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A Rare Association: Jadassohn Hamartoma, Basal Cell Carcinoma and Sebaceoma

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Abstract | **Case Report**

Jadassohn's sebaceous hamartoma or (NS), first described in 1895, is a congenital, non-melanocytic cutaneous hamartoma characterized by sebaceous gland differentiation. It typically presents as a solitary, well-defined pink or skin-colored plaque, often accompanied by alopecia. Although generally benign, NS carries a risk of malignant transformation, with basal cell carcinoma (BCC) being the most common malignancy. The association of NS with sebaceoma, a benign sebaceous gland tumor, is rare. A 49-year-old male with no significant medical history presented with a congenital erythematous lesion on the left temporal region, which had progressively enlarged and became painful over time. Clinical examination revealed a well-defined, pink, cerebriform tumor with an infiltrative base, located on an alopecic plaque. Biopsy confirmed the presence of BCC in association with sebaceoma and Jadassohn's hamartoma. Surgical excision with 5mm margins was performed under general anesthesia, followed by directed healing. Histopathological examination confirmed the diagnosis of a Jadassohn's hamartoma with BCC and sebaceoma. The patient healed without complications and was followed up regularly. NS, most commonly observed in the cephalic region, typically progresses through three clinical stages: a smooth, pink plaque in childhood, a warty and pigmented lesion in puberty, and a keratotic, firm lesion in adulthood. The risk of malignant degeneration, though rare, is a significant concern, particularly for BCC. This case highlights the rare co-occurrence of sebaceoma with NS and emphasizes the importance of early excision to prevent malignant transformation. While the risk of malignancy in NS is considered low, the psychological and cosmetic impacts of visible lesions should also be addressed. This case underscores the importance of monitoring and managing Jadassohn's sebaceous hamartoma to prevent malignant transformations such as BCC. Early surgical intervention is recommended not only for medical reasons but also to mitigate psychosocial concerns associated with visible congenital lesions. The rare association of sebaceoma with NS adds to the complexity of managing these lesions and reinforces the need for vigilant clinical surveillance.

Keywords: Benign Tumor, Sebaceous Gland Tumor, Sebaceoma, Basal Cell Carcinoma, Jadassohn's Sebaceous Hamartoma.

INTRODUCTION

Naevus sebaceus (NS), first described by Jadassohn in 1895 and thus also known as Jadassohn's NS, is a congenital non-melanocytic cutaneous hamartoma. It is characterized by sebaceous differentiation. Clinically, NS appears as a well-defined pink or skin-colored verrucous plaque accompanied by focal alopecia. It occurs in approximately 0.3% of newborns, affecting males and females equally across all races. Typically, NS presents as a solitary lesion, with a strong preference for the head and neck region in about 95% of cases [1].

Additionally, secondary neoplasms can develop in NS during adolescence and adulthood. The risk of malignant transformation reported in the literature varies between 0.8% and 22%.

The association between Jadassohn's hamartoma and basal cell carcinoma is relatively

common, whereas its association with sebaceoma is rare [2].

This article aims to report the clinical case of a patient with Jadassohn's NS associated with sebaceoma and a basal cell carcinoma who was treated surgically with excision and directed healing.

CASE PRESENTATION

Patient Presentation:

- A 49-year-old man, with no notable medical history, presented to the dermatology department for management of a congenital erythematous lesion on the left temporal region. The lesion, which has been painful and progressively enlarging since birth, prompted his consultation.

Clinical Findings:

- On clinical examination, the lesion appeared as a well-defined, oval-shaped, pink and cerebriform-like tumor measuring 4 cm (Figure *1*). It featured an infiltrated base that bled upon contact and was situated on an alopecic plaque.
- Furthermore, beneath the primary tumor, a plaque with a verrucous surface, covered by scales and small crusts, and exhibiting alopecia, was identified.

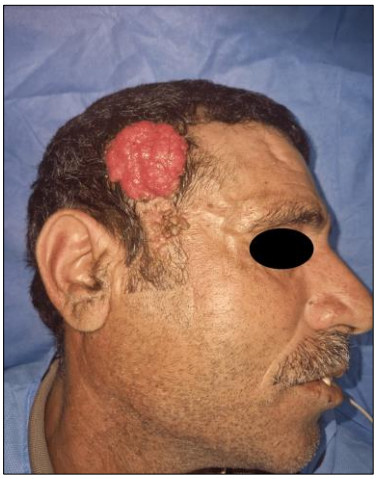

Figure 1: Preoperative image of the temporal lesion in our patient

Diagnostic Approach:

- A biopsy confirmed the presence of a basal cell carcinoma in association with a sebaceoma and a verrucous sebaceous Jadassohn's hamartoma.
- Some pre operative exams were taken then the planning of the surgery began.

Therapeutic Intervention:

- The patient underwent general anesthesia with orotracheal intubation and the temporal region was aseptically prepared using Povidone-Iodine Topical Solution (PVPI).
- Subsequently, a monobloc excision with 5mm margins was performed (Figure *2*), and it was decided to allow the surgical site to heal by directed healing.
- The specimen was labeled and sent for histological analysis.

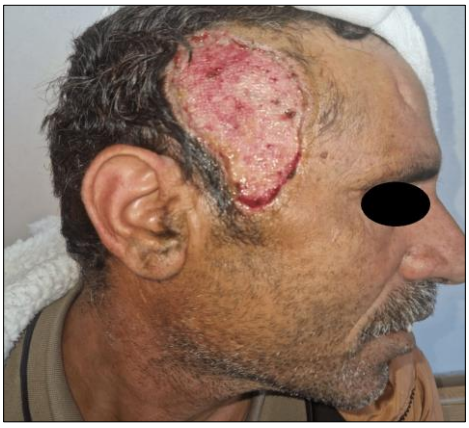

Figure 2: Image of our patient on postoperative day 1 (J1)

Histological Analysis:

Was conducted on a surgical specimen measuring 8 cm x 5 cm, containing a proliferative lesion. The excision margins were clear, and the report confirmed a diagnosis of a Jadassohn's hamartoma associated with a basal cell carcinoma (BCC) and a Sebaceoma.

Follow Up and Outcomes:

The patient was followed up weekly in the maxillofacial surgery outpatient department during the first 4 weeks (Figure *3*) and achieved complete healing without complications. He was then scheduled for follow-ups every 6 months (Figure *4*) and subsequently annually.

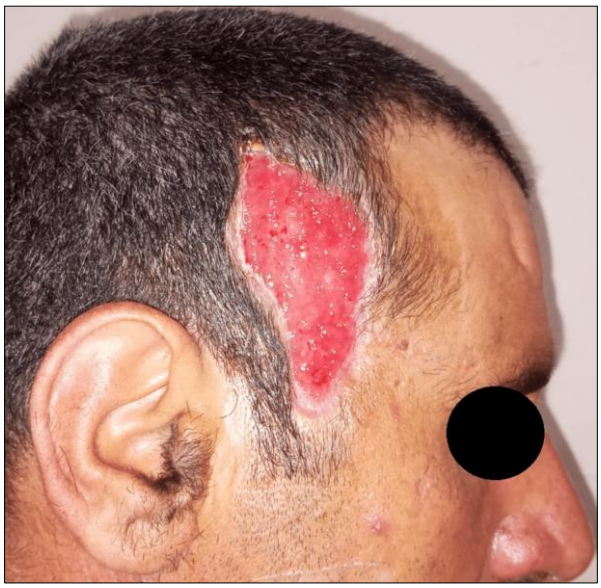

Figure 3: Image of our patient on one month post operative

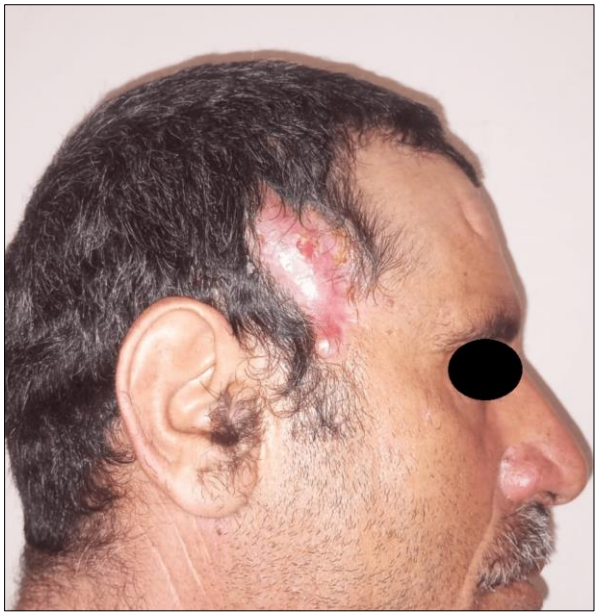

Figure 4: Image of our patient on three months post operative

DISCUSSION

Sebaceous hamartoma is a common congenital tumor, occurring in approximately 0.3% of newborns, and is most frequently observed on the cephalic region [3].

It most commonly affects the scalp, forming an alopecic plaque. This is due to the high concentration of sebaceous glands in the facial, preauricular, and temporal regions, with other locations being much less common, including an extremely rare intraoral form. It is usually a solitary lesion, generally oval in shape.

Our study aligns with literature, as in the presented case, the patient consulted for a unique lesion on the temporal region. HSJ progresses through three clinical and histological stages [4], depending on the patient's age. It first appears as a slightly raised, pinkish,

alopecic oval plaque in childhood, and later develops a mamillated and warty appearance during puberty.

The natural clinical progression is stereotyped, at birth: The child presents with a small, pink plaque with a finely granular texture, most commonly located in the cervicofacial region. At puberty: The surface of the lesion becomes mamillated and tends to become pigmented. In adulthood: The lesion becomes keratotic, firm, and elevated, and in rare cases, as seen in our observation, may take on a hypertrophic, multilobular, and extensive appearance.

The risk of degeneration is the main reason for early excision, with basal cell carcinoma being the primary concern despite its relatively low aggressiveness. Traditionally, NSJ is linked to the development of malignancies. A study of 155 cases found that the most common lesions arising from an NSJ are trichoblastomas. Of the 33 neoplasms observed in these cases, all were benign. Approximately one third were trichoblastomas and syringocystadenoma papilliferum, while the remainder included sebomatricomas, apocrine cysts, and apocrine poromas. Malignant tumors arising in NSJ are rare, with only 4 well documented cases of squamous cell carcinomas (SCCs) reported in the literature. Other malignancies noted include adenocarcinomas and sebaceous carcinomas [5].

In our patient, the Jadassohn's hamartoma was associated with a malignant tumor, specifically basal cell carcinoma (BCC), and a benign tumor, which was a sebaceoma.

A clinical audit published in 2007 concluded that prophylactic excision of NS is not necessary, especially in young children. Excision is only recommended when there is clinical suspicion of benign or malignant neoplasms, or for cosmetic reasons. While more recent studies suggest that the risk of malignant transformation [6] may be lower than previously thought, full excision was performed in this patient. In addition to the potential for secondary tumor development, psychosocial factors such as high social anxiety, social avoidance, discrimination, impaired social interactions, and disruption of personal life must also be considered. Dissatisfaction with facial appearance may also lead to behavioral challenges [7].

In conclusion, we suggest that NS should be excised at the earliest opportunity to prevent the onset of secondary neoplasms and to mitigate potential psychosocial concerns.

CONCLUSION

Jadassohn's sebaceous hamartoma is a benign tumor that typically regresses over time since childhood. However, the risk of complications from both benign and malignant tumors, particularly basal cell carcinoma (BCC), has been documented, highlighting the importance of surveillance.

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