid therapy (2 mg/kg prednisolone) just after the removal of the tick. Significant recovery of the fascial paralysis and the lisp were detected on the sixth day of the steroid therapy. The boy was fully recovered and released eleven days after being hospitalized.

2. Discussion

Many different species of insects and arachnids may present as foreign bodies in the external auditory canal. Ticks are found relatively rarely in the external auditory canal [7]. Ticks are the second-most important vectors, after mosquitoes, of viral, bacterial, and protozoal diseases, such as rickettsiosis, borreliosis, babesiosis, ehrlichiosis, tularemia and Crimean-Congo haemor-

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Fig. 1. (a and b) Pretreatment facial examination.

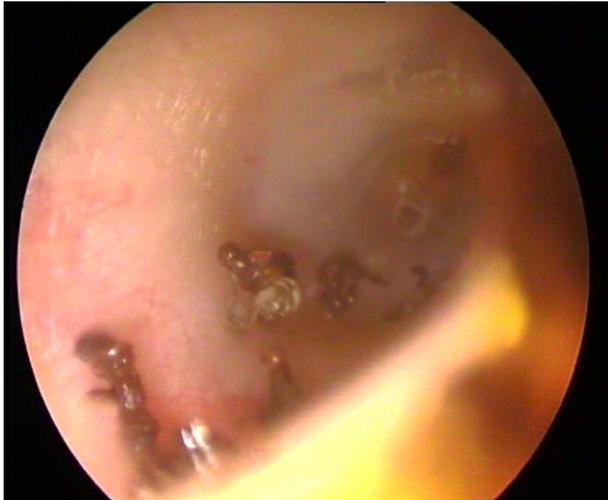


Fig. 2. Otoendoscopic view: the perforation at the postero-inferior quadrant of right tympanic membrane.

rhagic fever in humans all over the world [8,9]. CCHF epidemics have been reported in Central Asia, Africa, and Eastern Europe [10–14]. CCHF is an acute, infectious illness that can cause multi-organ failure and death. CCHF is a haemorrhagic syndrome presenting with fever, nausea, vomiting, myalgia, and bleeding from various sites. Leucopenia, thrombocytopenia, high levels of alanine aminotransferase, aspartate aminotransferase and lactate dehydrogenase are commonly detected [15,16]. A tick bite may cause isolated facial nerve paralysis by infestation of the ear canal. A very small number of cases of isolated facial nerve paralysis by tick infestation were reported in the literature [17–19]. Some theories were posited for the pathogenesis. Toxin, secreted from the salivary glands of the arthropode, may reach the facial nerve via perforation of the tympanic membrane and natural dehiscence of the Fallopian canal [4]. This toxin is found to interfere with the liberation or synthesis of acetylcholine at the motor end plate of muscle fibre [20]. In cases that the tympanic membrane is intact, direct extension of the inflammatory process to the fallopian canal is via persistent dehiscence or direct invasion of the infectious organisms into the facial canal through the middle ear which results in edema of the inflamed nerve within the canal [19]. Using

computerized tomography, we found perforation of the tympanic membrane but no dehiscence of the Fallopian canal. We think the toxin reached the facial nerve via the perforation of the middle ear and affected the facial nerve via a dehiscence of an unknown origin. In conclusion, a tick bite might cause peripheral facial paralysis in the endemic regions of CCHF so a careful otologic examination should be performed in addition to the routine examination.

Appendix A. Supplementary data

Supplementary data associated with this article can be found, in the online version, at doi:10.1016/j.pedex.2010.08.009.

References

- [1] C.A. Whitehouse, Crimean-Congo hemorrhagic fever, *Antiviral Res.* 64 (2004) 145–160.
- [2] O. Ergonul, A. Celikbas, N. Baykam, S. Eren, B. Dokuzoguz, Analysis of risk-factors among patients with Crimean-Congo haemorrhagic fever virus infection: severity criteria revisited, *Clin. Microbiol. Infect.* 12 (2006) 551–554.
- [3] R. Indudharan, A.S. Dharap, T.M. Ho, Intra-aural tick causing facial palsy, *Lancet* 348 (1996) 613.
- [4] R. Indudharan, M. Ahamad, T.M. Ho, R. Salim, Y.N. Htun, Human otoacariasis, *Ann. Trop. Med. Parasitol.* 93 (1999) 163–167.
- [5] P. Antonelli, A. Ahmadi, A. Prevatt, Insecticidal activity of common reagents for insect foreign bodies of the ear, *Laryngoscope* 111 (2001) 15–20.
- [6] G. Arias, J. Noguez, M. Manos, E. Amilibia, M. Dicenta, Bilateral facial nerve palsy: four case reports, *J. Otorhinolaryngol. Relat. Spec.* 60 (1998) 227–229.
- [7] S. Iwasaki, S. Takebayashi, T. Watanabe, Tick bites in the external auditory canal, *Auris Nasus Larynx* 34 (2007) 375–377.
- [8] R.N. Charrel, H. Attoui, A.M. Butenko, J.C. Clegg, V. Deubel, T.V. Frolova, et al., Tick-borne virus diseases of human interest in Europe, *Clin. Microbiol. Infect.* 10 (2004) 1040–1055.
- [9] P. Parola, D. Raoult, Tick-borne bacterial diseases emerging in Europe, *Clin. Microbiol. Infect.* 7 (2001) 80–83.
- [10] R. Swanepoel, A.J. Shepherd, P.A. Leman, S.P. Shepherd, G.B. Miller, A common-source outbreak of Crimean-Congo haemorrhagic fever on a dairy farm, *S. Afr. Med. J.* 68 (1985) 635–637.
- [11] A. Altaf, S. Luby, A.J. Ahmed, N. Zaidi, A.J. Khan, S. Mirza, et al., Outbreak of Crimean-Congo haemorrhagic fever in Quetta, Pakistan: contact tracing and risk assessment, *Trop. Med. Int. Health* 3 (1998) 878–882.
- [12] A.S. Khan, G.O. Maupin, P.E. Rollin, A.M. Noor, H.H. Shurie, A.G. Shalabi, et al., An outbreak of Crimean-Congo hemorrhagic fever in the United Arab Emirates, 1994–1995, *Am. J. Trop. Med. Hyg.* 57 (1997) 519–525.
- [13] L.N. Iashina, V.S. Petrov, I.D. Petrova, V.V. Gutorov, S.V. Kazakov, K.S. Ospanov, et al., Genetic identification of the Crimean-Congo hemorrhagic fever virus during epidemic outbreak in Kazakhstan in 2000, *Mol. Gen. Mikrobiol. Virusol.* (4) (2002) 31–35 (Russian).
- [14] A. Papa, S. Bino, A. Llagami, B. Brahijam, E. Papadimitriou, V. Pavlidou, et al., Crimean-Congo hemorrhagic fever in Albania, 2001, *Eur. J. Clin. Microbiol. Infect. Dis.* 2 (2002) 603–606.
- [15] R. Swanepoel, D.E. Gill, A.J. Shepherd, P.A. Leman, J.H. Mynhardt, S. Harvey, The clinical pathology of Crimean-Congo hemorrhagic fever, *Rev. Infect. Dis.* 11 (1989) 794–800.
- [16] H. Hoogstraal, The epidemiology of tick-borne Crimean-Congo hemorrhagic fever in Asia, Europe, and Africa, *J. Med. Entomol.* 15 (1979) 307–417.
- [17] M.K. Gürbüz, M. Erdoğan, N. Doğan, L. Birdane, C. Cingi, E. Cingi, Case report: isolated facial paralysis with a tick, *Turk. Parazitol. Derg.* 34 (2010) 61–64 (in Turkish).
- [18] A. Zamzil Amin, A. Baharudin, H. Shahid, S. Din Suhaimi, M.J. Nor Affendie, *Isolated Sci.* 2 (2007) 51–53.
- [19] M.K. Miller, Massive tick (*Ixodes holocyclus*) infestation with delayed facial-nerve palsy, *Med. J. Aust.* 176 (March(6)) (2002) 264–265.
- [20] V. Vedanarayanan, W.H. Sorey, S.H. Subramony, Tick paralysis, *Semin. Neurol.* 24 (2) (2004) 181–184.