Cotard delusion in a patient with Shapiro syndrome

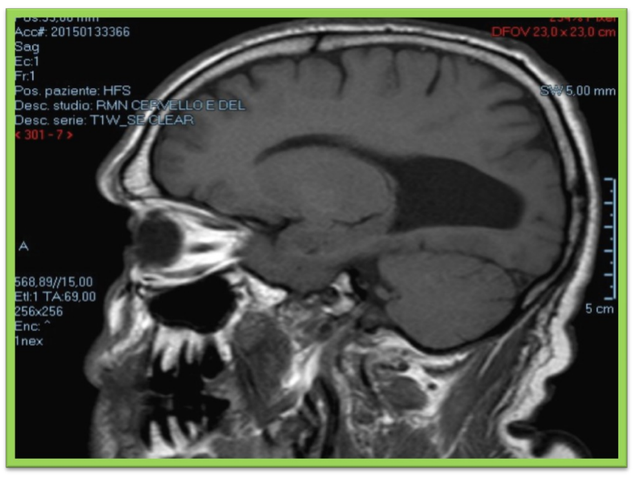
Filomena Barbone, M. De Angelis, F. Anzellotti, R. Di Giacomo, R. Telese, V. Di Stefano, M. Vitale, C. Ferrante, M. Onofrj

Department of Neurosciences, Imaging and Clinical Sciences - University G.d'Annunzio of Chieti-Pescara - Chieti

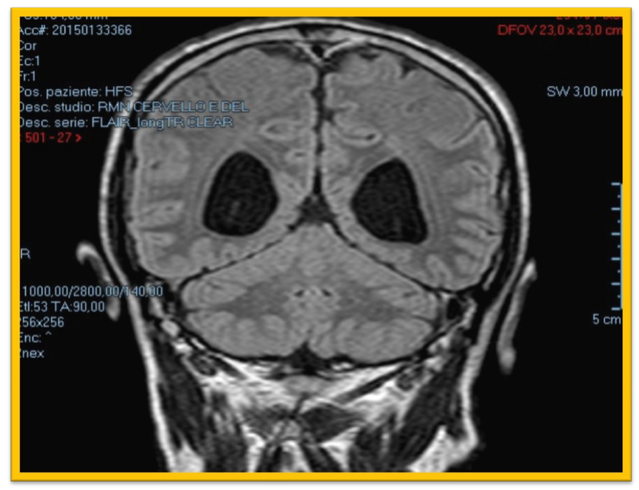
Objective: to describe an association between Shapiro syndrome and Cotard delusion.

Materials and methods: Revision of clinical and radiological data in a patient with Shapiro syndrome.

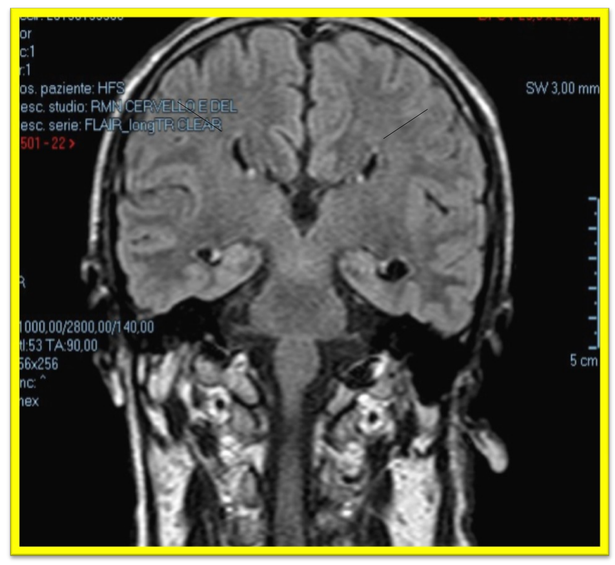
Case Report: A 55 year-old man was admitted to our emergency department because of profuse sweating, hypothermia and coma. Several years ago a similar attack had occurred and he was diagnosed as having Shapiro syndrome. His medical history included arterial hypertension. Physical examination revealed pulse rate at 80 beats per minute, blood pressure of 130/80 mmHg and axillary temperature of 33°C. At neurological examination patient was in comatose state with Glasgow Coma Scale: 3, direct and consensual light pupil reflex were preserved, while lower limbs areflexia and an inconstant extensor plantar response bilaterally could be observed. Routine blood analyses and electrocardiography were normal. Head Computed Tomography (CT) scan showed corpus callosum agenesis with no signs of acute ischemia or intracranial hemorrhage. Brain Magnetic Resonance Imaging (MRI) confirmed congenital corpus callosum agenesis. Medications administered prior to admission included clonazepam, clonidine, melevodopa/carbidopa and multi-vitamin integrators. Patient was treated with adequate hydration and heated up using thermic blanket in order to re-establish state of consciousness. Sweating and hypothermia attacks occurred almost daily. Patient was managed with Cyproheptadine 4 mg three times a day with significant symptoms improvement. Unfortunately, attacks did not halt completely and during the sixth day of hospitalization, he presented nihilistic delusion: “My bladder exploded”; “I’m dead because I’ve lost my organs”. He responded well to Haloperidol 2 mg intramuscular and Chlorpromazine 50 mg intramuscular to administered as necessary and totally remitted after two days. Electroencephalography was normal. Patient was discharged after 39 days of hospitalization with 34.5 C axillary temperature and normal neurological and cognitive state.



Sagittal T1-weighted showing corpus callosum agenesis



Coronal view revealing complete agenesis of the corpus callosum



Coronal image showing the ‘Viking helmet’ or ‘moose head’ appearance assumed by lateral ventricles in the absence of the corpus callosum (white arrows).

Discussion: Shapiro syndrome is an extremely rare disorder consisting of periodic hypothermia, hyperhidrosis and corpus callosum agenesis. There are approximately 50 described cases of Shapiro syndrome in literature. Cotard syndrome, a relatively rare condition comprising any one of a series of delusions that range from a belief that one has lost organs, blood, or body parts to insisting that one has lost one’s soul or is dead[1], may be seen in neurological patients[2]. Shapiro described an “agitated delirium” in his syndrome, but it wasn’t a proper Cotard syndrome[3].
We described a patient with Shapiro syndrome, who never suffered from psychiatric disorder and developed a Cotard syndrome. This case highlights that Cotard delusion may be associated with Shapiro syndrome.

References:
1.Ruminjo A, Mekinulov B. A case report of Cotard’s syndrome. Psychiatry (Edgmont) 2008 June; 5(6):28-29.
2.Pazderska Ramirez-Bermudez J, Aguilar-Venegas LC, Crail-Melendez D, Espinola-Nadurille M, Nente F, Mendez MF. Cotard syndrome in neurological and psychiatric patients. J Neuropsychiatry Clin Neurosci. 2010 Fall; 22(4):409-16.
3.Shapiro WR, Williams GH, Plum F. Spontaneous recurrent hypothermia accompanying agenesis of the corpus callosum. Brain. 1969;92:423–36.

Sin
XLVII CONGRESSO NAZIONALE
22-25 OTTOBRE 2016 – VENEZIA
WebPoster