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Case Report

Pituitary Abscess Causing Panhypopituitarism in a Patient With Neurobrucellosis: Case Report

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ABSTRACT

Background/Objective: Pituitary abscess is an uncommon life-threatening disease that could lead to panhypopituitarism. It is important to suspect its prevalence in regions with endemic infectious diseases.

Case Report: A 55-year-old man, a farmer, with a background of consumption of unpasteurized dairy products, presented with headache, impaired consciousness, and fever that started in February 2023. Initial test results were consistent with neuroinfection. Brain MRI showed ventriculitis; the pituitary gland was heterogeneous with the presence of an 8 × 8 mm abscess. The pituitary hormone axis was evaluated, and it showed results compatible with the results of panhypopituitarism with central hypothyroidism, central hypocortisolism, central hypogonadism, and growth hormone deficiency. Hormone replacement treatment with hydrocortisone and levothyroxine was started. The Rose Bengal test for *Brucella spp.* and 2-mercaptoethanol Brucella agglutination test showed positive results. After neurobrucellosis (NB) was diagnosed, antibiotic treatment was commenced. The patient was discharged 6 weeks later and treatment with prednisone, levothyroxine, recombinant somatropin, testosterone, as well as doxycycline, and rifampin was continued for another 4 months.

Discussion: NB and pituitary abscess are rare manifestations of brucellosis and are challenging to diagnose due to their nonspecific clinical presentation and cerebrospinal fluid (CSF) findings. NB diagnosis relies on neurologic symptoms and serological evidence of Brucella infection. Magnetic resonance imaging is the preferred diagnostic tool for pituitary abscesses. Medical management may be sufficient, while transsphenoidal drainage is not always necessary. Hormonal deficits typically remain permanent.

Conclusion: Pituitary abscess could be suspected in patients presenting with symptoms of neuroinfection, panhypopituitarism, and heterogenous image in the magnetic resonance imaging differential diagnosis. Opportune management can lead to reduced mortality and improved recovery of the pituitary hormone function.

Introduction

Brucellosis, caused by *Brucella melitensis*, is a frequent zoonotic disease with a global annual incidence of 500 000 cases.[1] It is common among farmers and people who consume unpasteurized milk products.[2] Patients may exhibit multiple manifestations and complications. Neurobrucellosis (NB) is a frequently missed complication.

Pituitary abscesses are infrequent abnormalities that develop within preexisting lesions, including adenomas, craniopharyngiomas, and Rathke's cleft cysts, or because of surgical interventions.[3] Existing literature documents approximately 200 reported cases.[4] Clinical presentation can be nonspecific with a lack of signs of systemic infection, making the diagnosis a challenge. The most presented signs and symptoms include headache, diabetes insipidus, and hypopituitarism.[4] We describe a case of a pituitary

Abbreviations: ACTH, adrenocorticotropic hormone; CSF, cerebrospinal fluid; CT, computed tomography; FSH, follicle-stimulating hormone; IGF-1, insulin-like growth factor 1; LH, luteinizing hormone; MRI, magnetic resonance imaging; NB, Neurobrucellosis; T3, triiodothyronine; T4, thyroxine; TSH, thyroid-stimulating hormone.

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abscess secondary to NB presenting with panhypopituitarism, which was treated with conservative management.

Case Report

A 55-year-old farmer from Mexico State, Mexico, with a clinical background of consumption of unpasteurized dairy products, presented with a 2-week history of headache, impaired consciousness, fever, and vomiting in February 2023. He went to a local health institution. On admission, vital signs were normal; physical examination showed a stiff neck. Initial head computed tomography (CT) scan did not report alterations, and a lumbar puncture, which is a cerebrospinal fluid (CSF) cytochemical study, showed 800 leukocytes, for which he was referred to our institution with diagnosis of acute meningitis.

The findings of the initial tests were compatible with the results of neuroinfection tests: CSF showed increased proteins (>200 mg/dL, n < 45 mg/dL), decreased glucose (14 mg/dL with a serum glucose concentration of 76 mg/dL), and pleocytosis (800 mm^3, n < 5 mm^3), and negative results were obtained for tuberculosis polymerase chain reaction. Blood and CSF cultures without isolates. C-reactive protein was slightly elevated (2.63 mg/dL, n < 5 mg/dL). A second head CT scan was taken and normal results were obtained. Treatment with ceftriaxone, vancomycin, ampicillin, and acyclovir was started due to the diagnosis of acute meningitis.

Subsequently, a brain magnetic resonance imaging (MRI) was performed, and it revealed ventriculitis at the 3rd and 4th ventricle with suprasellar extension and associated hypothalamic encephalitis. The pituitary gland was observed with a heterogeneous appearance with the presence of an 8 × 8 mm abscess (Fig. 1).

After obtaining the MRI, pituitary axis hormones were evaluated, and the results are compatible with the results of panhypopituitarism with central hypothyroidism, central hypocortisolism, central hypogonadism, and growth hormone deficiency (Table). Hormone replacement treatment was initiated with hydrocortisone and levothyroxine.

Owing to the epidemiologic background (consumption of unpasteurized dairy products), the Rose Bengal test for *Brucella spp.* and 2-mercaptoethanol Brucella agglutination test were performed, and positive results were obtained. NB was diagnosed, and treatment with doxycycline, rifampin, and ceftriaxone was started. Neurosurgery service suggested conservative management with antibiotics.

Laboratory tests performed 7 days after hospital admission revealed hyponatremia, serum hypoosmolarity, urinary hyperosmolarity, and high natriuresis, whereof, the additional diagnosis of syndrome of inappropriate antidiuretic hormone secretion (SIADH) was established.

The patient showed improvement in his neurologic status, and infection symptoms disappeared. Laboratory tests 2 weeks after admission showed normal serum sodium, normal serum osmolality, normal urine osmolality, normal natriuresis, normal thyroid-stimulating hormone, normal free T4, decreased total T3, normal serum cortisol, decreased follicle-stimulating hormone, decreased luteinizing-hormone, decreased testosterone, and normal prolactin concentrations. A CSF cytochemical study showed low pH, decreased glucose, elevated proteins, and increased monocytes (Table). Subsequent pituitary MRI showed an inflammatory process in the ependyma of the third and fourth ventricles, as well as in the hypothalamic region with sellar extension; it also showed a thickened infundibulum, with a pituitary intraglandular abscess with dimensions of 8 × 7 × 12 mm with a 15% decrease compared with the previous study (Fig. 2). The patient was discharged 6 weeks later and sent home on hormone replacements with prednisone, levothyroxine, and somatropin, to be taken indefinitely. Subsequently, the patient decided not to continue with testosterone replacement. Following discharge, the patient continued antibiotic treatment with doxycycline and rifampicin for an additional 4 months.

Laboratory tests were performed 2 weeks after discharge and the results show improvement in pituitary function including clinical improvement (Table). Subsequent laboratory tests were

Highlights

- Brucellosis can present with neurologic complications
- Pituitary abscesses, as a complication of brucellosis, are rarely reported
- This report highlights panhypopituitarism associated with brucellosis

Clinical Relevance

The simultaneous presence of neurobrucellosis and a pituitary abscess is documented infrequently in contemporary literature. The purpose of this case report was to document panhypopituitarism as a secondary consequence of this condition. Although other hormonal dysfunctions are frequently described, this report aimed to shed light on less frequently observed panhypopituitarism.

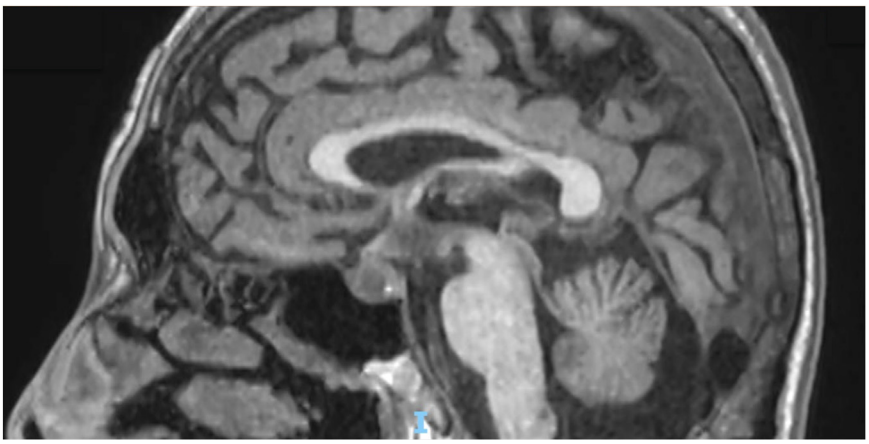

Fig. 1. Brain magnetic resonance imaging showing a heterogeneous pituitary gland of probable infectious/inflammatory etiology due to the presence of an 8 × 8 mm abscess.

Table
Patient's Laboratory Results at Admission, During hospitalization, and After Discharge

Laboratory parameters	Presentation	Discharge	2 wks after discharge	4 mo after discharge	Normal values
Sodium (mmol/L)	130	139	138	141	135-145
Serum osmolality (mOsm/kg)	264	282	283.3	287	275-295
Urine osmolality (mOsm/kg)	481	678	-	-	40-900
Natriuresis (mmol/L)	101.9	60.7	-	-	54-150
	Pituitary axis hormones				
TSH (mIU/L)	1.05	0.47	0.55	1.24	0.3-5
Total T3 (ng/mL)	0.47	0.6	0.79	1.16	0.64-1.81
Free T4 (ng/dL)	0.57	0.81	0.76	0.59	0.63-1.64
ACTH (pg/ml)	20	20	57	-	10-50
Serum cortisol (μg/dL)	8.88	5.72	10.26	-	6.7-22.6
IGF-1 (ng/mL)	9.99	-	29.9	76.89	61-206
LH (mUI/mL)	<0.20	<0.20	0.84	0.82	1.24-8.62
FSH (mUI/mL)	0.37	0.47	1.56	2.73	1.27-19.26
Prolactin (ng/mL)	5.14	7.81	4.40	-	3.9-29.5
Testosterone (ng/mL)	-	0.24	0.51	0.64	1.75-7.81
	Cerebrospinal fluid (CSF) analysis				
pH	8	7.00	-	-	7.35-7.45
Glucose (mg/dL)	14	40	-	-	40-70
Protein (mg/dL)	>200	129.4	-	-	15-45
Leukocytes (cells/μL)	37	-	-	-	<5
Monocytes (mm^3)	-	90%	-	-	0
Red blood cells (mm^3)	100	-	-	-	0
Crenocytes (mm^3)	2	-	-	-	0
Opening pressure (cmH_20)	5	-	-	-	<10

Abbreviations: ACTH = adrenocorticotropic hormone; FSH = follicle-stimulating hormone; IGF-1 = insulin-like growth factor 1; LH = luteinizing hormone; TSH = thyroid-stimulating hormone; T3 = triiodothyronine; T4 = thyroxine.

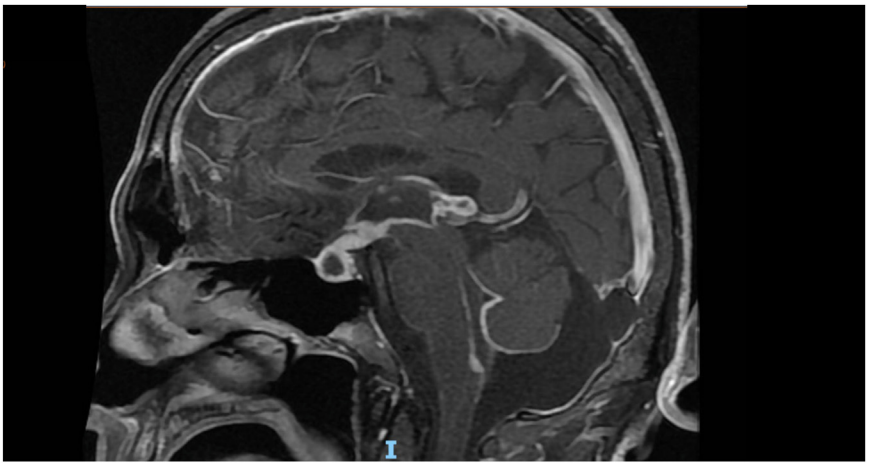

Fig. 2. Pituitary gland magnetic resonance imaging (4 weeks later) showing an inflammatory process in the ependyma of the third and fourth ventricle and the hypothalamic region with sellar extension, due to irregular thickening and enhancement. Thickened infundibulum, with an intraglandular pituitary abscess, which measures 8 × 7 × 12 mm, with a 15% decrease in the volume of the collection compared with the previous study.

conducted 4 months after discharge and demonstrated even greater improvement in the hormonal axes that were previously compromised (Table). These laboratory test results were obtained when the patient was under hormone replacement.

Discussion

In this case report, we present an exceptional case of a pituitary abscess secondary to NB presenting with panhypopituitarism.

Brucella invaded the reticuloendothelial system and caused a bacteremia, crossing the blood–brain barrier, causing NB, which is a rare manifestation that can occur in 0.5% to 25% of the cases of brucellosis.[5] NB may result in deleterious cytokine or endotoxin release, a direct neuropathic effect, and an inflammatory and immunologic response by the host.[6] NB is linked to a considerable mortality rate, reaching up to 7%.[7] NB may not exhibit both typical clinical presentation and specific CSF findings, which can delay its diagnosis from 2 to 12 months after the onset of the symptoms.[8] The main clinical manifestation is meningitis,[2] but patients can also present with severe complications, such as encephalitis, brain abscess, epidural abscess, myelitis, radiculoneuritis, subarachnoid hemorrhage, and psychiatric manifestations.[9]

Simmonds[10] described the first case of pituitary abscess in 1914; since then, the literature has documented >200 cases of pituitary abscess stemming from various causes, with most of them being isolated case reports.[11] Less than 1% of all pituitary lesions are

accounted for by this condition.[12] To our knowledge, only 2 cases of pituitary abscess secondary to NB have been reported in the existing literature.[3,13]

Vates et al[14] described 24 cases in which the main presentation of the pituitary abscess was a headache with no specific pattern (91.7%); approximately 50% had visual impairment, and only 25% had signs of meningism; furthermore, panhypopituitarism was observed in 54.2% of these cases. In our case, the patient did not exhibit any visual disturbances. Gao et al[15] described cases of pituitary abscess presenting with symptoms of pituitary dysfunction in >80% at hospital admission; the most common deficits included adrenocorticotropic hormone and thyroid-stimulating hormone; on the other hand, central hypogonadism was the least reported hormone impairment.

NB diagnosis is made based on neurologic symptoms alongside serologic proof of systemic Brucella infection. Serologic assays, including the standard agglutination test, Rose Bengal test, and enzyme-linked immunosorbent assay (ELISA), are widely recommended due to their ease of use and cost-effectiveness.[9] MRI is the preferred radiographic diagnostic method for the diagnosis of pituitary abscesses, which present as heterogeneous intrasellar lesions with a hyperintense peripheral capsule mimicking apoplexy.[14] In pituitary abscesses, negative cultures are frequently observed (50% of cases).[4] The indolent course of pituitary abscesses often leads to delays in recognition, because they are frequently misdiagnosed as pituitary adenomas.[11] If timely diagnosis and treatment are not initiated, pituitary abscesses may be linked to significant morbidity and mortality rates.[16]

In our patient, surgery was not indicated given his pituitary abscess size as well as the stability of the patient. However, a complete regimen of antibiotics was sufficient to treat the pituitary abscess. Although transsphenoidal drainage is the most effective treatment,[17] some cases may be treated based on medical management depending on each case. The probability of complete hormonal recovery is unlikely, just like in our patient's case; in most patients, a complete hormonal deficit remains permanent and requires continued follow-up.[7,11] In our patient, the last level of serum cortisol concentration was 10.26 μg/dL after he was discharge from the hospital. Prednisone was preferred over hydrocortisone as replacement therapy because it facilitates therapeutic adherence. In this case, a follow-up imaging study has been indicated 1 month after discharge to assess improvement, although the patient has not been able to visit the institution for follow-up imaging studies. The patient continues to receive treatment for the other compromised axis and is followed up.

Conclusion

This is a case of panhypopituitarism due to an abscess secondary to NB. NB should be suspected in a patient with clinical manifestations of neuroinfection of unknown etiology mainly when there is a history of unpasteurized dairy consumption. For patients presenting with symptoms of neuroinfection, panhypopituitarism, and heterogenous image in the MRI, pituitary abscess should be included as a potential diagnosis. Appropriate follow-up after the resolution of the pituitary abscess may result in a lower mortality and a higher recovery of the pituitary hormone function.

Disclosure

The authors have no conflicts of interest to disclose.

Acknowledgment

Patient consent was obtained. This case was presented at the Endocrine Society Meeting, June 15-18, 2023, Chicago, Illinois, United States, with the Poster Board number SAT-609.

References

1. Negrón M, Tiller R, Kharod G. *Brucellosis | CDC Yellow Book 2024*. Travelers Health on CDC Yellow Book. Published 2023. Accessed June 26, 2023. https://wwwnc.cdc.gov/travel/yellowbook/2024/infections-diseases/brucellosis
2. Soares CN, Angelim AIM, Brandão CO, Santos RQ, Mehta R, da Silva MTT. Neurobrucellosis: the great mimicker. *Rev Soc Bras Med Trop*. 2022;55:e05672021. https://doi.org/10.1590/0037-8682-0567-2021
3. Güven MB, Cirak B, Kutluhan A, Ugras S. Pituitary abscess secondary to neurobrucellosis. Case illustration. *J Neurosurg*. 1999;90(6):1142. https://doi.org/10.3171/JNS.1999.90.6.1142
4. Pekic S, Popovic V. Diagnosis of endocrine disease: expanding the cause of hypopituitarism. *Eur J Endocrinol*. 2017;176(6):R269–R282. https://doi.org/10.1530/EJE-16-1065
5. Chandrashekar N, Shome R, Kumari V, Netravathi M. Neurobrucellosis – a clinical masquerade in an Indian scenario; review of 12 cases by correlative diagnostic approach. Published online January 1, 2015. Accessed June 26, 2023. krishi.icar.gov.in/jspui/handle/123456789/9277
6. Kizilkilic O, Calli C. Neurobrucellosis. *Neuroimaging Clin N Am*. 2011;21(4):927–937. https://doi.org/10.1016/J.NIC.2011.07.008
7. Akdeniz H, Irmak H, Anlar Ö, Demiröz AP. Central nervous system brucellosis: presentation, diagnosis, and treatment. *J Infect*. 1998;36(3):297–301. https://doi.org/10.1016/S0163-4453(98)94279-7
8. Guven T, Ugurlu K, Ergonul O, et al. Neurobrucellosis: clinical and diagnostic features. *Clin Infect Dis*. 2013;56(10):1407–1412. https://doi.org/10.1093/CID/CIT072
9. Patra S, Kalwaje Eshwara V, Pai AR, Varma M, Mukhopadhyay C. Evaluation of clinical, diagnostic features and therapeutic outcome of neurobrucellosis: a case series and review of literature. *Int J Neurosci*. 2022;132(11):1080–1090. https://doi.org/10.1080/00207454.2020.1860969
10. Simmonds M. Ueber embolishe prozesse in der hypophysis. *Virchow Arch Pathol Anat*. 1914;217:226–239.
11. Pekic S, Popovic V. Alternative causes of hypopituitarism: traumatic brain injury, cranial irradiation, and infections. *Handb Clin Neurol*. 2014;124:271–290. https://doi.org/10.1016/B978-0-444-59602-4.00018-6
12. Jain KC, Varma A, Mahapatra AK. Pituitary abscess: a series of six cases. *Br J Neurosurg*. 1997;11(2):139–143. https://doi.org/10.1080/02688699746492
13. Yang X, Liu X, Li W, Chen D, Ma C. Neurobrucellosis presenting as a pituitary abscess: a case report and review of literature. *Int J Clin Exp Med*. 2017;10(6):9669–9674.. Accessed June 26, 2023. www.ijcem.com
14. Vates GE, Berger MS, Wilson CB. Diagnosis and management of pituitary abscess: a review of twenty-four cases. *J Neurosurg*. 2001;95(2):233–241. https://doi.org/10.3171/JNS.2001.95.2.0233
15. Gao L, Guo X, Tian R, et al. Pituitary abscess: clinical manifestations, diagnosis and treatment of 66 cases from a large pituitary center over 23 years. *Pituitary*. 2017;20(2):189–194. https://doi.org/10.1007/S11102-016-0757-7
16. Agyei JO, Lipinski LJ, Leonardo J. Case report of a primary pituitary abscess and systematic literature review of pituitary abscess with a focus on patient outcomes. *World Neurosurg*. 2017;101:76–92. https://doi.org/10.1016/J.WNEU.2017.01.077
17. Zhang X, Sun J, Shen M, et al. Diagnosis and minimally invasive surgery for the pituitary abscess: a review of twenty-nine cases. *Clin Neurol Neurosurg*. 2012;114(7):957–961. https://doi.org/10.1016/J.CLINEURO.2012.02.020