

Solitary bone plasmacytoma, a rare monoclonal gammopathy. A case report

Lisbet Labrada-Placeres 1 , Alain Areces-López 1 , Anadely Gámez-Pérez 2 , Anadely Gámez-Pérez

- 1 Universidad de Ciencias Médicas de La Habana. Facultad de Ciencias Médicas de Artemisa. Artemisa, Cuba.
- 2 Universidad de Ciencias Médicas de La Habana. Hospital General Docente "Comandante Pinares". Artemisa, Cuba

ABSTRACT

Solitary bone plasmacytoma is a rare presenting plasma cell tumor that has been observed as a precursor to multiple myeloma; it is located mainly in the spine, causing pain at the thoracic and/or lumbar level. We report the case of a 55-year-old female patient who presented long-standing low back pain without associated trauma, accompanied by paraparesis; a chest X-ray was indicated, which revealed an osteolytic image, corresponding to a solitary bone plasmacytoma that was confirmed by histological examinations. After pharmacological treatment and radiotherapy, the evolution of the patient was satisfactory. Solitary bone plasmacytoma is a rare monoclonal gammopathy that should be suspected in the presence of axially located bone pain without a history of trauma associated with paraparesis. Its main differential diagnosis is multiple myeloma, which is sometimes the final result of its evolution.

Keywords: Multiple Myeloma; Plasmacytoma; Paraparesis; Paraproteinemias.

lasma cell dyscrasias, currently monoclonal gammopathies, comprise a series of pathological processes that reflect a disorder in the proliferation of immunoglobulin-producing cells. The excessive proliferation of a single clone of plasma cells, which causes the synthesis of large amounts of a single type of the main classes of immunoglobulins (component M), is considered as such1.

Monoclonal gammopathies include Waldenström's macroglobulinemia, multiple myeloma (MM), heavy chain disease, monoclonal gammopathy of uncertain significance, amyloidosis, solitary plasmacytoma, and extramedullary plasmacytoma¹.

Solitary plasmacytoma accounts for fewer than 10 % of plasma cell neoplasms and presents itself as a localized clonal plasma cell tumor. This affects mostly males with a 4:1 ratio, between 50 and 60 years of age, with a predominance of presentation in black people (30 %) 2, 3, 4.



OPEN ACCESS

O Corresponding author: Alain Areces López e-mail: alainareces@gmail.com

Published: October 05th, 2021

Received: September 20th, 2021; Accepted: September 22nd, 2021 Cite as:

Labrada Placeres L. Areces López A. Gámez Pérez A. Plasmocitoma óseo solitario, una gammapatía monoclonal infrecuente. Informe de un caso. 16 de Abril [Internet], 2021 [citado:fecha de acceso]; 60 (282); e1456. Disponible en: http://www.rev16deabril.sld.cu/index.php/16 04/article/view/1456

Conflict of interests

The authors declare no conflict of interests.

Two main types are distinguished: bone plasmacytoma, which in most cases occurs in bones of the rachis, and extraosseous plasmacytoma, more common in the subepithelium of the respiratory (paranasal sinuses, nasopharynx, larynx) and upper digestive tracts 2.

Reportedly, 50 % of patients with solitary plasmacytoma progress to MM in the three years next to diagnosis, being more frequent the progression in those who do not receive timely treatment. The prognosis of this gammopathy is significantly more favorable than that the prognosis of MM, so it is essential to make an early diagnosis to prevent or delay its evolution and increase the survival of these patients ^{5,6}.

CASE REPORT

A 55-year-old female patient, black, an educator, with a personal pathological history of rheumatoid disease (for which she received pharmacological treatment with azathioprine and prednisone) came to the clinic about two years ago because she presented bone pain located mainly in the spine at the thoracic level, of great intensity and stabbing nature, which radiated to the entire lumbar region, with three months of evolution. A history of associated trauma was not reported.

In the following months, the pain progressed in intensity, with limited relief after the use of pain relievers and anti-inflammatories. It was accompanied by weakness in the lower limbs, which caused some inability to walk, without loss of sensation or paresthesia.



CC BY-NC-SA 4.0

1

As a family pathological history stands out the suffering of arterial hypertension on the part of maternal relatives. The patient has undergone surgical interventions on three occasions (hysterectomy, appendectomy, and umbilical hernia raffia), without the existence of postoperative complications. She has received blood transfusions on two occasions. She did not report any toxic habits and has a drug allergy to cotrimoxazole and tramadol.

During the physical examination hypocoloured and normohydric mucous membranes were found, as well as bilateral thoracic and lumbar paravertebral pain on palpation with positive Lassegue sign. The rest of the physical examination showed no alterations.

The hematometric parameters initially showed decreased hemoglobin (90 g/L) which confirmed the existence of suspected anemia on physical examination, accelerated erythrocytosis (ESR) (115 mm/h), slight thrombocytosis (450 x 109 /L), and elevation of uric acid (345 µmol/L) and total proteins (100 g/L).

Chest X-ray showed radiopaque lesions at the level of the eighth thoracic vertebra. Computerized Tomography (CT) revealed normal lung parenchyma, without pleural effusion, with slight septal thickening; when evaluating the bone structures, the existence of destruction of the posterior costal area was observed in T8 at the level of the joint with the vertebral body, presenting a hyperdense image whose density oscillated around 74 HU, with a diameter of 22 x 35 mm. The rest of the vertebral body, the transverse and spinous processes at this level were normal. Magnetic Resonance Imaging (MRI) reported the presence of a pathological fracture at the T12, L1, and L3 levels, which caused the patient to accentuate the kyphosis.

Pathological studies showed, in the bone marrow aspiration, an infiltration by plasma cells greater than 60 %. These cells were binucleated and with vacuoles. Immunohistochemistry with CD138, a specific marker for plasma cells, was positive for Lambda-type tumor cells.

Soft tissue cytology confirmed the diagnosis of plasmacytoma. The patient did not meet the criteria for multiple myeloma (MM), so she was diagnosed with solitary bone plasmacytoma.

Therapeutic intervention

Immediately based on diagnosis, pharmacological treatment with dexamethasone 100 mg diluted in 200 mL of 0,9% physiological saline solution was established for 3 hours, once a day, for 3 days intravenously. A blood transfusion with 700 mL of red blood cells for a post-transfusion hemoglobin of 100 g/L was given. Subsequently, she received treatment with radiotherapy (15 sessions), with a favorable response, and a monthly re-evaluation in consultation was decided.

Monitoring and results

The complementary evolutionary examinations showed an improvement in the patient's health status concerning those performed at the time of diagnosis, with a decrease in ESR (36 mm/h), a decrease in total proteins (73 g/L), and an increase in the hematocrit.

Currently, the patient remains without disease progression, with a good response to treatment and a good general condition. A rehabilitation of joint function has been achieved, which has allowed her reincorporation to social and work life.

She is still under pharmacological treatment: dexamethasone (4 mg bulb) 20 mg for 3 days, intravenously, diluted in 300 mL of 0,9 % NaCl to last 2 hours each month, and thalidomide (100 mg) one tablet daily.

Two years after diagnosis, symptoms or clinical and analytical signs that point to a recurrence or extension of the process have not been evidenced, that's why the course of the disease is considered satisfactory in relation to the therapy adopted.

DISCUSSION

The cases of solitary bone plasmacytoma reported in the literature indicate a low incidence of the disease, which manifests itself mainly in men, with an age of presentation after the fifth decade of life. However, several cases have been described in women, including the reports of two patients aged 14 and 19, respectively, which coincides in terms of sex with the patient in the case presented^{4,7}.

Solitary bone plasmacytoma is located mainly in the spine (50% of cases) or in the peripheral long bones. More unusual presentations have been described, such as the hip joint, the thoracic wall, the maxillary sinus, or the skull 8,9,10,11. In 40 % of patients, the initial manifestation is paraparesis, localized pain is also frequent¹², which has been observed in this case.

The diagnosis of plasmacytoma is established by the finding of a monoclonal plasma cell histopathology, demonstrated by immunohistochemistry. This disease has the same histological characteristics as multiple myeloma, so this is the main differential diagnosis to rule out ¹³.

Some authors agree that solitary bone plasmacytoma is an early stage in the evolution of MM, which can remain latent for years. They also suggest that 40-75 % of cases with this disease subsequently developed multiple myeloma ^{5,6,14}.

As stated by Caers et al¹⁵, solitary bone plasmacytoma has four possibilities in its evolution: progression to MM (65 %), local recurrence (12 %), a new solitary lesion at distance (15 %), and complete healing.

The average time of evolution of solitary bone plasmacytoma is 2 to 3 years, with a mean survival

of 10 years. In contrast, in the absence of treatment, the survival prognosis for MM is less than two years¹⁵.

In the case presented, there is no evidence so far of torpid evolution or progression to MM. In patients with this type of diagnosis, monitoring is very important since it allows the health professional to act early in the face of manifestations of disease progression or recurrence.

Radiation therapy, with or without surgical excision of the tumor, is the treatment of choice for solitary plasmacytoma because it is a highly radiosensitive neoplasm ^{5,12,15}.

Remission occurs in 90 % of cases, only with local control of the lesion. The possibility of local relapse after radiotherapy is fewer than 5 % and 30 % for distant relapse, the latter tend to occur within 2 or 3 years after the initial diagnosis. The recurrence rate is higher in the elderly and those with axial skeletal involvement. About 60 % of patients survive at least 10 years or more^{16,17.}

There is controversy about the efficacy of adjuvant chemotherapy as a preventive therapeutic measure in the progression to MM. Predictive factors of progression to MM are tumor size, the presence of osteopenia, and the non-reduction of the monoclonal peak after treatment¹⁷.

It is understandable that avoiding the evolution to MM, through the timely diagnosis and management

of solitary bone plasmacytoma, constitutes a tool in favor of greater survival and better quality of life for the patient.

CONCLUSIONS

Solitary bone plasmacytoma is a rare monoclonal gammopathy that should be suspected in the presence of axially located bone pain without a history of trauma associated with paraparesis. Its main differential diagnosis is multiple myeloma, which is sometimes the final result of its evolution. The timely treatment of this entity prevents its evolution and improves the patient's prognosis.

AUTHORSHIP

LLP: conceptualization, research, methodology, draftoriginal writing, review.

AAL: conceptualization, data curation, resources, draft-original writing, review, validation and editing. AGM: data curation, resources, monitoring, validation and editing.

FINANCING

The authors did not receive funding for this article.

BIBLIOGRAPHIC REFERENCES

- 1. Perdomo Delgado J, Espinosa Martín L, del Valle Díaz S. Discrasia de células plasmáticas (gammapatías monoclonales). En: Noya Chaveco ME, Moya González NL. Roca Goderich Temas de medicina interna. Vol 3. 5ª ed. La Habana: Editorial de Ciencias Médicas; 2017. p. 435-444.
- 2. Bladé Creixentí J, San Miguel JF. Gammapatías monoclonales. En: Rozman C, Cardellach López F. Farreras Rozman Medicina interna. Vol 2. 19ª ed. Barcelona: Elsevier; 2020. p. 1619-1628.
- 3. Fernández A, Sancho M, Gras J, Talavera J. Plasmocitoma solitario de cabeza y cuello. Presentación de tres casos y revisión de la literatura. Act Otorrinolaringol Esp [Internet]. 2001 [cited 19/07/2021]; 52(8):715-720. Available from: https://doi.org/10.1016/S0001-6519(01)78271-4
- 4. Chacha N, Vera J, Ramírez F, Chisesi T, Posligua K. Plasmocitoma óseo solitario: caso clí-

- nico. Rev Oncol Ecu [Internet]. 2020 [cited 20/07/2021]; 30(1): 24–30. Available from: https://doi.org/10.33821/468
- 5. Xie L, Wang H, Jiang J. Does radiotherapy with surgery improve survival and decrease progression to multiple myeloma in patients with solitary plasmacytoma of bone of the spine? World Neurosurg [Internet]. 2020 [cited 20/07/2021]; 134(1):e790-e798. Available from: https://doi.org/10.1016/j.wneu.2019.11.002
- 6. Salguero AJ, Cuneo BN, Olguín HF, Osse ME, Villanueva IME. Plasmocitoma solitario de costilla como presentación inicial de mieloma múltiple: reporte de un caso. Rev Cir. [Internet]. 2019 [cited 20/07/2021]; 71(4):341-344. Available from: http://www.scielo.cl/scielo.php?script=sci_arttext&pid=s2452-45492019000400341&l-ng=es
- 7. Gil Agramonte M, Serrano Mirabal J, Machín García S, Rivera

- Keelling C, Valdés Cabrera F, González Otero A et al. Plasmocitoma solitario en pediatría: una rara entidad. Rev Cubana Hematol Inmunol Hemoter [Internet]. 2017 [cited 20/07/2021]; 33(3):102-107. Available from: http://scielo.sld.cu/scielo.php?script=sci_arttext&pid=s0864-02892017000300012&lng=es
- 8. Muñoz-Medina CE, Alemán-Frías LD, Moreno-Brito RN, Fleitas-Moreno MP, Yépez-González PJ, Soto-Alfaro TV. Plasmocitoma óseo solitario coxofemoral. Presentación de un caso. RAM [Internet]. 2019 [cited 20/07/2021]; 7(2):109-112. Available from: http://revistasam.com.ar/index.php/RAM/article/view/304
- 9. Montoya-Castillo M. Plasmocitoma óseo solitario en la pared torácica: una manifestación inusual y una bomba de tiempo. Revista de Hematología [Internet]. 2020 [cited 20/07/2021]; 21(2):115-119. Available from: https://

doi.org/10.24245/rev_hematol. v21i2.3588

10. Medina Pérez VM, Vasallo Díaz AL, Peñate Tamayo FD. Plasmocitoma solitario extramedular de órbita con extensión al seno maxilar. Medicentro Electrónica [Internet]. 2017 [cited 20/07/2021]; 21(4):351-355. Available from: http://scielo.sld.cu/scielo.php?script=sci_arttext&pid=S1029-30432017000400011&l-ng=es.

11. Zumaeta J, Lazón M, Murga A. Plasmocitoma solitario intracraneal: Reporte de caso. Peru J Neurosurg [Internet]. 2021 [cited 20/07/2021]; 3(1):37-40. Available from: http://perujournalneurosurgery.org/sites/default/files/PLAS-MOCITOMA%20SOLITARIO%20IN-TRACRANEAL%20REPORTE%20%20DE%20CASO%20Spanish.pdf
12. Pham A, Mahindra A. Solitary Plasmacytoma: a review of diagnosis and management. Curr Hema-

tol Malig Rep [Internet]. 2019 [cited 20/07/2021]; 14(2):63-69. Available from: https://doi.org/10.1007/s11899-019-00499-8

13. Gálvez-Prieto F, Lorenzo-Franco F, Luaces-Rey R, López-Cedrún Cembranos JL. ¿Cuál es su diagnóstico? Rev Esp Cirug Oral y Maxilofac [Internet]. 2017 [cited 20/07/2021]; 41(4):202-205. Available from: http://scielo.isciii.es/scielo.php?script=sci_arttext&pid=S1130-05582017000100050&l-ng=es

14. Grammatico S, Scalzulli E, Petrucci MT. Solitary Plasmacytoma. Mediterr J Hematol Infect Dis [Internet]. 2017 [cited 20/07/2021]; 9(1):e2017052. Available from: https://doi.org/10.4084/MJHID.2017.052

15. Caers J, Paiva B, Zamagni E, Leleu X, Bladé J, Kristinsson SY, et al. Diagnosis, treatment, and response assessment in solitary plasmacytoma: updated recommendations

from a European Expert Panel. J Hematol Oncol [Internet]. 2018 [cited 20/07/2021]; 11(1):10-16. Available from: https://doi.org/10.1186/s13045-017-0549-1

16. Tsang RW, Campbell BA, Goda JS, Kelsey CR, Kirova YM, Parikh RR, et al. Radiation Therapy for Solitary Plasmacytoma and Multiple Myeloma: Guidelines From the International Lymphoma Radiation Oncology Group. Int J Radiat Oncol Biol Phys [Internet]. 2018 [cited 20/07/2021]; 101(4):794-808. Available from: https://doi.or- g/10.1016/j.ijrobp.2018.05.009 17. Mheidly K, Lamy De La Chapelle T, Hunault M, Benboubker L, Benchalal M, Moreau P, et al. New insights in the treatment of patients with solitary bone plasmacytoma. Leuk Lymphoma [Internet]. 2019 [cited 20/07/2021]; 60(11):2810-2813. Available from: https://doi.org /10.1080/10428194.2019.1605067

Plasmocitoma óseo solitario, una gammapatía monoclonal infrecuente. Informe de un caso

RESUMEN

El plasmocitoma óseo solitario es un tumor de células plasmáticas de presentación poco habitual que se ha observado como estado precursor del mieloma múltiple; se localiza principalmente en la columna vertebral, provocando dolor a nivel torácico y/o lumbar. Se informó el caso de una paciente femenina de 55 años que presentó dolor lumbar de larga evolución sin traumatismo asociado, acompañado de paraparesia; le fue indicada una radiografía torácica que reveló una imagen osteolítica, correspondiente a un plasmocitoma óseo solitario que se confirmó mediante exámenes histológicos. Luego de tratamiento farmacológico y radioterapia la evolución de la paciente fue satisfactoria. El plasmocitoma óseo solitario es una gammapatía monoclonal infrecuente que debe sospecharse ante la presencia de un dolor óseo de localización axial sin antecedentes de trauma, asociado a paraparesia. Su principal diagnóstico diferencial es el mieloma múltiple, el cual en ocasiones constituye el resultado final de su evolución.

Palabras clave: Mieloma Múltiple; Plasmacitoma; Paraparesia; Paraproteinemias.



Este artículo de Revista 16 de Abril está bajo una licencia Creative Commons Atribución-No Comercial 4.0. Esta licencia permite el uso, distribución y reproducción del artículo en cualquier medio, siempre y cuando se otorgue el crédito correspondiente al autor del artículo y al medio en que se publica, en este caso, Revista 16 de Abril.