

## LETTER TO THE EDITOR

## INTRA-ORAL SPITZ NAEVUS: A CASE REPORT

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## INTRODUCTION

The Spitz nevus, also known as benign juvenile melanoma or a nevus of epithelioid and spindle cells<sup>2</sup>, was first described by Sophie Spitz in 1948<sup>1</sup> and is most common in children and adolescents.<sup>3</sup> They can present in three different ways: solitary nodular, multiple grouped, and multiple disseminated.<sup>3</sup> The great majority of lesions are solitary, nodular, and are present in children and adolescents (57%-70%), with a slight preference for the female gender. The regions where they are most frequently found are the lower extremities, the head and neck, and the trunk.<sup>4</sup> A Spitz nevus is usually pink, brown, or black in color. After an initial period of growth, the majority of Spitz nevi tend to stabilize their growth, reaching, in 95% of patients, a final size smaller than 6 mm.<sup>5</sup>

Histopathologically, a Spitz nevus is very similar to a melanoma due to the large size of the spindle or polygonal cells, frequently containing considerable cellular and nuclear pleomorphisms, the presence of inflammatory infiltrate, and, on the base of the lesion, a diminished cell size and spread among the collagen fibers of the conjunctive tissue. The difference between the Spitz nevus and a malignant melanoma can be hard to detect, but its structural pattern, which resembles a normal nevus, and its cytological traits, such as the large spindle and epithelioid cells, can be useful in making this distinction. The majority of Spitz Nevi are small and well circumscribed.<sup>5</sup>

Those lesions that are particularly hard to distinguish from malignant melanoma are called atypical or malignant Spitz nevi and Spitzoid cells.<sup>6,7,8</sup> Immuno-histochemical studies can help, but they are still not definitive.<sup>9,7</sup> A Spitz Nevus expresses the S100 protein, but HMB-45 tends to be distributed more within the junctional and superficial components of the skin. HMB-45 correlates with melanosome production and thus, HMB-45-positive cells

are of melanocytic origin. Therefore, HMB-45 may correlate best with factors that stimulate melanocytic proliferation and the production of melanosomes.<sup>8</sup>

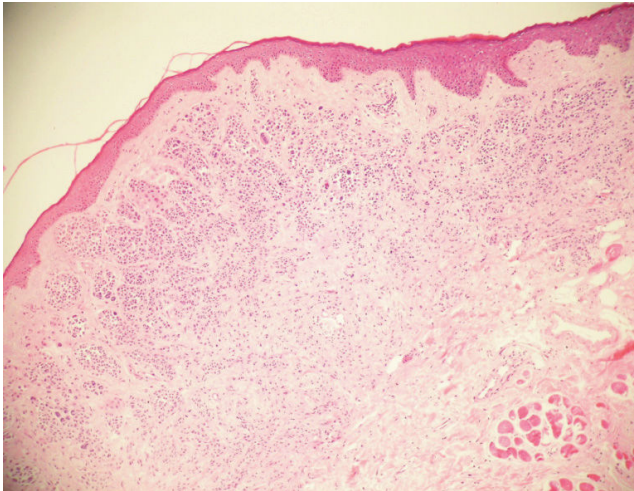
The treatment indicated is complete excision to determine the lesion's depth and extension.<sup>10</sup> Despite the fact that the Spitz nevus is a benign lesion, its transformation into a malignant lesion has been reported. Therefore, its proservation must be recommended.<sup>11</sup>

## CASE REPORT

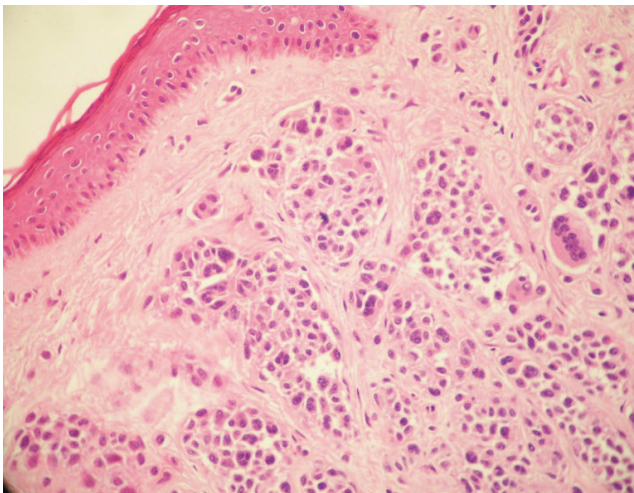
Patient D. B. S., a 32-year-old female, with pheoderma, who was a non-smoker and non-drinker, came to the Service of Oral Diagnostics at the Federal University of Rio Grande do Norte - Brazil, complaining about a lesion on the superior lip, which she had for 5 years. The intra-oral exam showed the presence of a nodule with irregular edges, a pinkish, asymptomatic, flat surface with a soft consistency, and sessile implantation. The lesion was exophytic with no bleeding, soreness or local traumatic factor, and its largest diameter was 0.5 cm. The intra-oral exam did not detect cervical lymphadenopathy. The patient said the lesion had increased its volume over a period of 5 years and had then stopped.

An excisional biopsy was performed and the fragment was removed and sent to the Service of Pathological Anatomy of the Discipline Oral Pathology, Federal University of Rio Grande do Norte - Brazil, where it was processed, stained using routine techniques, and examined. Histologically, an oral mucosa covered by a stratified orthokeratinized squamous epithelium was evident, showing atrophy and soft hyperplasia. Clear cytoplasm cells were seen all around the tissue density, suggesting nevoid cells. On the underlying conjunctive tissue, the proliferation of nevoid cells could be seen, which, on more superficial portions, organized themselves into variable-sized nests, exhibiting a raised volume, multi-nucleation and hyperchromatism due to their melanin deposition. At deeper levels, these cells lost their cohesion and acquired a more oval-shaped and fusiform aspect (figures 1 and 2). Because

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**Figure 1** - Photomicrography on H/E - 100x – where the organization of nests can be seen more superficially and, more deeply in the conjunctive tissue, the spread of nevusoid cells.



**Figure 2** - Photomicrography on H/E - 400x – showing the cellular and nuclear pleomorphism, hyperchromatism and multi nucleation.

of these findings, the histopathological diagnosis was a case of Spitz nevus.

## DISCUSSION

The occurrence of the intra-oral Spitz nevus is very rare, having been reported previously in the international literature only twice. In 1990, Nikai et al<sup>12</sup> reported a Spitz nevus case on the palate, and in 2007, Seehra et al<sup>11</sup> reported another intra-oral case on the superior lip mucosa. Our case, then, would be the third intra-oral case, and only the sec-

ond to be reported of a Spitz Nevus on the superior lip.

Herreid and Shapiro<sup>13</sup>, in comparing the distribution of the Spitz nevus and malignant melanoma by age, verified that the age range for the Spitz nevus was between 6 months and 72 years old, with an average of 21 years. The patient in the case presented here was 32 years old, which is within this age range, also corroborating the study of Cesinaro et al<sup>14</sup>, where adults represented the highest proportion of Spitz nevus patients.

Since it can be difficult to distinguish between a Spitz nevus and malignant melanoma, the complete removal of a solitary Spitz nevus is recommended. Research done by the American Academy of Dermatology found that 80% recommend removal of the lesion with 1 to 2 mm borders.<sup>15</sup> Since the case presented here is of a pigmented lesion, the surgeon decided to include a 3mm safety margin and, since the histopathological diagnosis indicated a case of Spitz nevus, the treatment was in accordance with the recommendations in the literature.

Gelbard et al.<sup>15</sup> report that 74% of the dermatologists interviewed believe that Spitz Nevus is a benign entity, however, 4% believe it is a precursor lesion to a malignant melanoma. Seven percent of general dermatologists and 4% of pediatric dermatologists have seen metastatic melanomas that originated from lesions initially histologically diagnosed as Spitz nevi, thus, the follow-up recommended by Seehra et al<sup>11</sup> must be done.

Some studies have suggested clinical and histological criteria to evaluate the malignant potential of the Spitz nevus, including diameter greater than 1 cm, subcutaneous tumor depth, the presence of soreness, and high mitotic index.<sup>16,17</sup> The lesion of the case presented here measured 0.5 cm at its largest diameter without fat involvement, ulceration, bleeding, or a high mitotic index, characterizing the lesion as low risk. However, these studies focus mainly on clinical and histological characteristics rather than the immunohistochemical patterns. Under the gradation system suggested by Spatz<sup>16</sup>, despite its applicability only to children and adolescents, the patient presented here had a low risk lesion.

Since the Spitz nevus presents a different diagnosis from that of the extremely important malignant melanoma, it is recommended that pigmented lesions be completely and safely removed with adequate patient follow up after all due procedures, since there are reports in the literature about the malignant potential of this entity.

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