ABSTRACT

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Predictors of mortality and tumor recurrence in desmoplastic infantile ganglioglioma and astrocytoma-and individual participant data meta-analysis (IPDMA).

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PURPOSE: Desmoplastic infantile astrocytoma (DIA) and desmoplastic infantile ganglioglioma (DIG) are classified together as grade I neuronal and mixed neuronal-glial tumor of the central nervous system by the World Health Organization (WHO). These tumors are rare and have not been well characterized in terms of clinical outcomes. We aimed to identify clinical predictors of mortality and tumor recurrence/progression by performing an individual patient data meta-analysis (IPDMA) of the literature.

METHODS: A systematic literature review from 1970 to 2020 was performed, and individualized clinical data for patients diagnosed with DIA/DIG were extracted. Aggregated data were excluded from collection. Outcome measures of interest were mortality and tumor recurrence/progression, as well as time-to-event (TTE) for each of these. Participants without information on these outcome measures were excluded. Cox regression survival analyses were performed to determine predictors of mortality and tumor recurrence / progression.

RESULTS: We identified 98 articles and extracted individual patient data from 188 patients. The cohort consisted of 58.9% males with a median age of 7 months. The majority (68.1%) were DIGs, while 24.5% were DIAs and 7.5% were non-specific desmoplastic infantile tumors; DIAs presented more commonly in deep locations ($p = 0.001$), with leptomeningeal metastasis ($p = 0.001$), and was associated with decreased probability of gross total resection (GTR; $p = 0.001$). Gender, age, and tumor pathology were not statistically significant predictors of either mortality or tumor recurrence/progression. On multivariate survival analysis, GTR was a predictor of survival (HR = 0.058; $p = 0.007$) while leptomeningeal metastasis at presentation was a predictor of mortality (HR = 3.27; $p = 0.025$). Deep tumor location (HR = 2.93; $p = 0.001$) and chemotherapy administration (HR = 2.02; $p = 0.017$) were associated with tumor recurrence/progression.

CONCLUSION: Our IPDMA of DIA/DIG cases reported in the literature revealed that GTR was a predictor of survival while leptomeningeal metastasis at presentation was associated with mortality. Deep tumor location and chemotherapy were associated with tumor recurrence / progression.

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